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Linguistic and psychological variables in children with autism spectrum disorder and their relatives: Implications on family quality of life

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A mi tesoro más preciado,

mi familia

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SUMMARY

VARIABLES LINGÜÍSTICAS Y PSICOLÓGICAS EN LOS NIÑOS CON TRASTORNO DEL ESPECTRO AUTISTA Y SUS FAMILIARES: IMPLICACIONES EN LA CALIDAD DE VIDA FAMILIAR

El trastorno del espectro autista (TEA) es un trastorno del neurodesarrollo caracterizado por presentar déficits en la interacción social y en la comunicación y patrones de conducta restringidos y repetitivos. Se estima que afecta a 1 de cada 59 niños, y su irrupción dentro del seno familiar supone un gran impacto en el bienestar y la calidad de vida familiar (CdVF). Sin embargo, aunque se trata de un trastorno con una alta incidencia y con características incapacitantes tanto para las personas con TEA como para sus familiares, aún no se han estudiado en profundidad y de forma integrada tres aspectos de gran importancia que potencialmente podrían tener impacto sobre la CdVF en familias de niños con TEA desde su nacimiento hasta el final de la edad escolar: (1) la detección de señales tempranas en personas con TEA, (2) la identificación de factores lingüísticos y psicológicos que afectan a la CdVF en personas con TEA, y (3) la caracterización de factores familiares (relacionados con los padres y los hermanos de niños con TEA) que afectan directamente a la CdVF.

Por ello, en la presente tesis doctoral se propone un modelo integrado general sobre cómo diferentes factores psicológicos y lingüísticos influyen en la CdVF en TEA. Entre los factores que investigamos se incluyen: las habilidades lingüísticas y socio–comunicativas, las habilidades motrices, las habilidades numéricas y otros factores psicológicos como el apoyo social y la CdVF. En este trabajo utilizamos un enfoque interdisciplinar. En concreto, empleamos teorías y métodos propios de diversas áreas, entre los que se incluye la psicología del desarrollo, la psicología cognitivo–conductual y la logopedia. Asimismo, empleamos dos metodologías científicas principalmente: la investigación de campo y el estudio experimental. En particular, llevamos a cabo y analizamos siete estudios de caso–control en niños en riesgo o con un diagnóstico de TEA, sus padres y/o hermanos, una intervención con padres de niños con TEA y un meta–análisis de la literatura. Las muestras de participantes incluyen tanto niños en riesgo o con TEA, como padres y hermanos de niños con TEA procedentes de Estados Unidos y España. Además, registramos un amplio abanico de variables independientes y moderadoras/mediadoras en nuestros modelos (p.

ej., severidad del TEA, lenguaje receptivo, lenguaje expresivo, motricidad fina, motricidad gruesa, apoyo social, problemas emocionales y conductuales, intenciones conductuales, conducta, y habilidades numéricas).

Los resultados de esta tesis tienen implicaciones tanto teóricas como aplicadas para la detección temprana del TEA, la promoción de la CdVF y su relación con características de los padres y los hermanos de niños con TEA. Entre ellas podemos destacar a nivel teórico: (1) la caracterización de algunos de los aspectos relacionados con una detección temprana del TEA, (2) la comprensión de las variables que influyen sobre la CdVF en TEA, y (3) la definición y mejora de algunas características presentes en padres y hermanos que median o explican en parte la CdVF. Para poder aplicar las intervenciones en niños con TEA, donde se pretende mejorar la CdVF, estas intervenciones deberían incorporar objetivos adicionales relacionados con estos niños (como el incremento en la comprensión lingüística y la conducta adaptativa), con los padres de niños con TEA (como la incorporación de estrategias de aprendizaje para incrementar las habilidades de interacción y comunicación con sus hijos y la mejora en las habilidades numéricas) y con los hermanos de niños con TEA (como el trabajo en habilidades relacionadas con la comunicación y el apoyo social). A nivel aplicado, los resultados de esta tesis plantean sugerencias que pueden mejorar la detección y el diagnóstico del TEA y potenciar el diseño de los objetivos de las intervenciones para mejorar la CdVF, especialmente aquellas destinadas a las intervenciones con niños con TEA, padres y hermanos.

En conjunto, los resultados de esta tesis doctoral indican que las habilidades lingüísticas además de influir sobre la detección y el diagnóstico temprano en TEA, también ejercen un impacto sobre la CdVF una vez recibido el diagnóstico junto con otras variables de índole psicológica. En concreto, una mejor comprensión lingüística y una mayor conducta prosocial en los niños con TEA, unas mejores habilidades numéricas en los padres y un mejor apoyo social en los hermanos de niños con TEA ejercen un impacto positivo sobre la CdVF. Adicionalmente, los resultados de la intervención mediada por padres muestran que tras la intervención mejoran las estrategias de comunicación, la interacción de los padres con sus hijos con TEA y el manejo de la conducta de los padres, y que esta mejoría repercute positivamente en la satisfacción de la CdVF (papel de padres, bienestar emocional e interacción familiar). Por tanto, la comprensión paterna de las

características de la comunicación de las personas con TEA, los principios de refuerzo en las aproximaciones de lenguaje oral y la sensibilidad y el interés hacia los intentos de comunicación intencional de los niños con TEA, a su vez, no sólo mejoran la CdVF, sino que también ayudan a los padres a entender su potencial papel dentro del desarrollo de sus hijos.

LINGUISTIC AND PSYCHOLOGICAL VARIABLES IN CHILDREN WITH AUTISM SPECTRUM DISORDER AND THEIR RELATIVES: IMPLICATIONS ON FAMILY QUALITY OF LIFE

Autism spectrum disorder (ASD) is a neurodevelopmental disorder characterized by symptoms in two broad domains, i.e., remarkable deficits in communication and social interaction, and the presence of restricted and/or repetitive interests and behaviors. ASD is estimated to affect 1 out of 59 children, and its outbreak within the family has a huge impact on well-being and family quality of life (FQoL). Indeed, ASD shows a high incidence and disabling features for both people with ASD and their relatives. However, there is a dearth of published research on three important topics that could have a potential impact on FQoL in families with a child with ASD. These topics are (1) detection of early signs of ASD, (2) identification of linguistic and psychological variables that affect the FQoL in people with ASD, and (3) characterization of family factors (related to parents and siblings of children with ASD) that directly affect FQoL.

The goal of this thesis is to conduct research on these topics and to propose a general integrative model on the impact of different psychological and linguistic factors on FQoL in ASD. In particular, we investigate the influence of linguistic and socio–communicative skills, motor skills, numeracy skills, and other psychological factors such as social support and FQoL. We use an interdisciplinary approach. Specifically, we use theories and methods from several areas, including developmental psychology, cognitive behavioral psychology, and speech science, and two main scientific methodologies (i.e., field research and experimental studies). We conduct seven case–control studies with children at risk for or with a diagnosis of ASD, and their parents and/or siblings. We also conduct an intervention mediated by parents of children with ASD, and a meta-analytic review. Our participants' samples include both children at risk for or with ASD and parents, as well as siblings of children with ASD from several cultures, including USA and Spain. Moreover, we record a wide range of independent variables and moderators and/or mediators in our models, including severity of ASD, receptive language, expressive language, fine motor skills, gross motor skills, social support, emotional and behavioral problems, behavioral intentions, and numeracy skills.

Results in this research can have important theoretical and applied implications for the detection of ASD, promotion of the FQoL, and its relation with characteristics of parents and siblings of children with ASD. Among these theoretical implications, we would include: (1) the characterization of several linguistic factors related to an early detection of ASD, (2) the understanding of those variables influencing the FQoL in ASD, and (3) the definition and improvement of some aspects about parents and siblings that could mediate their FQoL. To implement interventions in children with ASD to improve their FQoL, these interventions can aim to improve additional factors related (1) to these children (such as increasing linguistic comprehension and the adaptive behavior), (2) to their parents (such as teaching strategies to improve language and communication together with numeracy skills), and (3) to their siblings (such as improving those skills related to communications and social support). At an applied level, the results of this thesis provide suggestions that could improve the detection and the diagnosis of ASD, and it would help design interventions which improve the FQoL in children with ASD, parents, and siblings.

On balance, the results of this thesis suggest that besides the influence on early detection and diagnosis of ASD by linguistic skills, these abilities and additional psychological variables have a significant influence on FQoL once ASD has been diagnosed. Overall, a better linguistic comprehension and prosocial behavior in children with ASD, more adequate numerical skills in parents, and a better social support in siblings of children with ASD make a positive impact on FQoL. In addition, results in our intervention show that parents can improve their communication and their interactions with their children with ASD. Moreover, this improvement is positively correlated with satisfaction on FQoL (i.e., parental role, emotional well-being, and family interaction). Hence, parental comprehension of communication characteristics of children with ASD, reinforcement of language's approximations, parental responsiveness, behavioral management, and their interest in the intentional communication attempts of their children with ASD, in turn, not only improve their FQoL, but help parents to understand their potential role in their children's development.

THESIS PUBLICATIONS

Publications included in the thesis

Garrido, D., Carballo, G., Artis, J., & Garcia-Retamero, R. (2018). Timing of parents' concerns related to Autism Spectrum Disorder and its diagnosis: A mediation analysis. *The Spanish Journal of Psychology*, 21, e59, doi:10.1017/sjp.2018.64

Garrido, D., Watson, L. R., Carballo, G., Garcia-Retamero, R., & Crais, E. R. (2017). Infants at–risk for autism spectrum disorder: Patterns of vocalizations at 14 months. *Autism Research*, *10*(8), 1372–1383, doi:10.1002/aur.1788

Garrido, D., García–Fernández, M., Garcia-Retamero, R., & Carballo, G. (2017). Perfil comunicativo y de adaptación social en población infantil con trastorno del espectro autista: Nuevo enfoque a partir de los criterios del DSM–5. *Revista de Neurología*, 65, 49–56, doi: 10.33588/rn.6502.2017019

Garrido, D., Carballo, G., Ortega, E., & Garcia-Retamero, R. (under review). Conducta adaptativa en niños con autismo y su efecto sobre la calidad de vida familiar.

Garrido, D., Carballo, G., Franco, V., & Garcia-Retamero, R. (2015). Dificultades de comprensión del lenguaje en niños no verbales con trastorno del espectro autista y sus implicaciones en la calidad de vida familiar. *Revista de Neurología, 60,* 207–214, doi: 10.33588/rn.6005.2014226

Garrido, D., Garcia-Retamero, R., & Carballo, G. (under review). Improving social–communication management and family quality of life through parent-mediated intervention in autism spectrum disorder.

Garrido, D., Petrova, D., Cokely, E., Carballo, G., & Garcia-Retamero, R. (under review). Parental numeracy may be associated with higher quality of life in families with a child with autism spectrum disorder.

Garrido, D., Petrova, D., Watson, L. R., Garcia-Retamero, R., & Carballo, G. (2017). Language and motor skills in siblings of children with autism spectrum disorder: A meta–analytic review. *Autism Research*, *10*(11), 1737–1750, doi:10.1002/aur.1829

Garrido, D., Carballo, G., & Garcia-Retamero, R. (under review). Siblings of children with autism spectrum disorders: Social support and quality of life.

Related publications

Garrido, D., Carballo, G., y Garcia-Retamero, R. (under review). Detección de las dificultades lingüísticas expresivas en la edad escolar

Garrido, D., Carballo, G., y Garcia-Retamero, R. (under review). Análisis de la comprensión gramatical en el trastorno del espectro autista y el retraso del lenguaje

Garrido, D., Garcia-Retamero, R., and Carballo, G., (in prep). Developmental profiles in siblings of children with autism spectrum disorder: A cross sectional developmental trajectories comparison

Garrido, D., Carballo, G., y Garcia-Retamero, R. (in prep). Perfil de comprensión gramatical en los trastornos del Lenguaje y la Comunicación

Garrido, D., Garcia-Retamero, R., and Carballo, G., (in prep). Reading abilities and language skills in school-age children with autism spectrum disorder

Garrido, D., Carballo, G., and Garcia-Retamero, R. (in prep). Identifying language impairments in school-age children with autism spectrum disorder using language sample analysis

Note: Each chapter of the thesis consists of one article that either has been published or is in the process of being published in a scientific journal. The content presented here may not fully coincide with the final published articles.

PART I

INTRODUCTION

CHAPTER 1

Chapter 1

Autism spectrum disorder, language, and family quality of life

"Qué bonito, pero a la vez qué duro es ser madre … No saber dónde le duele, no escuchar un "mamá" o un "te quiero" de sus labios… No cambiaría a mi hijo por nada del mundo, pero sí cambiaría el mundo por mi hijo…"

María, madre de un niño con TEA

El trastorno del espectro autista (TEA) se caracteriza por desafíos en la comunicación social y por presentar conductas estereotipadas, repetitivas y restringidas (APA, 2013). Este trastorno presenta una alta prevalencia (afecta a 1 de cada 59 niños) y afecta más a niños que a niñas, ya que aparece en 4 niños por cada niña (Baio et al., 2018). Debido a las características que presenta el TEA, el bienestar y la calidad de vida familiar (CdVF) se ven afectados. Por ello, esta tesis tiene como principal objetivo analizar el impacto que diversas variables lingüísticas y psicológicas pueden ejercer sobre el diagnóstico y el bienestar en niños con TEA y en su entorno familiar. Además, se propone como objetivo secundario el realizar propuestas para mejorar la CdVF en familias de niños con este trastorno.

Para completar estos objetivos hemos analizado diversas variables y hemos considerado tres poblaciones diferentes: niños con diversos niveles de severidad o apoyo, sus padres y hermanos. Para ello, hemos explorado tres enfoques (como se indican en cada una de las partes que componen esta tesis) que afectan directamente a la CdVF y que hemos estudiado en profundidad y de forma integrada: (1) aspectos previos al diagnóstico, es decir, la detección de señales tempranas en TEA, (2) aspectos tras el diagnóstico como la identificación de factores lingüísticos y psicológicos en niños con TEA que afectan a la CdVF, y (3) la caracterización de factores familiares (relacionados con los padres y los hermanos de niños con TEA).

A lo largo de estas partes hemos tenido en cuenta a los diferentes miembros que conforman la familia más cercana (es decir, niños en riesgo de tener TEA, niños con TEA con diferentes niveles de apoyo, padres y hermanos) que pueden tener un impacto tanto sobre el diagnóstico como en la CdVF desde el nacimiento hasta el final de la edad escolar

de los niños con TEA (ver Figura 1.1 para una visión general de las poblaciones objetivo en cada una de las partes de este trabajo).

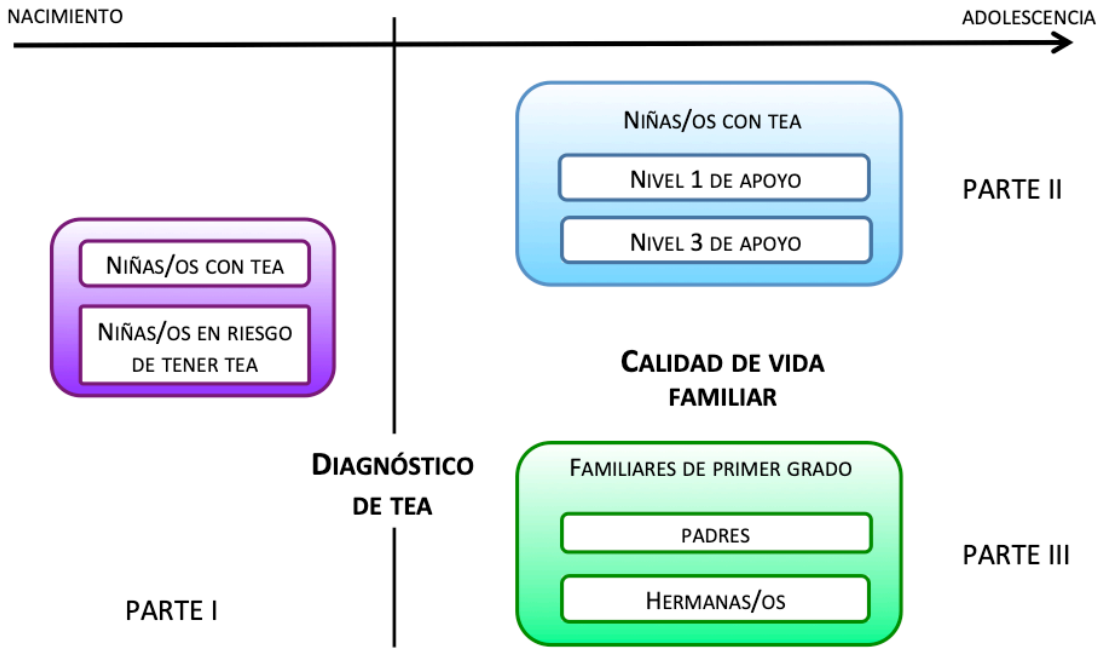

Figura 1.1. Visión general de las poblaciones objetivo en las partes de la tesis

Además, hemos evaluado en cada una de las partes que componen esta tesis tanto variables lingüísticas como psicológicas que potencialmente podrían tener un impacto en el diagnóstico en TEA y en la CdVF (ver Figura 1.2 para una visión general de las principales variables analizadas en función de cada parte). Además, a lo largo de esta tesis hemos utilizado el conocimiento y la teoría de diversas áreas científicas como la Psicología del Desarrollo, la Ciencia Cognitiva, la Psicología de la Salud, la Logopedia y los Juicios y la Toma de Decisiones.

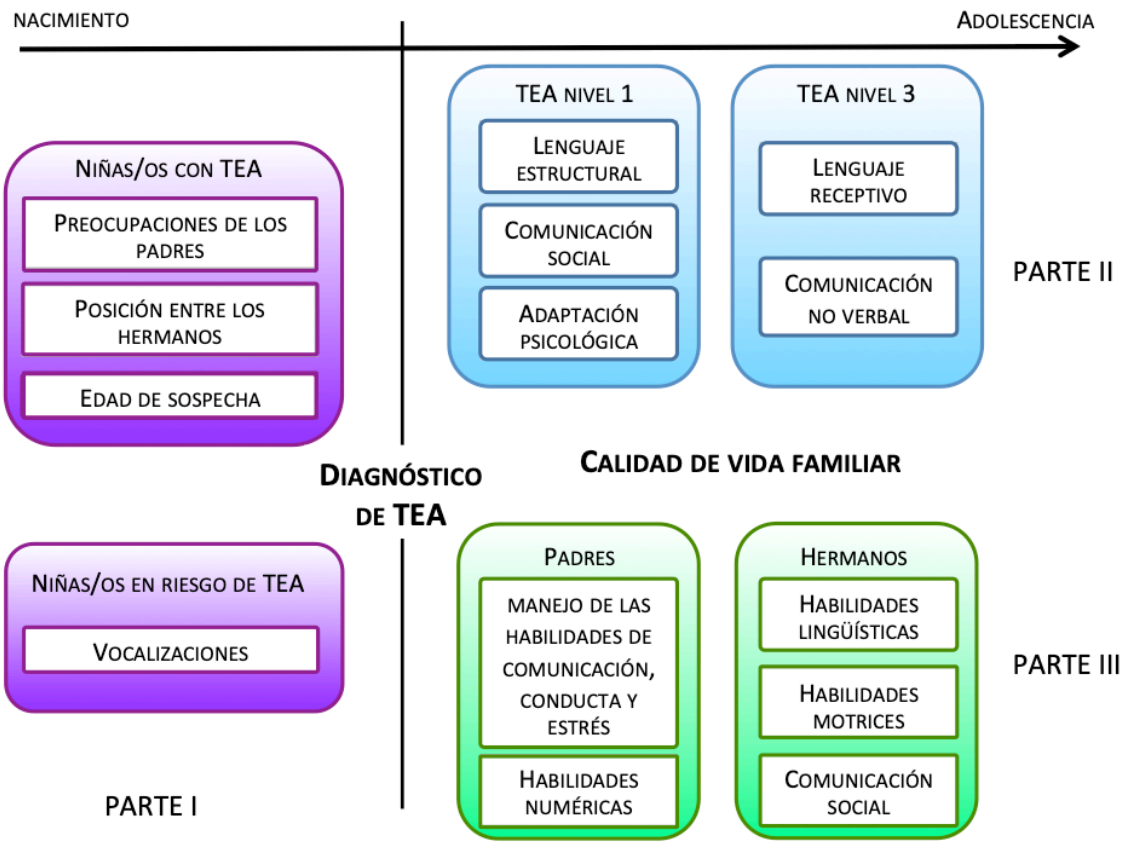

Figura 1.2. Principales variables analizadas en las partes de la tesis

Parte II: El desarrollo de los niños con TEA antes de recibir un diagnóstico

En esta segunda parte analizamos uno de los momentos más críticos en las familias de niños con TEA y una de las primeras etapas en sus vidas como familia de un niño con un trastorno del neurodesarrollo: la sospecha, la detección, y el diagnóstico.

En el TEA, recibir un diagnóstico temprano es fundamental para acceder a la intervención temprana. Sin embargo, debido a la ausencia de claros marcadores biológicos, el diagnóstico de este trastorno está relacionado con el reconocimiento de ciertos indicadores tempranos en el desarrollo socio–comunicativo y conductual del niño. Sorprendentemente, aunque la incidencia del TEA se ha incrementado significativamente en los últimos años, la edad de diagnóstico del TEA en España y Europa sigue produciéndose de media a los 3–3.5 años de edad (Díaz-Atienza, García-Pablos & Martín-Romera, 2004; Salomone, Charman, McConachie, & Warren, 2016). Por todo ello, tanto la

detección de señales de riesgo como el diagnóstico temprano es un claro desafío y una necesidad esencial en Psicología.

En España, se estima que en alrededor del 80% de los casos de niños con TEA, son los padres los primeros en detectar aquellas señales en el desarrollo o síntomas tempranos relacionados con el trastorno (Fortea–Sevilla, Escandell–Bermúdez, & Castro–Sánchez, 2013). Se ha establecido que entre el 30–50% de las señales tempranas se expresan en torno al primer año de vida, y entre el 80–90% de estas señales se pueden detectar a los 2 años (Volkmar, Chawarska, & Klin, 2008). Las dificultades relacionadas con el retraso en el lenguaje y la comunicación son dos de las primeras preocupaciones que los padres suelen informar (Meek, Robinson, & Jahromi, 2012). Algunas de estas señales están relacionadas con retrasos en la adquisición de hitos evolutivos (p. ej., retraso en el habla), pérdida de habilidades adquiridas previamente (regresión antes de los 36 meses), o la falta de progresión (estancamiento) de ciertos pre–requisitos como una menor orientación social, la ausencia de la sonrisa social y el pobre contacto visual (Kishore & Basu, 2011; Maestro et al., 2005; Mandell, Novak, & Zubritsky, 2005; Mishaal, Ben–Itzchak, & Zachor, 2014).

Además de las señales y/o dificultades que aparecen relacionadas con el desarrollo, otros factores también pueden influir a la hora de detectar el TEA. Por ejemplo, tener un hijo mayor puede propiciar una detección más temprana debido a que los padres pueden comparar el desarrollo del hijo que presenta ciertos rasgos o características inusuales con el de su hijo con desarrollo típico (DT) (Herlihy, Knoch, Vibert, & Fein, 2015). Otra variable que también puede influir es el nivel educativo de los padres o el género del propio niño. Por ejemplo, un nivel educativo bajo se ha asociado con un retraso en la detección de señales relacionadas con TEA (Mishaal et al., 2014). Además, el género del niño podría influir en la edad de sospecha dado que quizás los padres esperan de las niñas una mayor conducta social, lo que podría fomentar que actúen de forma más social y por lo tanto, retrasar la detección del TEA (Begeer et al., 2013). Sin embargo, no existe un consenso general sobre qué características son determinantes o ejercen un efecto sobre el momento de la detección temprana en el TEA. Por ello, el principal **objetivo** de la segunda parte de esta tesis, compuesta por dos estudios, es el de profundizar en los factores que están relacionados con la detección y el diagnóstico en TEA.

En particular, en el **segundo capítulo** investigamos de forma integrada en qué medida aquellas variables que a lo largo de la literatura se han estudiado de forma aislada (preocupaciones de los padres acerca del desarrollo de su hijo, la edad y el nivel educativo paterno, la edad y el género del niño y la existencia de un hermano/a mayor en la familia) predicen conjuntamente el tiempo de espera entre el momento de la sospecha y el diagnóstico. En este estudio presentamos los resultados de dos modelos derivados de un análisis retrospectivo en una muestra española de 48 familias de niños con TEA. Examinamos la influencia de aquellas señales tempranas más determinantes (preocupaciones relacionadas con aspectos socio–comunicativos, preocupaciones relacionadas con aspectos de conductas estereotipadas y repetitivas y otras preocupaciones del desarrollo no relacionadas directamente con los criterios diagnósticos de TEA) que pueden hacer pensar a los padres que su hijo podría tener TEA.

Ofrecer señales que puedan contribuir a detectar el trastorno lo antes posible es otro aspecto esencial en el diagnóstico temprano debido a (1) la falta de marcadores biológicos y (2) al hecho de que los padres son los primeros en detectar el TEA. Íntimamente relacionado con los criterios diagnósticos son las habilidades socio–comunicativas. La investigación más reciente muestra que no existen diferencias en el desarrollo entre niños con TEA y niños con DT hasta los 6 meses de vida (Ozonoff et al., 2010). Sin embargo, sí se han descrito diferencias a los 9–12 meses (Paul, Fuerst, Ramsay, Chawarska, & Klin, 2011) y a los 15–18 meses (Patten et al., 2014). Concretamente, los niños con TEA producen menos vocalizaciones relacionadas con el habla (p. ej., baja locuacidad), un menor balbuceo canónico (es decir, sílabas completas formadas por consonantes y vocales), un mayor número de vocalizaciones no relacionadas con el habla o vocalizaciones atípicas (gritos y chillidos) comparados con los niños con DT (Paul et al., 2011; Patten et al., 2014; Plumb & Wetherby, 2013; Schoen, Paul, & Chawarska, 2011). Otros estudios han mostrado que en algunos niños con TEA se produce una regresión en el balbuceo más complejo alrededor de los dos años de vida (Werner & Dawson, 2005).

Un aspecto relevante en las señales relacionadas con las habilidades socio–comunicativas es la evaluación de la intención comunicativa. En este sentido, se ha encontrado que en el primer año de vida los niños que reciben un diagnóstico de TEA producen menos vocalizaciones dirigidas hacia otros (Ozonoff et al., 2010; Winder,

Wozniak, Parlade, & Iverson, 2013). Sin embargo, aunque la evidencia científica disponible sugiere que existen algunos marcadores que podrían ayudar a los padres a detectar un TEA emergente, pocos estudios han evaluado estos marcadores en una muestra de alto riesgo de TEA de forma prospectiva. Por ello, en el **tercer capítulo** ampliamos los resultados presentados en el segundo capítulo y ofrecemos una perspectiva más completa de aquellos factores lingüísticos (es decir, vocalizaciones con un propósito comunicativo) que pueden mejorar la confianza de los padres y pediatras en el momento de sospechar y detectar el TEA. En este capítulo exponemos los resultados de un estudio prospectivo que examina las vocalizaciones de una muestra de 82 niños estadounidense en riesgo de tener TEA identificados a través de un instrumento de cribado a los 14 meses. El objetivo de este capítulo es el de informar en qué medida los diferentes tipos de vocalizaciones analizadas (vocalizaciones relacionadas con el habla [canónicas, no canónicas, con intención comunicativa y sin intención comunicativa], y vocalizaciones no relacionadas con el habla [atípicas, estrés, y de placer]) predicen la pertenencia al grupo con TEA, al grupo dentro del espectro o al grupo sin diagnóstico de TEA a los 22 meses.

Parte III: El trastorno del espectro autista y sus implicaciones en la calidad de vida familiar

Tras el diagnóstico, atender a las necesidades individuales de cada niño con TEA y de sus respectivas familias es fundamental para poder mejorar sus habilidades, aptitudes y el bienestar familiar. Una de las grandes dificultades a la hora de ofrecer intervenciones eficaces para los niños con TEA es la gran heterogeneidad en cuanto a severidad de síntomas y alteraciones que se presentan. La clasificación del DSM–5 (APA, 2013) sugiere que cada niño con TEA se puede localizar a lo largo de varios continuos. Desde este enfoque es indispensable conocer dónde se sitúa cada niño a lo largo del espectro para poder ofrecer una intervención personalizada e individualizada en función de sus necesidades. Dado que esta tesis está centrada en los aspectos del lenguaje y comunicación social, las muestras de los diversos trabajos presentados en la tercera parte se han definido en función del nivel de apoyo o severidad (según el DSM–5; APA, 2013) a lo largo de este continuo. Para optimizar las diferencias encontradas a lo largo de los niveles de apoyo, hemos considerado los dos extremos en el trastorno para clasificar a los

participantes en los estudios. Además, a lo largo de la literatura, la mayoría de los estudios en TEA no incluían muestras de niños no verbales (Tager-Flusberg et al., 2017) por lo que consideramos fundamental evaluar también a esa población. Para ello, hemos distinguido entre muestras de niños con TEA nivel de apoyo 1 (niños con mínimo apoyo) y niños con nivel de apoyo 3 (niños con TEA no verbales).

Siguiendo con la línea argumental de esta tesis, en la tercera parte analizamos otro factor esencial que tiene un impacto en las familias de niños con TEA: el papel del propio niño con TEA. El **objetivo** que perseguimos en esta tercera parte, formada por tres estudios, es el de ampliar la literatura previa relacionada con la CdVF en las familias de niños con TEA tras recibir el diagnóstico y hasta el final de la etapa escolar. Aunque los niños con TEA nivel 1 de apoyo se definen por no presentar problemas de lenguaje ni retraso cognitivo, hay algunos aspectos en los que sí difieren de los niños con DT (Helland, Biringer, Helland, & Heimann, 2012; Martín–Borreguero, 2005; Saalasti et al., 2008). Por ejemplo, se ha mostrado que los niños con TEA nivel 1 de apoyo tienen (entre otras) dificultades en las habilidades pragmáticas, los aspectos comunicativos no verbales, el cambio de tema en la conversación y también a la hora de hacer inferencias, utilizar y comprender conceptos abstractos, metáforas, y dobles sentidos (Bishop & McDonald, 2009; Loukusa & Moilanen, 2009).

Adicionalmente a las características lingüísticas y socio–comunicativas, aparecen otras dificultades de forma generalizada, como las dificultades conductuales, la falta de atención/hiperactividad, los problemas con los compañeros o las dificultades emocionales (Hervás & Rueda, 2018; Russell, Rodgers, & Ford, 2013). A lo largo de la literatura, se ha mostrado que algunos de estos aspectos alterados (como ciertas variables lingüísticas) se relacionan con problemas en habilidades sociales y pueden tener impacto sobre las habilidades de adaptación social y las relaciones interpersonales (Gilmour, Hill, Place, & Skuse, 2004; Meyer, Mundy, Van Hecke, & Durocher, 2006). Además, las habilidades lingüísticas y comunicativas también ejercen diversos efectos en la interacción y en el funcionamiento familiar (Gardiner & Iarocci, 2012; Lee, Harrington, Louie, & Newschaffer, 2008). Dadas las repercusiones que las alteraciones lingüísticas pueden conllevar en las futuras habilidades socio–comunicativas y el patrón conductual en los niños con TEA, se hace necesario conocer el patrón existente de habla, lenguaje, habilidades comunicativas y

adaptación social. Por ello, en el cuarto y quinto capítulos de esta tesis, investigamos tanto el perfil comunicativo y el funcionamiento emocional y conductual en niños con TEA nivel 1 de apoyo, como su relación con una medida que refleja el nivel familiar general de bienestar: la CdVF.

Concretamente, en el **cuarto capítulo** analizamos los resultados de un estudio de caso–control realizado con una muestra de 37 niños españoles con TEA con nivel de apoyo 1 o con DT. En este estudio investigamos las diferencias en niños con TEA nivel 1 de apoyo en términos de variables de lenguaje estructural (expresión y comprensión), variables de interacción social (pragmática) y factores de adaptación social (síntomas emocionales y problemas con los compañeros) que podrían establecer diferencias entre estos niños y otros que reciben el mismo diagnóstico (basado en el DSM–5; APA, 2013).

En el **quinto capítulo** ampliamos los resultados previos relacionados con los niños con TEA con nivel de apoyo 1. Analizamos la adaptación social, que aunque no se trata de un criterio diagnóstico, podría tener un efecto en la CdVF. Presentamos los resultados de un estudio de caso–control en el que participaron 49 familias españolas de niños con TEA nivel 1 de apoyo y niños con DT. En este estudio investigamos la influencia de aquellas variables relacionadas con el ajuste psicológico (como síntomas emocionales, conducta disruptiva, hiperactividad, problemas con los iguales y conducta prosocial) en la satisfacción en la CdVF.

Debido a la gran heterogeneidad que se presenta en el trastorno, hemos analizado también las características de lenguaje y comunicación de los niños con TEA que se sitúan en el otro extremo del continuo (es decir, niños con TEA nivel 3 de apoyo). De hecho, alrededor de un 25% de los niños con TEA son no verbales y muestran limitaciones comunicativas severas (Luyster, Kadlec, Carter, & Tager–Flusberg, 2008). Se considera (a nivel cualitativo) que un niño es mínimamente verbal si produce un repertorio muy limitado de palabras habladas o frases fijas que se usan con fines comunicativos (Kasari, Brady, Lord, & Tager-Flusberg, 2013). Sin embargo, aunque el término mínimamente verbal no está definido operacionalmente, a lo largo de las investigaciones se ha considerado como un criterio cuantitativo el producir un repertorio menor de 20 palabras

funcionales (Brignell et al., 2016; Kasari et al., 2013; Tager–Flusberg & Kasari, 2013; Yoder & Stone, 2006), criterio utilizado en nuestro trabajo.

Dadas las limitaciones a nivel de lenguaje y comunicación, la evaluación de estas habilidades en los niños con TEA no verbales (nivel de apoyo 3) es muy compleja porque entre otras dificultades, presentan niveles bajo de atención (Tager–Flusberg & Kasari, 2013). En este sentido, algunos estudios han mostrado que en esta población existe un retraso temprano en el lenguaje, ausencia de habilidades verbales y alteraciones en habilidades pragmáticas (Luyster et al., 2008). Además, se ha mostrado que las habilidades de los niños para comunicarse de forma funcional están relacionadas con el estrés emocional de los padres de niños con TEA (Ello & Donovan, 2005). Sin embargo, no se ha profundizado en el estudio de la comprensión de dichos aspectos estructurales del lenguaje ni en las posibles repercusiones que estas limitaciones pueden tener sobre la CdVF.

Por ello, en el **sexto capítulo** de esta tesis investigamos la influencia de las habilidades receptivas del lenguaje en niños con TEA no verbales sobre la CdVF. Este capítulo amplía los resultados previos obtenidos en niños con TEA con nivel 1 de apoyo en una población con diferente nivel de severidad (niños con TEA con nivel de apoyo 3). En este estudio analizamos el impacto de los aspectos receptivos del lenguaje (p. ej., estructuras gramaticales, comprensión y comunicación no verbal) en la CdVF en una muestra de 52 familias españolas con hijos con TEA no verbales y con DT igualados en función del nivel de vocabulario comprensivo.

Parte IV: El papel de los padres y los hermanos en el trastorno del espectro autista

Como se ha constatado en la literatura, cuando el TEA irrumpe en una familia, el riesgo de experimentar una baja CdVF aumenta. Sin embargo, aunque de forma clásica se han investigado factores que afectan a la CdVF de forma negativa, en los últimos años, algunas investigaciones se están acercando hacia un posicionamiento más comprensivo de la CdVF en el TEA (Vasilopoulou & Nisbet, 2016). Este posicionamiento comprensivo busca encontrar factores protectores que mitigan el impacto de tener un hijo con TEA. Dado que

la CdVF se considera como uno de los predictores más fiables de la condición de vida familiar y refleja el nivel de bienestar general y las relaciones entre los principales componentes de una familia, no sólo se deben analizar las variables relacionadas directamente con niños con TEA sino también incidir en otras variables relacionadas con el resto de familiares (Mannan, Summers, Turnbull, & Poston, 2006). Por ello, en la cuarta parte de esta tesis perseguimos el **objetivo** de investigar en qué medida diferentes variables en los familiares de niños con TEA (padres y hermanos) pueden mejorar la satisfacción en la CdVF. En esta parte presentamos un total de cuatro estudios, en los que investigamos tanto los efectos de una intervención mediada por padres como las diferencias y el potencial efecto protector de diversas variables relacionadas con los familiares de primer grado en la CdVF (padres y hermanos).

Como se deriva de la investigación y de los resultados presentados en el sexto capítulo de esta tesis, las habilidades lingüísticas y comunicativas de los niños con TEA −nivel de apoyo 3− ejercen un impacto sobre la CdVF. Por ello, promover una mejora en dichas habilidades es otro de los aspectos relevantes y necesarios en la literatura científica más actual. Sin embargo, a la hora de intervenir en niños con TEA no sólo hay que considerar al propio niño. En las intervenciones en esta población hay diversos agentes que deben incluirse para generalizar los resultados a otros ámbitos y contextos. Uno de estos agentes son los padres, que comparten la mayor parte del tiempo libre que tienen los niños fuera del ámbito escolar o de terapia. Por ello las intervenciones mediadas por padres son un tipo de intervenciones especializadas que se centran en la enseñanza y entrenamiento de ciertas habilidades en los padres que pueden mejorar los niveles de estrés, su percepción de autoeficacia y confianza y que se espera que tengan un impacto directo sobre el bienestar familiar y la CdVF (Ayuda-Pascual, Llorente-Comi, Martos-Perez, Rodriguez-Bausa, & Olmo-Remesal, 2012; Feinberg et al., 2014).

Algunas de estas intervenciones se han centrado en el manejo de las habilidades de comunicación y lenguaje (p. ej., Responsive Teaching, RT; Mahoney & McDonald, 2007) y han informado de mejorías en la calidad de las interacciones de los padres (Karaaslan, Diken, & Mahoney, 2013; Karaaslan & Mahoney, 2013; Mahoney, Nam, & Perales, 2014; Mahoney & Perales, 2003, 2005; Mahoney, Wiggers, Nam, & Kralovic, 2014). Otras intervenciones han tenido como objetivo la mejora en el manejo conductual (p. ej.,

Applied Behavior Analysis, ABA; Cooper, Heron, & Heward, 2007), informando también de mejorías en las habilidades de los padres, su sentido de competencia y su satisfacción como padres (Dillenburger, Keenan, Gallagher, & McElhinney, 2004; Green et al., 2010; Kasari, Gulsrud, Wong, Kwon, & Locke, 2010; Kasari, Gulsrud, Paparella, Hellemann, & Berry, 2015). Adicionalmente, otros trabajos han abarcado el estrés en padres, aunque de forma indirecta. Estos estudios han encontrado un descenso en el estrés parental y un incremento en la auto–eficacia tras participar en una intervención con padres (Feinberg et al., 2014).

Sin embargo, las intervenciones mediadas por padres suelen ser muy demandantes en tiempo y la adherencia podría verse comprometida. Este hecho se explica debido a las altas demandas que ya tienen que afrontar los padres de niños con TEA (como acompañarlos a terapias, colegios o centros de salud) (Roberts & Ridley, 2004). Por lo tanto, se hace necesario analizar y promover intervenciones breves en padres de niños con TEA que identifiquen posibles vías que puedan ayudar a los padres a mejorar su CdVF de forma eficiente en pocas sesiones.

Por ello, en el **séptimo capítulo** de esta tesis analizamos los resultados de una intervención breve mediada por padres cuyo propósito es incrementar las respuestas de los padres a los intentos comunicativos de sus hijos con TEA no verbales, la mejora en las habilidades de manejo de los problemas conductuales de sus hijos y el control del estrés paterno. En este estudio participaron un total de 40 familias de niños con TEA no verbales. Este capítulo tiene como objetivo el ofrecer una intervención breve mediada por padres en la que se desarrollan cuatro componentes principales: conocimiento sobre TEA, manejo del lenguaje y la comunicación, manejo conductual y manejo del estrés parental. Además, evaluamos tanto la adherencia a la intervención como el impacto que tiene la intervención sobre las habilidades paternas (como el manejo de la comunicación, conducta y el estrés parental) y en la CdVF tras participar en una intervención mediada por padres de 6 semanas de duración.

Por otro lado, algunos estudios constatan que los padres de niños con TEA que tienen un alto apoyo social también informan de una mejor salud mental y tienen una percepción más positiva de su CdVF (Khanna et al., 2011; Kuhlthau et al., 2014). Sin embargo, el apoyo social se puede ver limitado en estas familias debido a la gran carga y el

estrés diario al que están sometidas. Se hace necesario por tanto ofrecer variables individuales que protejan su CdVF. Además, otro aspecto fundamental al que los padres deben hacer frente, es la gran cantidad de información que reciben (ej., nuevos tratamientos, nuevas investigaciones y avances, nuevas informaciones desde las propias asociaciones de autismo y procedentes de otros padres y/o profesionales) y que deben integrar para tomar decisiones tan importantes como el tipo de intervención o tratamiento al que van a adherirse o la escolarización de su hijo/a (Romanzyk & Gillis, 2005). En este sentido, se hace necesario detectar habilidades o destrezas que puedan ejercer un impacto positivo sobre la toma de decisiones y que promuevan una mejoría en la CdVF.

Concretamente, en el **octavo capítulo** de esta tesis exploramos el potencial efecto de una de las variables que se ha demostrado que ejerce un efecto protector en la toma de decisiones en la población general: las habilidades numéricas (Cokely et al., 2018). Por este motivo, en este capítulo presentamos los resultados de un estudio de caso–control de 65 padres de niños con TEA y DT en el que analizamos diversas variables que pueden tener un efecto positivo sobre la CdVF como las habilidades numéricas tanto objetivas como subjetivas, el nivel educativo y el apoyo social.

Al igual que los padres de los niños con TEA, los hermanos de estos niños también ejercen una influencia dentro de la CdVF. Los hermanos de los niños con TEA tienen un 20% más de probabilidad de desarrollar TEA comparados con los hermanos de niños con DT (Ozonoff et al., 2011) y un 25% adicional de estos hermanos también presentan síntomas subclínicos de TEA (Georgiades et al., 2013; Messinger et al., 2013). Estos síntomas o características, que pueden diferenciarlos de los hermanos de niños con DT, pueden también ejercer un impacto sobre la CdVF. Por ello, evaluar las diferencias en el desarrollo de los hermanos de niños con TEA es imprescindible para conocer cómo pueden afectar dichas características a la CdVF. Algunas de las habilidades de desarrollo están íntimamente relacionadas entre sí. Por ejemplo, la adquisición del lenguaje y de la motricidad se desarrollan de forma paralela en el DT desde un punto de vista teórico (Alcock & Krawczyk, 2010). También existe evidencia científica de esta relación, que apoya la idea de que las habilidades motrices facilitan la interacción y la comunicación social (Bhat, Landa, & Galloway, 2011; Karasik, Tamis–LeMonda & Adolph, 2011). Belmonte et al.

(2013) informan que las habilidades motrices pobres podrían predecir menores avances en intervenciones de lenguaje expresivo.

Así, en el **noveno capítulo** de esta tesis realizamos una revisión meta–analítica de la literatura sobre las diferencias en las habilidades lingüísticas y motrices de los hermanos de niños con TEA como grupo, desde el nacimiento hasta los 3 primeros años de vida. Un total de 34 estudios fueron incluidos en el meta-análisis. En este capítulo resumimos las diferencias que se han publicado en hermanos de niños con TEA comparados con hermanos de niños con desarrollo típico en lenguaje (receptivo y expresivo) y en motricidad (fina y gruesa).

Debido a que la mayor parte de las investigaciones que evalúan el lenguaje y la motricidad en hermanos se realizan en edades muy tempranas, es necesario evaluar qué ocurre cuando crecen y llegan a la edad escolar. Sin embargo, hay muy pocos estudios que evalúen dichas habilidades en niños de mayor edad. Algunos estudios no encuentran diferencias entre los hermanos de niños con TEA y los hermanos de niños con DT a los 7 y 11 años (Drumm, Bryson, Zwaigenbaum, & Brian, 2015; Shepard et al., 2017). Una posible explicación es que, a medida que los niños se van desarrollando y estas diferencias se van disipando, aparecen otras habilidades que pueden ejercer un papel importante dentro de su desarrollo además de las habilidades lingüísticas y motrices. Por ejemplo, las estrategias de afrontamiento, el estrés y el apoyo social pueden ejercer un impacto sobre el ajuste de los hermanos de niños con TEA en edad escolar (Tsai, Cebula, & Fletcher–Watson, 2017). Por este motivo, en el **décimo capítulo** ampliamos los resultados presentados en el noveno capítulo y ofrecemos una visión general de las trayectorias del desarrollo en hermanos de niños con TEA en edad escolar. Específicamente, en este capítulo evaluamos las habilidades lingüísticas (expresivas y receptivas), socio–comunicativas, motrices (finas y gruesas) y el apoyo social de hermanos de niños con TEA en edad escolar. En concreto, en este estudio hemos examinado las diferencias en estas variables del desarrollo y su influencia en la CdVF en una muestra de 82 hermanos de niños con TEA y DT en edad escolar (entre los 4 y los 12 años).

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PART II

DEVELOPMENT OF CHILDREN BEFORE THE DIAGNOSIS OF AUTISM SPECTRUM DISORDER

CHAPTER 2

Chapter 2
Parents' concerns related to ASD and its diagnosis

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Timing of parents' concerns related to Autism Spectrum Disorder and its diagnosis: A mediation analysis.

Parents are the first to indicate concerns about their child's development in up to 80% of children with autism spectrum disorder (ASD). They often notice symptoms related to ASD around the first two years, but the average age of diagnosis is 3.5 years old. This study examined the relationships between parents' early concerns and the time lag between suspicion and diagnosis. Forty–eight Spanish–speaking families were enrolled in this study. Parents were asked about early signs that made them think that their child could possibly have ASD. Mediation analyses showed that the child's age at suspicion mediated between sibling status and the time lag between suspicion and a formal diagnosis ($\beta = –.53$, $p < .01$). Having another child with typical development accelerated parents' detection of ASD signs ($\beta = –.62$, $p < .001$). The number of social–communication concerns that parents detected mediated this relationship ($\beta = –.28$, $p < .01$). Parents who reported more social–communication concerns perceived these signs earlier, but have to cope with a longer time lag until diagnosis than those who reported more concerns related to restrictive and repetitive behaviors and interests, or other developmental concerns. Moreover, this relationship between concerns of ASD and the diagnoses was explained by the child's age. Training pediatricians on how to respond to parent questions and concerns could reduce the time lag between parents' concerns and diagnosis of ASD.

I. Introduction

An early diagnosis of autism spectrum disorder (ASD) is a critical need in psychology due to the significant role it plays in early intervention of ASD. Although the prevalence of ASD is currently estimated at around 1.5% of the general population, the average age of diagnosis in Europe is not until 3.5 years old (Developmental Disabilities Monitoring Network Surveillance Year 2010 Principal Investigators, & Centers for Disease Control and Prevention, 2014; Salomone, Charman, McConachie, & Warreyn, 2016). Given that an early diagnosis allows for early intervention and therefore, may lead to better outcome, there are continual efforts to provide earlier diagnosis. Diagnosing ASD is challenging and the current diagnostic

criteria are related to impairments in social interaction and communication, and restricted interests and repetitive behaviors (American Psychiatric Association, APA, 2013). These symptoms are usually not detected until the first two years of life. The purpose of this study was to examine the association between parents' early concerns related to ASD's criteria and age at diagnosis and to estimate to what extent socio–demographic variables could predict and/or mediate the time lag between early concerns and diagnosis.

In the absence of early biological markers for ASD, the early diagnosis of ASD is closely related to the recognition of the behavioral signs and symptoms of ASD. These signs and symptoms are present early in life and usually are detected by parents (Barbaro & Dissanayake, 2009; Guinchat et al., 2012). In Spain, parents are the first to see signs in up to 79 % of children with ASD at 18 months (Fortea–Sevilla, Escandell–Bermúdez, & Castro–Sánchez, 2013), but these early concerns are not specific to ASD.

The first signs of ASD generally become visible prior to 36 months of life (Guinchat et al., 2012); furthermore, some studies found that between 30–50% of early concerns related to ASD are expressed around 12 months of life, and 80–90% by 24 months (Volkmar, Chawarska, & Klin, 2008). In Spain, although parents may be aware of some signs early, often their child does not receive a diagnosis until 37 months of life (Díaz–Atienza, García–Pablos, & Martín–Romera, 2004; Salomone et al., 2016), and the time lag between early concerns and a formal ASD diagnosis is close to 13 months (Fortea–Sevilla et al., 2013).

The time that it takes for children to receive a diagnosis after initial concerns are expressed by parents can be influenced by the manner in which pediatricians elicit and answer parents' developmental concerns (Zuckerman, Lindly, & Sinche, 2015). Parents and pediatricians may find it difficult to differentiate ASD from other developmental disorders at an early stage (Monteiro et al., 2015). This is due to the similarity of symptoms of ASD to symptoms of other developmental disorders (e.g., speech and language disorders or general developmental delay). The wide heterogeneity of ASD and the similarity of symptoms between different disorders could generate a delay in diagnosis of ASD because pediatricians have a wait–and–see attitude (Daniels & Mandell, 2014; Klin, Klaiman, & Jones, 2015).

Language delay and communication difficulties are two of the initial ASD–related concerns frequently reported by parents (Meek, Robinson, & Jahromi, 2012). These

symptoms have been associated with early detection, but also with a later diagnosis (Volkmar et al., 2008; Zwaigenbaum, Bryson, Rogers, Roberts, Brian, & Szatmari, 2005). Other features directly related to ASD diagnosis have been delays in reaching developmental milestones (e.g. speech and language delay), slower rate of development, loss of previously acquired skills (e.g. speech regression) (Kishore & Basu, 2011; Mishaal, Ben–Itzchak, & Zachor, 2014), less frequent instances of smiling, orienting, and vocalizing to people (Maestro et al., 2005), lack of joint attention (Kishore & Basu, 2011), poor eye contact, lack of play and interaction, and hand flapping and toe walking (Mandell, Novak, & Zubritsky, 2005). Concerns related to repetitive behaviors often emerge later, and are often reported for children with other developmental disorders too (Mooney, Gray, & Tonge, 2006).

Beside these early difficulties, several factors have been shown to influence age of early concerns specifically related to ASD. For instance, having another child without ASD has been associated with an early detection and diagnosis, as parents could compare the development of the child at risk of ASD with their child with typical development (TD) (Fountain, King, & Bearman, 2011; Herlihy, Knoch, Vibert, & Fein 2015; Mishaal et al. 2014; Rosenberg, Landa, Law, Stuart, & Law, 2011). In general, higher maternal education would be associated with more knowledge of child development. Shattuck et al. (2009) showed that a lower level of maternal education and younger maternal age were associated with older children's ages for early concerns related to ASD. However, Mishaal et al. (2014) did not find significant correlations between parental age and educational attainment and age of ASD diagnosis. Furthermore, the gender of children with ASD has been another variable of scientific interest that might have an impact on the age in which children are diagnosed. Some studies have found no differences between gender and the age of diagnosis (Begeer et al., 2013; Fountain et al., 2011). Nevertheless, parents may expect more social behavior from girls, and encourage them to act more socially, which could delay detection.

There is no clear consensus about which features are more important in the earlier detection of ASD and which variables have a significant impact on the time lag between type of parents' early concerns and the diagnosis of ASD. It is important to consider features that improve clinical practice in eliciting these concerns and as a result, help to improve the early diagnosis of ASD. In addition, an early and accurate diagnosis helps parents cope with the stress related to the disorder (Pozo & Sarria, 2014). Thus, this retrospective study

investigates the association between parents' early concerns related to ASD symptom criteria and age at diagnosis. In particular, we investigated to what extent socio–demographic variables (i.e., type of parents' early concerns, parental level of education, parental age, child's age, sibling status, and child's gender) predict the time lag between early concerns and diagnosis.

II. Method

II.I. Participants

A total of 48 families from Granada (Spain) of a child with ASD (range: 4.1–10.1 years old) were enrolled in this study. All parents signed the informed written consent before participation, and The Ethics Committee of the University of Granada approved the methodology of the study. Inclusion criterion for each child was having an ASD diagnosis based on Diagnostic and statistical manual–text revision (DSM–TR–IV; APA, 2000) and the Autism Diagnostic Interview–Revised (ADI–R; Le Couteur, Lord, & Rutter, 2003) or Autism Diagnostic Observation Schedule–WPS (ADOS–G; Lord, Rutter, DiLavore, & Risi; 2002). The diagnosis was confirmed by an independent tester with the Guilliam Autism Rating Scales (GARS; Guilliam, 2004). In addition, one of our purposes was to investigate whether having an older sibling with typical development could have delayed or accelerated parents' doubts. As a result, an additional inclusion criterion was that for the children with older siblings, the older siblings must be deemed typically developing in order to be included in this analysis

Parents were interviewed and they completed an open–ended questionnaire measuring several aspects related to detection of ASD and diagnosis. This kind of questionnaire enabled us to obtain more spontaneous information and to run both qualitative and quantitative analyses (Chamak, Bonniau, Oudaya, & Ehrenberg, 2011; Guinchat et al., 2012). Families were asked (1) about early concerns that made them think that their child could have ASD, (2) what was their child's age when these signs were detected, and (3) when the child received the diagnosis of ASD.

II.II. Coding procedure and analysis

We focused on parents' early concerns about ASD. The coding procedure consisted of breaking down early concerns into 2 fields related to two domains from DSM–5 (APA, 2013): (1) Social communication (SC), and (2) restrictive and repetitive behaviors and interests (RRBI). Moreover, because many parents usually reported concerns that were not specific to autism criteria (Guinchat et al., 2012), we included a third category (3) other concerns non–specific to ASD (OC). The operational definitions of what variables were considered as SC, RRBI, or OC are mentioned in Table 2.1. Two independent coders identified the three different types of concerns. Mean percentage agreement (based on Cohen's Kappa inter–rater agreement measurement) was over 95% for all symptoms. Disagreements were resolved through discussion.

Table 2.1. First signs related to ASD reported by parents

Categories	Parents' early concerns about ASD
SC	Does not talk; impression of deafness; no language; language appeared later; language regression; language comprehension problems; no response to name; no reaction to social solicitation; no response to demands or questions; lacks social engagement; no joint attention; would not share games with peers (social reciprocity); dislikes being cuddle (social avoidance); no pointing; no eye contact; no gaze; no imitation.
RRBI	No interest in toys and games; only interest in strange and specific objects; resistant to change; stereotypies; strange behavior; hand–flapping; temper tantrums; walks on tiptoes; scared by certain noises; places hands over ears.
OC	Cries all the time; delayed walking; sleeping problems (routines); feeding problems (selectivity); screams for no reason; hyperactivity.

Note. SC= Social communication signs; RRBI= Restrictive and repetitive behaviors and interest signs; OC= Other developmental concerns non–specific to ASD.

II.III. Data analysis

All statistical analyses were performed using Statistica 13 software. To evaluate relationships between the two outcome variables (i.e., child's age at suspicion, and time lag between suspicion and diagnosis) and four predictor variables (i.e., sibling status, SC, RRBI, and OC) bivariate Pearson correlations were conducted. To determine the influence of each predictor, we run several multiple regression analyses. Furthermore, to examine potential meditational effects, path analyses were computed. Parental age, education, and child's gender were entered as covariates. We consulted the bootstrapping method with bias–corrected confidence estimates to test our results. The 95% confidence interval of the indirect effect was obtained with 5000 bootstrap samples.

III. Results

Forty–eight families were enrolled in this study. Descriptive statistics for all study variables are presented in Table 2.2. From the whole sample, 12.5% (n = 6) reported child's age at diagnosis less or equal to 24 months; 54.2% *(*n = 26) reported child's age at diagnosis between 25 and 35 months; the remain 33.3% (n = 16) reported child's age at diagnosis longer than 36 months.

Table 2.2. Socio–demographic variables from families with a child with ASD

Families with a child with ASD	n = 48
Child's gender	
Male	34 (71%)
Female	14 (29%)
Child's age at suspicion [a] (SD)	22.58 (.59)
SC (SD)	3.17 (1.22)
RRBI (SD)	1.25 (1.27)
OC (SD)	.75 (.86)
Time lag between suspicion and diagnosis [a] (SD)	9.29 (.91)
Age of Diagnosis [a] (SD)	32.25 (1.10)
Parental age [b] (SD)	35.42 (.64)
High Parental education [c]	41 (85%)
Sibling status [d]	22 (46%

Note. SC = Social communication signs; RRBI = Restrictive and repetitive behaviors and interest signs; OC = Other developmental concerns non–specific to ASD.
[a] Months. [b] Years. [c] Post–secondary and university education. [d] Having an older sibling with typical development.

Bivariate Pearson correlations are presented in Table 2.3. Correlations between parents' early concerns (i.e., SC, RRBI, and OC), parental level of education, parental age, child's age at suspicion, time lag between suspicion and diagnosis, sibling status, and child's gender were computed. We focused on two of these variables: Child's age at suspicion and time lag between suspicion and diagnosis. Younger child's age at suspicion was related to more number of SC reported by parents ($p < .05$), longer time lag between suspicion and diagnosis ($p < .001$), and having an older sibling with typical development ($p < .001$). Additionally, longer time lag between suspicion and diagnosis was positively related to having an older sibling with typical development ($p < .05$).

Table 2.3. Correlations coefficients values between symptoms related to ASD, and variables related to parents and children

Variables	1	2	3	4	5	6	7	8	9
1. SC signs	–								
2. RRBI signs	–.09	–							
3. OC signs	–.12	–.06	–						
4. Parental level of education	–.14	–.25	.09	–					
5. Parental age	.01	.16	.08	–.06	–				
6. Child's age at suspicion	–.42**	.27	.16	–.21	.04	–			
7. Time lag between suspicion and diagnosis	.18	–.13	–.23	–.16	–.02	–.62**	–		
8. Sibling status	.12	–.07	–.03	.15	.03	–.68**	. 38**	–	
9. Child's gender	–.16	.13	.03	–.12	.29*	.16	–.18	–.23	–

Note. SC = Number of social communication signs; RRBI = Number of restrictive and repetitive behaviors and interests signs; OC = Other developmental concerns non–specific to ASD.
*$p < .05$; **$p < .01$

We then conducted two multiple linear regression analyses with outcomes child's age at suspicion, and time lag between suspicion and diagnosis respectively. The results of these regressions are presented in Table 2.4, including standardized regression coefficients for each predictor.

Child's age at suspicion. This model accounted for 56% of the total variance, $F(4, 42) = 15.612$, $p < .001$. From all potential predictors, the number of SC and having an older sibling with typical development were the statistically significant variables ($p < .005$, and $p < .001$ respectively).

Time lag between suspicion and diagnosis. This model accounted for 35% of the total variance, $F(5, 41) = 5.896$, $p < .001$. In this model, from all potential predictors, child's age at suspicion was the only statistically significant variable ($p < .05$).

Table 2.4. Regression analyses to determine the influence of each predictor on child's age at suspicion and time lag between suspicion and diagnosis

	Outcomes					
	Child's age at suspicion			Time lag between suspicion and diagnosis		
	β	SE	p	β	SE	p
Child's age at suspicion	–	–	–	–.53*	.18	.007
SC	–.28*	.55	.008	–.06	.47	.640
RRBI	.17	.52	.104	.01	.62	.991
OC	.09	.77	.368	–.17	.90	.176
Sibling Status	–.62**	1.00	<.001	.06	1.61	.724

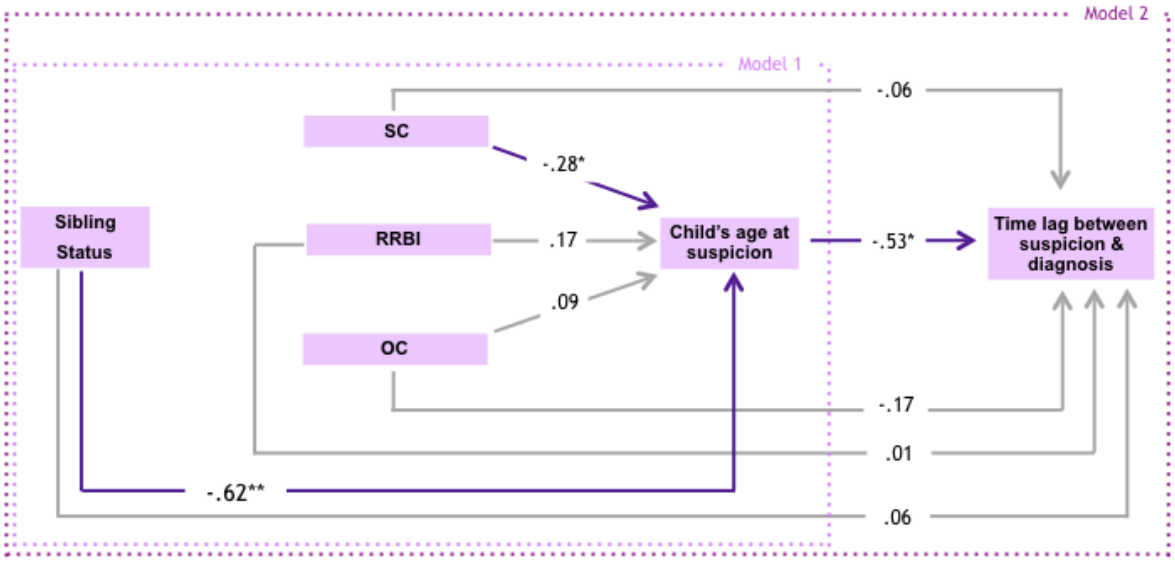
SC = Number of social communication signs; RRBI = Number of restrictive and repetitive behaviors and interests signs; OC= Other developmental concerns non–specific to ASD.
* $p < .01$, ** $p < .001$

Path analysis. To describe a comprehensive model, we carried out path analyses. We tested a path model that was based on the regression analyses and was consistent with the actual timeline of events (i.e., that having an older sibling with typical development influences the potential signs noticed by parents, which can in turn influence their suspicions of ASD, and finally the diagnosis).

The number of SC was related to child's age at suspicion, suggesting that it could be a potential mediator of the relationship between sibling status and the child's age at suspicion (see Figure 2.1). To test this possibility, we estimated indirect effect on number of SC via child's age at suspicion (see Table 2.5). The results showed that there was a significant indirect effect of number of SC on child's age at suspicion via sibling status (95% confidence interval excluding 0).

Regarding the second model, child's age at suspicion was related to time lag between suspicion and diagnosis, suggesting that it could be a potential mediator of the relationship between sibling status and the time lag between suspicion and diagnosis. To test this possibility, we estimated indirect effect on child's age at suspicion via time lag between suspicion and diagnosis. The results showed that there was a significant indirect effect of child's age at suspicion on time lag between suspicion and diagnosis through sibling status

(95% confidence interval excluding 0). Moreover, results showed another significant indirect effect of SC on child's age at suspicion through sibling status.



Note: Coefficients are standardized β; SC= Number or social communication signs; RRBI= Number of restrictive and repetitive behaviors and interest signs; OC= Other developmental concerns non–specific to ASD. * p < .01; ** p < .001

Figure 2.1. Results from mediation analyses

Table 2.5. Estimated indirect effects from sibling status via path analysis

	Outcomes					
	Child's age at suspicion			Time lag between suspicion and diagnosis		
	Coeff	95% CI		Coeff	95% CI	
		LL	UL		LL	UL
Child's age at suspicion	–	–	–	3.199	.098	6.810
SC	–.320	–1.363	–.257	–.016	–.744	.435
RRBI	–.119	–1.122	.383	–.007	–.435	.346
OC	–.026	–.647	.211	.043	–.422	.853

Note. SC = Number of social communication signs; RRBI = Number of restrictive and repetitive behaviors and interests signs; OC = Other developmental concerns non–specific to ASD.

IV. Discussion

To our knowledge, this is the first study that includes several variables to develop a mediation model between parents' early concerns, sibling status, child's age, and time lag between suspicion and diagnosis. Our study suggests that some variables are mediating over others that could explain the period of time between the suspicion and the diagnosis of ASD.

First, our results indicated that if the child with ASD has an older sibling with typical development, this precipitates parents' detection, as other authors have found (see for example, Herlihy et al., 2015; Mishaal et al., 2014). This can be explained by the fact that those parents who have another child, can compare the development of their child with ASD with a typical development, and can detect these signs earlier. This relationship is partially mediated by the type of concerns noted by parents. If we include SC, RRBI, and OC concerns in our model as predictors, the strength of the association between sibling status and child's age at suspicion decreases. Moreover, as previously has been found by Volkmar et al. (2008) and Zwaigenbaum et al. (2005), concerns related to SC and language hasten the suspicion of ASD. This can be explained by the fact that SC concerns such as smiling and orienting to others or speaking are detected earlier by parents because they appear before than those symptoms related to RRBI, such as interest in toys, resistance to change, stereotypic

behaviors, or motor delay (Herlihy et al., 2015). On the other hand, our results did not find statistically significant other concerns such as RRBI, OC, or socio–demographic variables that were included in the model (e.g. parental level of education, parental age, and child's gender). One possible explanation might be that our sample showed similar levels of education and parental age.

Second, we found that the child's age at suspicion is a mediator between parents' early concerns and time lag between suspicion and diagnosis. This mediation can be explained by the fact that the age of suspicion is crucial in the ASD diagnosis. We need to keep in mind that these children have been diagnosed through DSM–IV–TR (APA, 2000), in which it was stated that the onset of ASD must be prior to 3 years of age, but the diagnosis was not usually made until 3 years of age (Díaz–Atienza et al., 2004). This further explains why pediatricians decided to wait until children were 3 years old. Pediatricians may have understood that every child had their own pace of development and some of the early concerns could be related to other disorders (such as specific language disorder). So, it seems that parents may help their child receive a diagnosis earlier by relaying their concerns to their pediatricians but because every child has his/her own rate of development, pediatricians chose to wait and see (Klin et al., 2015).

Finally, we found a relationship between sibling status and time lag between suspicion and diagnosis. Our results are partially consistent with those obtained by Fountain et al. (2011), Herlihy et al. (2015), and Rosenberg et al. (2011), because having another child without ASD was associated with an early detection. Surprisingly, our results showed that having another child make longer the time lag between suspicion and diagnosis. However, this relationship was totally mediated by the time when ASD was suspected and parental concerns (i.e., SC, RRBI, and OC). We found that the earlier the age of suspicion, the more time lag between the suspicion and the diagnosis of ASD they have to wait. This explanation sheds light on the previous association. This would explain why children who have older siblings have to endure longer wait times in order to receive an ASD diagnosis.

This study has several limitations. First, by using an open–ended questionnaire there is a possibility of parental recall bias and parents' limited knowledge about ASD. Nevertheless, this approach is an efficient way to collect the most spontaneous concerns and retrospective parental information has been widely used as a source of information about

ASD (Barbaro & Dissanayake, 2009). In that sense, we encourage future research to observe if parents report the same kind of concerns by comparing different procedures, such as a checklist and an open–ended questionnaire. Second, our sample consists of families with a high level of parental education, so we have not been able to assess whether educational level affects their ability to detect ASD signs. Third, due the fact that our parental sample belonged to the same age cohort, we could not evaluate this variable. It would be interesting to study possible differences between the signs detected by parents from different age cohorts. Maybe younger parents would detect more signs because of the huge amount of information about ASD available nowadays.

We conclude that there are some variables related to the time lag between parents' early concerns and diagnosis of ASD. These relationships are not linear, but mediating variables are included. Parents who detected signs earlier reported more concerns related to SC than those who detected ASD later. Moreover, parents earlier detected ASD signs if their child had an older sibling with typical development. However, when they shared their concerns with the pediatrician, they often received the advice of wait and see. This could mean that pediatricians should give more attention to these signs and this also underscores the importance of promoting awareness about early signs of ASD.

Our study and others (Daniels & Mandell, 2014; Guinchat et al., 2012) have shown that a significant delay exists between the moment when parents first become concerned about their child's development and the moment when their child receive a formal ASD diagnosis. We consider that training pediatricians on how to respond to parents' questions or concerns might enhance their response to parents' concerns and consequently, decrease the time until diagnosis of children with ASD. One potential response includes promoting parents' informed decisions and advising them to contact a specialist as soon as concerns arise.

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CHAPTER 3

Chapter 3

Patterns of vocalizations at 14 months in children at–risk for ASD

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Infants at–risk for autism spectrum disorder: Patterns of vocalizations at 14 months

Differences in the early development of children are crucial for early detection of autism spectrum disorder (ASD). Previous studies have shown large differences between children later diagnosed with ASD and their typically developing peers in the early use of canonical vocalizations (i.e., vocalizations that include well–formed consonant–vowel syllables) and the use of vocalizations for communicative purposes. In this prospective study, we examined the extent to which infant vocalizations at 14 months would predict Autism Diagnostic Observation Schedule (ADOS) diagnostic symptom groups, that is, Autism, Spectrum, and Non–ASD, for 82 community identified at–risk infants at 23 months. Thirty–minute video samples were coded with the intention to categorize and quantify speech (canonical/noncanonical and directed/nondirected) and nonspeech vocalizations (atypical, distress, and pleasure vocalizations). Our results revealed that more canonical directed (OR = 1.039, p = .036), and fewer noncanonical directed (OR = .607, p = .002) and noncanonical nondirected (OR = 1.200, p = .049) vocalizations were associated with a greater likelihood of being in the Non–ASD group versus the Autism group, with no variables significantly predicting Autism versus Spectrum group membership. Despite some statistically significant findings, models performed poorly in classifying children into correct ASD symptom group at age 23 months based on vocalizations at 14 months. Thus, the utility of infant vocalizations alone for predicting toddler clinical outcomes among infants initially identified at an elevated risk for ASD appears limited; however, considering the structure and function of early vocalizations combined with other early developmental and behavioral features may improve the confidence for clinicians in making an early diagnosis of ASD.

I. Introduction

Understanding the early behavioral symptoms of autism spectrum disorder (ASD) not only can elucidate the developmental course of infants later diagnosed with this disorder, but also contribute to early identification and intervention planning. Compared to their peers, infants

who go on to be diagnosed with ASD show significant group differences related to a wide range of behavioral risk markers, including eye contact, imitation, shared affect, orienting to name, response to caregiver language input, joint attention, gestures, and sensory modulation difficulties (Baranek, 1999; Cohen et al., 2013; Luyster, Seery, Talbott, & Tager–Flusberg, 2011; Mulligan & White, 2012; Ozonoff et al., 2010; Watson, Crais, Baranek, Dykstra, & Wilson, 2013; Zwaigenbaum et al., 2005). However, it is important to note that individual behavioral risk markers for ASD are rarely specific to categorical differences in clinical outcomes, but instead reflect different distributions in the frequency or intensity of behaviors for groups of infants later diagnosed with ASD versus those who are not. Thus, early ASD symptoms are generally manifested as more subtle differences across multiple behaviors rather than a clear presence or absence of one or two behaviors. This is consistent with the wide heterogeneity in the specific behavioral symptoms and severity of the impact of these symptoms among older children diagnosed with ASD (e.g., Johnson, Myers, & American Academy of Pediatrics Council on Children With Disabilities, 2007).

Both to advance understanding the early development of children prior to an ASD diagnosis and to contribute to the applicability of research findings to the clinical goals of early identification and intervention planning, it is important to know about the broad range of early behaviors that show different distributions among infants later diagnosed with ASD and those who are not. For the purposes of this paper, we define infants as children under the age of 24 months. Findings from several studies address the use of speechlike vocalizations (i.e., vocalizations with vowels or consonant–vowel combinations recognizable by adults as similar to sounds used by mature speakers) in infants with ASD. Retrospectively, parents of children with ASD indicate that their children's use of speech–like vocalizations at around 12 months of age differed significantly from what is reported by parents of typically developing children; however, parents' recall of their infants' vocalizations at 12 months does not consistently discriminate children with ASD from those with other developmental disabilities (DD) (Watson et al., 2007; Werner, Dawson, Munson, & Osterling, 2005).

Findings using other methods have been fairly consistent with the findings from retrospective parent reports related to the use of speech–like vocalizations. For example, through a retrospective analysis of early home videos made by parents of children with later diagnoses of ASD versus children later confirmed to be typically developing, Patten et al.

(2014) reported large differences at both 9–12 months and 15–18 months. Infants with ASD produced fewer speech–like vocalizations (i.e., lower volubility) and less canonical babbling (syllables including well–formed consonants and vowels) than typically developing infants. Typically developing infants were 17 times more likely to meet an established criterion for being in the canonical stage (i.e., >.15 ratio of canonical syllables to all syllables; Oller, 2000) than infants with ASD at 9–12 months, and still six times more likely to meet this criterion level at the relatively late age of 15–18 months. Although Patten et al. did not include a comparison group of children with other DD, other studies have looked at this comparison. An earlier retrospective video analysis of first birthday parties yielded no between group differences in vocalizations among infants later diagnosed with ASD only, ASD plus intellectual deficits, intellectual deficits only, or with typical development (Osterling, Dawson, & Munson, 2002); however, it is not clear whether vocalizations in this study were confined to speech–like vocalizations or included both speech–like and nonspeech–like vocalizations. In a later study, researchers analyzed home videos of first and second birthdays for three groups of infants: (a) infants with ASD with reported early onset of symptoms; (b) infants with ASD whose parents reported regression after 12 months of age; and (c) typically developing infants [Werner & Dawson, 2005]. In this study, infants with ASD with regression showed higher rates of “complex babbling” (which was undefined in the study) at 12 months than infants with early onset ASD and used complex babbling twice as frequently as typically developing infants. By 24 months, however, typically developing infants used twice as much complex babbling as either subgroup of infants with ASD, who by this age were very similar to one another.

In a prospective study of toddlers identified through a community screening as being at heightened risk for communication disorders, including ASD, Plumb and Wetherby (2013) observed that of the vocalizations produced by 18– to 24–month–old infants later diagnosed with ASD, a lower proportion contained speech–like sounds compared to their typically developing peers, with a similar but nonsignificant trend in comparison to infants later diagnosed with other DD. Additional evidence from a study of infants at elevated risk for ASD due to having an older sibling diagnosed with ASD indicated these infants produced fewer total speech–like vocalizations at 6, 9, and 12 months, as well as a lower proportion of vocalizations with canonical syllables at 9 months, compared to low–risk infants (i.e., those

who had an older sibling without ASD (Paul, Fuerst, Ramsay, Chawarska, & Klin, 2011). In this study, however, it is not clear whether group differences were driven largely by the high–risk infants who went on to be diagnosed with ASD, or instead represented a more generalized reduction in speech–like vocalizations and canonical syllables among high–risk infants.

Taken together, the existing studies lead to an expectation of generally fewer speech–like vocalizations and canonical syllables among infants later diagnosed with ASD compared to their typically developing peers. What is less clear is the extent to which specific patterns in early speech–like vocalizations may predict clinical outcomes among high–risk infants, either those who are at familial risk for ASD or those who screen as at–risk for ASD based on early behavioral symptoms related to ASD.

In addition to studying speech–like vocalizations, some researchers have also examined whether the production of nonspeech vocalizations is different for infants with ASD compared to other infants. Evidence suggests that a larger proportion of vocalizations of infants with ASD are nonspeech vocalizations than for typically developing infants (Plumb & Wetherby, 2013). Toddlers with ASD (16 to 36 months old) have been found to produce speech–like vocalizations at similar rates to 12–month–old typically developing peers matched for language level, but to produce more atypical nonspeech vocalizations than either language level matched or chronological age–matched typically developing peers (Schoen, Paul, & Chawarska, 2011). Atypical vocalizations included squeals, growls, and yells. Infants at heightened risk for ASD based on having an older sibling with ASD showed a similar pattern of producing more nonspeech vocalizations than low–risk infants across the ages of 6, 9, and 12 months (Paul et al., 2011).

A final question of particular relevance to the current study is whether infants later diagnosed with ASD are different from other infants in their use of vocalizations for communicative purposes. A lower frequency of vocalizations directed to others has been observed in infant siblings of children with ASD who themselves were later diagnosed with ASD compared with infants with typical development at 12 months but not at 6 months of age (Ozonoff et al., 2010). In addition, another study found that infants with an older sibling with ASD (compared with low–risk infants) used fewer nonword speech–like vocalizations for communicative purposes at both 13 and 18 months (Winder, Wozniak, Parlade, & Iverson, 2013).

Thus, the available evidence suggests that low volubility of speech–like vocalizations, delayed onset or restricted use of canonical babbling, a greater use of nonspeech vocalizations (including unusual vocalizations), and fewer speech–like vocalizations directed to others are potential markers of emergent ASD. However, relatively few studies have compared vocalizations of infants later diagnosed with ASD to infants diagnosed with other DDs. Also, in the few available studies, much more consistent differences have emerged between infants with ASD outcomes when compared to typically developing infants than to infants with other DD. In addition, studies of infant siblings of children with ASD have not clarified how specific the differences in any of these vocalization features are to infant siblings who go on to be diagnosed with ASD versus those who exhibit broader autism phenotype characteristics and/or those who do not show elevated ASD symptoms. In general, very limited prospective research is available on infants identified as at–risk for ASD through community screenings. Specific to the focus of the current study, we only identified one such study providing data on infant vocalizations, with data collected when infants were 18 to 24 months of age (Plumb & Wetherby, 2013).

In the current study, we extend the available evidence related to vocalizations among infants at–risk for ASD by analyzing data from a sample identified by a community screening with the First Year Inventory, version 2.0 (FYIv2.0; Baranek Watson, Crais, & Reznick, 2003; Reznick, Baranek, Reavis, Watson, & Crais, 2007). Our aim was to examine the extent to which infant vocalizations would be related to later ASD outcomes within this at–risk sample. Vocalizations were recorded during an assessment of the infants when they were between the ages of 13 and 15 months (mean of 13.74). Coded vocalizations were then used to predict ASD symptom group membership for the infants at 20 to 25 months of age (mean of 22.57). These symptom groups were based on Autism Diagnostic Observation Schedule Module 1 algorithm scores at the second assessment, using the cutoff criteria for “Autism,” “Spectrum,” and “Non–ASD.”

Our research question was as follows: To what extent is ASD symptom group membership at 23 months predicted by infant vocalizations at 14 months, including their (a) speech–like vocalizations—canonical/noncanonical and directed/nondirected, (b) nonspeech vocalizations— atypical, distress and pleasure, and (c) volubility.

II. Method

The current study was a secondary analysis of data collected as part of an efficacy study of a parent–mediated early intervention for infants at–risk for ASD, led by the second and last authors (Watson et al., 2017), which we will refer to as the parent study. The parent study was designed as a pretest–posttest randomized controlled trial. The vocalization data coded for the current study were drawn from the pretest assessment.

II.I. Participants

A total of 82 participants (55 male, 27 female) between 13 and 15 months at entry into the parent study were included in the present analyses. These infants all met criteria for being at–risk for ASD based on a parent report screening tool (see below). As part of participant recruitment, infants with identified genetic conditions associated with developmental disorders (e.g., Down syndrome), severe physical or sensory impairments, or in families speaking English less than 50% of the time in the home were excluded from the study.

II.II. Measures

The *First Year Inventory version 2.0* (FYIv2.0; Baranek et al., 2003) is a 63–item parent questionnaire, normed for 12–month–old infants and designed to screen for risk of ASD. The FYIv2.0 items assess infant behaviors in two broad domains: social–communication and sensory regulation. Using state birth records, the FYIv2.0 was distributed to families in the catchment area for the parent study approximately a week prior to each infant's first birthday. To be eligible for the parent study, infants had to meet pre–established criteria for being at–risk for later diagnosis of ASD on this questionnaire (i.e., 94%ile for risk in the social–communication domain and 88%ile in the sensory–regulatory domain).

The *Mullen Scales of Early Learning* (MSEL; Mullen, 1995) is an examiner–administered developmental assessment standardized for children from birth to 48 months. It has four scales: Visual Reception, Receptive Language, Expressive Language, and Fine Motor. We used the Visual Reception T–score (a standard score with a mean of 50 and a standard deviation of 10) to characterize the infants' nonverbal cognitive functioning.

The *Communication and Symbolic Behavior Scales– Developmental Profile* (CSBS; Wetherby & Prizant, 2003) is an examiner administered assessment of social, communication, and symbolic skills standardized for infants from 9 to 24 months of age. We used the total standard score for this tool, with a mean of 100 and a standard deviation of 15, to characterize the infants' general communication functioning.

The *Autism Diagnostic Observation Schedule* (ADOS; Lord, Rutter, DiLavore, & Risi, 1999) is a semistructured observational measure to evaluate symptoms of ASD in children and adults. At the initiation of the parent study, a validated version of the Toddler Module, currently recommended for assessing infants and toddlers, was not yet available; thus, all infants were assessed at 23 months using the ADOS Module 1, scored using the revised algorithms (Gotham, Risi, Pickles, & Lord, 2007). To examine the association of vocalizations with later outcomes, we subgrouped the infants into one of three symptom categories, based on their ADOS algorithm scores: (a) Autism, (b) Spectrum, or (c) Non–ASD. Although the Diagnostic and Statistical Manual of the American Psychiatric Association—5th edition [American Psychiatric Association, 2013] does not recognize diagnostic subcategories within ASD, we maintained the divisions between Autism, Spectrum and Non–ASD as a broad indicator of ASD symptom severity at 23 months. It is important to recognize that these categories did not represent definitive clinical diagnoses for these infants, due to an a priori decision in the parent study not to give clinical diagnoses at posttest due to the young age of the children. We will refer to these groups as "ASD symptom groups" to reinforce the point that they are based on the quantitative outcomes of the ADOS algorithm scoring. Although it is reasonable to question whether the infants' ASD symptom groups at 23 months may have been affected by exposure to the experimental parent–mediated intervention tested in the efficacy study, the parent study documented no statistically significant differences between the experimental and control groups for ADOS algorithm total scores at 23 months (Watson et al.,2017). As further confirmation of the lack of a clinically important impact on autism symptoms, the effect size for the comparison of ADOS algorithm scores was small, $d = .18$. Thus, for the purposes of the current study, we did not include treatment group in our models.

II.III. Procedures

As part of the parent study, participants were evaluated with the MSEL (Mullen, 1995) and the CSBS–DP (Wetherby & Prizant, 2003) at 14 and 23 months. Parents and infants also were videotaped during a 10– min free–play interaction with access to a standard set of toys at both assessment time points. At 23 months, participants were evaluated with the ADOS (Lord et al., 1999 by trained, experienced examiners who met established research standards for reliability of scoring.

II.IV. Coding Procedure and Observer Agreement

Samples were coded from audiovisual recordings of the evaluation in a clinical context, so the audio–video quality was appropriate and all videos were recorded in similar rooms, with the same situational context and toy set. All participants included in this study except two had 30 min of video recording. The first 10 min of video coded for vocalizations were from the parent– infant free play session. The next 20 min of coded video were recorded while an examiner assessed the infant with the CSBS–DP. We chose these two contexts in an effort to get a more representative sample of the infants' vocalizations. That is, the parent–infant free play was more naturalistic, with parents only being asked to, "Play with your child as you would if you were at home," whereas the CSBS–DP involves a protocol through which an examiner attempts to elicit social, communication, and play behaviors from the infant via direct cues (e.g., questions and directions) and indirect strategies (e.g., showing toys to the infant and waiting to see how she/he will respond). Per standard administration procedures, the parent sits beside the infant during the CSBS–DP assessment and is encouraged to respond to the infant if she/he tries to engage with the parent.

Based on previous literature (Oller, Eilers, Neal, & Schwartz, 1999; Oller, Eilers, Steffens, Lynch, & Urbano, 1994; Patten et al, 2014; Paul et al., 2011; Scherer, Boyce, & Martin, 2013; Sheinkopf, Mundy, Oller, & Steffens, 2000), we coded two types of speech vocalizations (canonical and noncanonical) and three types of nonspeech vocalizations (atypical, distress, and pleasure vocalizations). Vocalizations were coded as discrete events. The onset of a vocalization occurred when the infant initiated the sound. A vocalization ended if there was at least one second break in the infant's vocalizing. Furthermore, we

coded whether the infant directed the vocalization to another person, as evidence of intentionality. Vocalizations were considered to be "directed" if the infant accompanied the vocalization with gestures (e.g. touching a person, pointing to an object, shaking head "no") and/or eye contact, if the vocalization occurred within an interactive context involving reciprocal vocalizations/verbalizations of the parent and infant (e.g., while both were looking at a book, or both were engaged in a game), or if the vocalization was an imitation of an adult's preceding vocalization (Colgan et al., 2006; Sheinkopf et al., 2000; Watson et al., 2013; Wetherby, Cain, Yonclas, & Walker, 1988). In the absence of these indicators of intentionality (e.g., not sharing interest with another person, not looking at another person's face/eyes, not touching person, no imitation), speech vocalizations were coded as nondirected. Each vocalization was coded, and frequencies of occurrence were calculated for each vocalization type. The decision–making process is shown in Figure 3.1 and operational definitions of vocalizations are provided in Table 3.1.

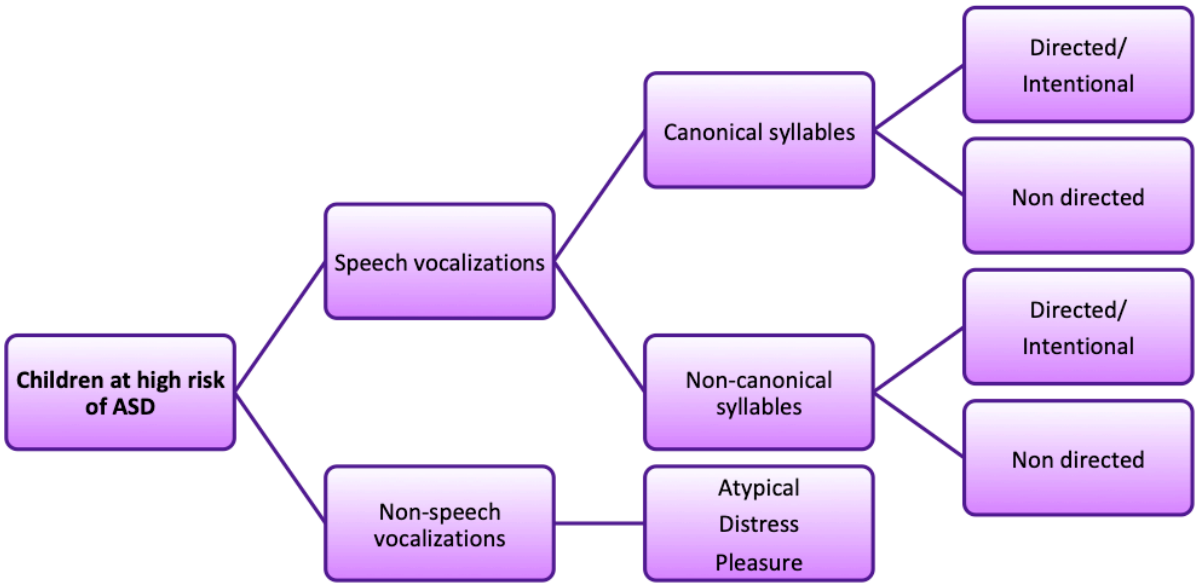

Figure 3.1. Coding tree for infant vocalizations

Table 3.1. Definitions of vocalizations used in the coding procedure

Vocalization	Definition
Speech	The production of consonants and/or vowels that have a speech–like vocal quality (Ozonoff et al., 2010; Paul et al., 2011).
Canonical vocalizations	Vocalizations that include at least one well–formed syllable (syllables containing the basic consonant–vowel (CV) structure, including single CV syllables such as [da], reduplicated sequences such as [bababa] and sequences with variable consonants such as [dʌgʌ]). Each canonical vocalization must have at least one full vowel–like element and at least one consonant–like element, and must have a rapid formant transition between consonant and vowel (examples, [ba][ata][nunu][di][dada] (Oller et al., 1999; Paul et al., 2011; Scherer et al., 2013).
Non–canonical vocalizations	Speech–like vocalizations that do not include any well–formed syllables with C–V structure; these vocalizations include those with marginal syllables (i.e., having a C–V structure but with a long C–V transition rather than a rapid formant transition), isolated consonants (mmmmmmm), speech–like full vowels, isolated vowels, and quasivowels. (Oller et al., 1994; Scherer et al., 2013; Sheinkopf et al., 2000).
Non–speech	Vocalizations characterized by the production of non–speech resonance and vocal quality (e.g., squeals, growls) without recognizable consonants (Paul et al., 2011).
Atypical	Squealing (vocalizations at high pitch), growling (vocalizations at low pitch, often in squeaky voice), glottal fricative and glottal stop sequences (vowels or quasivowels occurring in syllable–like sequences with glottal consonants, which require no supraglottal articulation), raspberries (labial trills or vibrates) (Oller et al., 1994).
Pleasure	Laughing or giggling, an audible vocalization that is usually associated with pleasure (Paul et al., 2011).
Distress	A vocalization associated with a negative emotional state like crying, whining, screaming or fussing (Paul et al., 2011).

Three coders who were blind to infant outcome (i.e., ADOS results at 23 months of age) completed the coding for this study. The three coders first completed a training program on identifying different kinds of vocalizations. They achieved 90% agreement on event–by–event coding during training. The samples used during training were not included in the analysis. All videos were randomized and distributed among the three coders. To assess interrater reliability, two coders independently coded 20% of the videotapes, selected at random. They were unaware of which videos would be coded for the evaluation of interrater reliability. Following independent coding of a video (these data were used to compute interrater reliability), disagreements were resolved through discussion, to help prevent coder drift across the data coding period. Reliability was estimated from intraclass correlation coefficients (ICCs), using SPSS statistics version 22.0, using a two–way random effects model for absolute agreement.

The results indicated that interrater agreement was excellent, with ICCs (single measures) as follows: speech vocalizations, r = .990, 95% CI [.970, .996]; nonspeech vocalizations, r = .986, 95% CI [.961, .995]; canonical directed vocalizations (CD), r = .995, 95% CI [.985, .998]; canonical nondirected vocalizations (CND), r = .987, 95% CI [.965, .995]; noncanonical directed vocalizations (NCD), r = .958, 95% CI [.820, .987]; noncanonical nondirected vocalizations (NCND), r = .964, 95% CI [.903, .987]; pleasure vocalizations (P), r = .942, 95% CI [.851, .979]; distress vocalizations (D), r = .992, 95% CI [.979, .997]; and atypical vocalizations (A), r = .907, 95% CI [.764, .965].

Our initially planned metric for volubility was the rate of speech–like vocalizations during the full 30 min (i.e., speech–like vocalizations per minute). During the coding process, however, coders noted salient differences across the two contexts, with generally lower volubility during the parent–infant interaction sessions. This led to a post hoc decision to explore volubility separately in each context (parent–infant interaction and CSBS–DP).

III. Results

We used SPSS statistics version 22.0 to carry out the analyses. First, we compared demographic variables for the three groups and documented no significant differences on chronological age, $p = .71$, gender, $p = .97$, or race, $p = .98$ (see Table 3.2). In addition, there were no significant differences in MSEL VR T–scores, $p = .15$, or CSBS–DP total SS, $p = .08$, at 14 months. At 23 months, the comparison of MSEL VR T–scores remained nonsignificant, $p = .35$, but the model for the CSBS–DP total SS indicated significant differences, $p = .009$. Post hoc tests indicated that infants in the Autism group had significantly lower scores than both the Spectrum group and the Non–ASD group, whereas the latter two groups were not statistically different from one another.

Table 3.2. Participant demographics and developmental test performance

ADOS classification	AUTISM	SPECTRUM	NON–ASD
N	34	25	23
Mean age (*sd*) at 13–15 m	13.79 (.77)	13.64 (.76)	13.78 (.74)
Mean age (*sd*) at 20–25 m	22.79 (.91)	22.48 (.65)	22.35 (1.03)
Child's gender (M:F)			
Male	23	17	15
Female	11	8	8
Child's race[a]			
White	24	18	16
Afrian-American	7	4	5
Other	3	3	2
Mean (*sd*) MSEL VR T–score, 13–15 m	43.00 (12.49)	48.36 (9.89)	40.06 (8.71)
Mean (*sd*) MSEL RL T–score, 13–15 m	33.21 (12.34)	34.88 (12.58)	31.00 (6.16)
Mean (*sd*) MSEL EL T–score, 13–15 m	34.26 (11.71)	35.64 (12.06)	32.91 (9.53)
Mean (*sd*) CSBS–DP Total SS, 13–15 m	84.29 (11.39)	86.29 (13.45)	89.87 (15.31)
Mean (*sd*) MSEL VR T–score 20–25 m	43.88 (15.13)	48.92 (10.28)	46.26 (12.49)
Mean (*sd*) MSEL RL T–score, 20–25 m	38.26 (17.99)	48.16 (14.29)	48.65 (13.86)
Mean (*sd*) MSEL RL T–score, 20–25 m	38.85 (14.63)	45.32 (12.10)	40.17 (8.95)
Mean (*sd*) CSBS–DP Total SS, 20–25 m	83.64 (16.53)	96.12 (16.24)	92.87 (13.29)

Note: ASD = autism spectrum disorder; m = months; MSEL = Mullen Scales of Early Learning T–score = standard score with a mean of 50 and *sd* of 10; VR = Visual Reception; RL = Receptive Language; EL = Receptive Language; CSBS–DP = Communication and Symbolic Behavior Scales–Developmental Profile; SS = standard score with a mean of 100, and *sd* of 15.

For descriptive purposes, Figure 3.2 shows the mean frequency data for each vocalization type by ASD symptom group. The means for total vocalizations per group were very similar: Autism = 115.3 (55.2), Spectrum = 107.0 (56.5), non–ASD = 121.5 (46.8), F = .45, p = .64. The standard deviations reflect the large variability within each group.

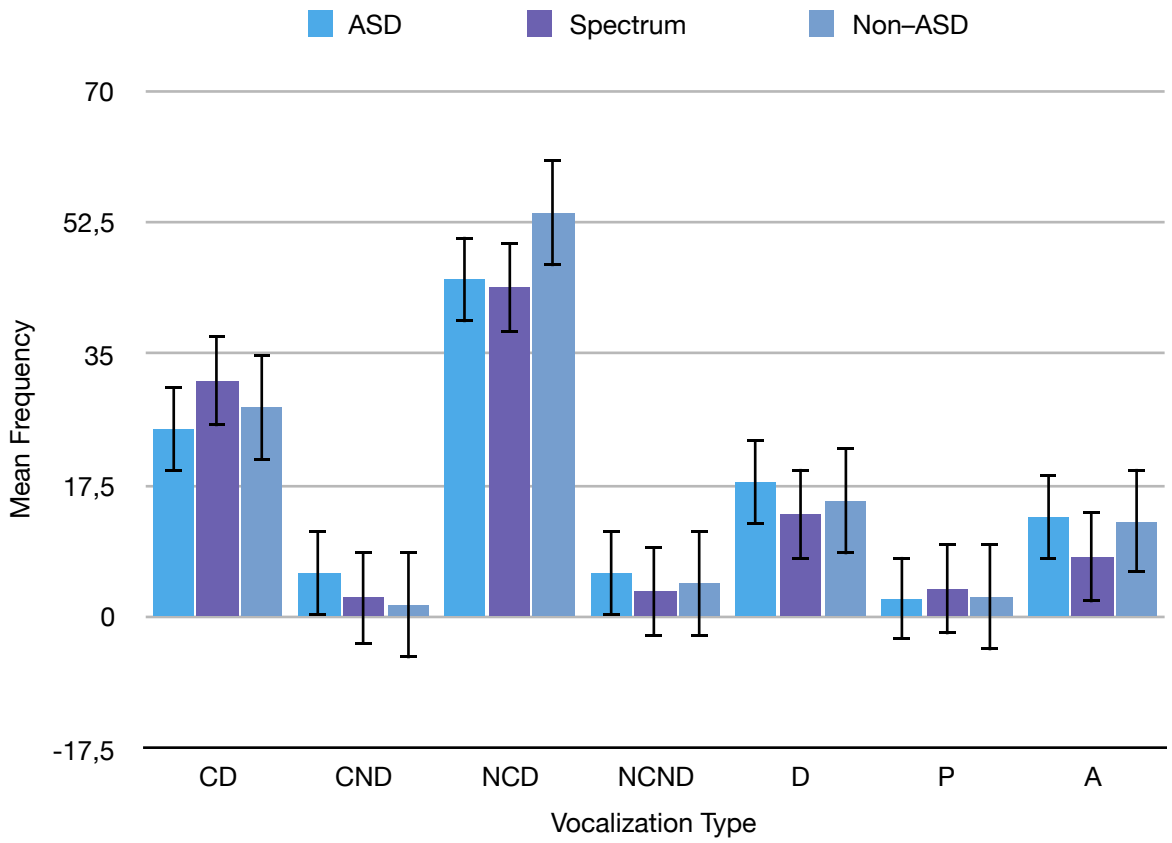

Figure 3.2. Mean frequency of each type of vocalization for infants in ADOS algorithm groups (Autism, Spectrum, and Non–ASD) during a 30–min sample.

Descriptive statistics for the mean frequency and range of each vocalization type, by group are provided in Table 3.3. The number of vocalizations was used in the regression analyses to examine the extent to which group membership could be predicted by vocalization type. Prior to the regression analyses, we conducted descriptive statistics in order to find statistical outliers, and detected three outliers who produced very high numbers of CD vocalizations, exceeding the mean in all groups by more than 3 SDs. Coincidentally, one outlier was detected in each group. In order to minimize the potential impact of bias, we winsorized the data. Comparing results from the winsorized data to results without any transformation, we determined that the outcomes were relatively unaffected by irksome data. Thus, the final analyses included untransformed data from all participants.

Table 3.3. Descriptive statistics for vocalization types and volubility

		Group								
		AUTISM			SPECTRUM			NON-ASD		
		M	SEM	Range	M	SEM	Range	M	SEM	Range
Speech	CD	24.94	3.19	0-65	31.32	5.42	2-107	27.74	5.11	0-97
	CND	5.76	1.23	0-38	2.56	1.17	0-29	1.57	.387	0-6
	NCD	44.97	4.93	6-124	43.80	6.15	9-99	53.87	5.64	4-116
	NCND	5.85	.993	0-21	3.44	1.00	0-20	4.57	.873	0-13
	Total Directed	69.91	7.01	12-161	75.12	.9.99	12-188	81.61	7.77	7-155
	Total Non-directed	11.62	2.04	0-57	6.00	2.06	0-49	6.13	1.04	0-16
Non-speech	A	13.24	1.96	0-50	8.04	1.04	0-19	12.78	2.04	1-36
	D	18.03	3.01	0-59	13.60	2.35	0-38	15.39	3.38	2-76
	P	2.50	.516	0-10	3.80	1.01	0-17	2.57	.482	0-8
Total	Speech	81.53	7.83	15-194	81.48	10.99	13-237	87.74	8.01	7-167
Total	Non-speech	33.76	3.86	0-98	25.52	2.63	9-55	30.74	3.71	8-83
Volubility	10MIN	1.92	1.28	0-6.4	2.73	1.76	.3-6.2	3.03	1.85	.6-7.0
	20MIN	3.12	2.23	0-8.40	2.50	2.03	0-11.7	2.94	1.92	0-5.8
	30MIN	2.72	1.52	.5-6.5	2.50	1.51	.5-7.9	2.92	1.28	.2-5.6

Note: Vocalization types are represented as number of vocalizations. Volubility is represented as the rate of speech–like vocalizations per minute.

CD, canonical direct; CND, canonical nondirected; NCD, non canonical directed; NCND, non canonical nondirected; A, atypical; D, distresss; P, pleasure;

10MIN, parent–infant context; 20MIN, CSBS–DP context; 30MIN, parent–infant + CSBS–DP contexts

We addressed the components of our research question using multinomial logistic regression analysis (an extension of regression that allowed us to predict from our vocalization variables to our categorical outcome— Autism, Spectrum, and Non–ASD). We tested models for speech and nonspeech vocalizations separately, followed by a set of models examining whether volubility discriminated between the groups. The Autism group was used as the referent group in each model. Results are presented in Table 3.4. Odds ratios (ORs) from these models reflect the increase (or decrease) in the odds of a child being in the contrast group relative to the Autism group, based on a one–unit increase in the predictor variable—in this case, an increase of one vocalization. Thus, although these ORs may seem small as presented, they should be considered with respect to the range of vocalizations of each type. To facilitate interpretation of the ORs for each predictor variable, we include the range of vocalizations of each type seen in the full sample.

Table 3.4. Multinomial logistic regression, based on autism group as reference

			AUTISM vs. SPECTRUM			AUTISM vs. NON-ASD		
	Range		OR	95% CI	*P*	OR	95% CI	*P*
Speech model	0 – 107	CD	1.033	.998 –1.068	.062	1.039	1.003 –1.077	.036[a]
	0 – 38	CND	.830	.674 –1.022	.079	.607	.445 –.827	.002[a]
	4 – 124	NCD	.994	.972 –1.017	.624	1.008	.985 –1.031	.501
	0 – 21	NCND	1.028	.866 –1.220	.753	1.200	1.001 –1.439	.049[a]
Non-Speech model	0 – 50	A	.915	.848 –.989	.024[a]	.995	.943 –1.050	.857
	0 – 76	D	.985	.950 –1.021	.414	.989	.956 –1.024	.545
	0 – 17	P	1.141	.972 –1.340	.107	1.008	.845 –1.203	.928
Volubility	.00 – 7.70	10MIN	1.122	.772 –1.630	.546	1.570	1.090 –2.261	.015[a]
	.00 – 11.70	20MIN	.988	.780 –1.251	.919	.981	.769 –1.251	.876
	.23 – 7.90	30MIN	1.016	.718 –1.436	.930	1.141	.806 –1.614	.456

Note:Range: the range of vocalizations of each type seen in the full sample.

CD, canonical direct; CND, canonical nondirected; NCD, non canonical directed; NCND, non canonical nondirected; A, atypical; D, distresss; P, pleasure; 10MIN, parent–infant context; 20MIN, CSBS–DP context; 30MIN, parent–infant + CSBS–DP contexts; OR, Odds ratio

*a, $p < .05$

The first multinomial logistic regression model tested whether there were group differences in different types of speech vocalizations (CD, CND, NCD, NCND). Results revealed there were significant differences between infants in the Autism vs. Non–ASD groups on CD, CND and NCND (CD = OR: 1.039, p = .036; CND = OR: .607, p = .002, and NCND = OR: 1.200, p = .049). This model did not find any significant difference between infants in the Autism versus Spectrum groups. We will use the finding for the CD vocalizations in the comparison of the Autism group to the Non–ASD group to illustrate the interpretation of the ORs in these analyses. The OR of 1.039 indicates the increased odds of being in the Non–ASD group rather than the Autism group associated with producing one additional CD vocalization in the 30–min sample. Producing 20 additional CD vocalizations would increase the odds of being in the Non–ASD group rather than the Autism group to 2.149. Given the range of CD vocalizations from 0 to 107, infants at the extremes of this range of CD vocalizations clearly have much different likelihoods of ending up in the Autism versus Non–ASD group. The second model revealed a significant difference on A (OR: .915, p = .024) between infants in the Autism versus Spectrum groups, indicating each additional A vocalization is associated with a decrease in the likelihood of being in the Spectrum group rather than the Autism group. This model did not find any significant difference between infants in the Autism versus Non–ASD groups.

The Nagelkerke coefficient exhibited evidence that our model including the four subtypes of speech–like vocalizations explained 28% of the variance among groups, and demonstrated significant differences on chi–squared distribution χ^2= 23.55, p = .003. However, the model showed a weak performance in classifying children accurately into the correct ASD symptom group with only 48.8% of the children correctly classified overall. As shown in Table 3.5, although 76.5% of children were correctly predicted to be in the Autism group, the model over–assigned Non–ASD children to the Autism group and over–assigned Spectrum children to the Non–ASD group.

Table 3.5. Classification table for vocalization model

		Predicted		
Observed	Autism	Spectrum	Non–ASD	Percentage correct
Autism	26	4	4	77 %
Spectrum	12	4	9	16 %
Non–ASD	10	3	10	44 %
Overall Percentage	59 %	13 %	28 %	49 %

In contrast to the results reported in speech vocalizations, we did not observe any difference between Autism versus Non–ASD groups on any of the nonspeech vocalization types of distress, pleasure or atypical vocalizations (see Table 3.4). This model found a significant difference on A (OR: .915, p = .024) between infants in the Autism versus Spectrum groups.

Finally, we evaluated the volubility of vocalizations, or the mean rate of speech–like vocalizations per minute (see the lower portion of Table 3.5). A series of three models indicated that volubility across the entire 30–min video samples did not make a significant contribution to predicting group membership, nor did volubility during the 20 min coded from the CSBS–DP. However, during the 10–min parent–infant interaction sessions, the model reported statistically significant differences between the Autism versus Non–ASD groups (OR: 1.570, p = .015). Infants in the Autism group showed lower volubility than infants in either of the other groups. The Nagelkerke coefficient exhibited evidence that the 10–min model explained 9% of the variance among groups, and demonstrated significant differences on chi–squared distribution $\chi^2 = 56.99$, p = .03.

IV. Discussion

In this study, we examined the extent to which characteristics of vocalizations at 14 months served to discriminate between infants falling into one of three ASD symptom groups at 23 months of age. The symptom groups, Autism, Spectrum, and Non–ASD are based on ADOS Module 1 algorithm score criteria. It is important to remember that these groups are based

only on the algorithm scores and do not reflect clinical diagnoses for the infants. Also impacting the interpretation of our study results is the fact that all of the infants in this study were identified as being at–risk for later diagnosis of ASD with the FYIv2.0. Based on estimates from a prior study (Turner–Brown, Baranek, Reznick, Watson, & Crais, 2013) approximately one–third of 12–month–old infants who meet the FYIv2.0 criteria for being considered at–risk for ASD will show symptoms warranting an ASD diagnosis at age 3, and approximately 85% of these infants will have developmental concerns of some type (ASD or other) at age 3. Thus, none of the three symptom groups can be considered a "typically developing" or "low risk" comparison group. The nature of this sample is congruent with our study aim. That is, previous research has yielded replicated findings of differences in vocalizations between infants and toddlers with ASD and their peers who are typically developing (Patten et al., 2014; Plumb & Wetherby, 2013). In order to extend the existing literature, our primary aim was to examine the utility of vocalizations for predicting ASD–related outcomes among a group of infants at–risk for ASD.

To first consider canonical vocalization types (directed or nondirected), infants in the Autism group in the current study used fewer CD vocalizations and more CND vocalizations than infants in the Non–ASD group, and showed nonsignificant trends in the same directions when compared to the Spectrum group. Then turning to noncanonical vocalizations, the tendency of infants in the Autism group to use more nondirected vocalizations was seen again in their higher use of NCND vocalizations than the Non–ASD group. Thus, the differences seen among the groups in their use of speech–like vocalizations seem to be related more to the communicative use of vocalizations (i.e., directing them to other people or not) than to canonical versus noncanonical syllable shapes. This is consistent with the findings reported by Ozonoff et al. (2010) and Winder et al. (2013), as well as being congruent with the later deficits in social–communication that are part of the diagnostic criteria for ASD.

Our findings run counter to the implications of findings of Patten et al. (2014) that syllable structure early in the second year of life might be a powerful risk marker for autism specifically. While acknowledging methodological differences between Patten et al. (2014) and the current study, infants in the Autism group in our study appeared to be producing canonical vocalizations much more consistently at 14 months than those studied by Patten et

al. at either 9–12 months or 15–18 months, and the syllable structure of speech–like vocalizations per se was not significantly predictive of ASD symptom group at 23 months. Possibly syllable structure is more closely related to global developmental delays and/or delays in general communication development than to autism specifically. The credibility of this explanation could be explored more fully in future prospective studies of infants at–risk for ASD, particularly if children are followed sufficiently long to make definitive clinical diagnoses. Comparing groups that are predefined in ways that create noncontinuous distributions of relevant variables (e.g., ASD versus typical, as in Patten et al., 2014) is likely to over–estimate the extent to which early markers predict ASD–related clinical outcomes.

We observed differences in the use of atypical nonspeech vocalizations among these high–risk infants based on their ASD symptom group at 23 months of age. Although Plumb and Wetherby (2013) documented that infants later diagnosed with ASD used proportionately more nonspeech vocalizations than typically developing children, they did not find a difference on this variable between the ASD group and the group of infants later diagnosed with other DD. Thus, our results support previous findings that infants' use of speech versus nonspeech vocalizations may not be helpful in predicting which infants will later be diagnosed with ASD (or present more symptoms consistent with an ASD diagnosis) among infants who are demonstrating some signs of ASD and developmental risk.

Our findings related to volubility extend the previous literature by suggesting that low volubility within relatively unstructured, more naturalistic contexts (such at the parent–infant interactions sessions in our study or the home videos used by Patten et al., 2014) may indicate risk for a later autism diagnosis, whereas volubility in a context specifically designed to elicit communicative behaviors from infants may not. This finding has a parallel in a study of pretend play in preschoolers, where children with ASD produced significantly fewer spontaneous than scaffolded pretend play acts, whereas children with other developmental disorders and children with typical development did not show these discrepancies (Rutherford, Young, Hepburn, & Rogers, 2007). This finding highlights the importance of eliciting information from parents regarding a child's characteristic behaviors at home as well as observing children under unstructured conditions during clinical assessments related to ASD diagnosis, rather than relying solely on observations during structured assessments.

Despite the emergence of this and other significant differences related to vocalizations among the subgroups of at–risk infants in our cohort, the heterogeneity within the groups and considerable overlap between them is perhaps more striking than the between–group differences. Evidence of the heterogeneity and overlap is supported both by the group means and standard deviations shown in Table 3.3 as well as by the low overall accuracy of our models in classifying children into ASD symptom groups, as illustrated in Table 3.5. Certainly, vocalizations could be salient risk markers for ASD in an infant who shows extreme patterns of limited rate of speech–like vocalizations, few canonical syllables, and few speech–like vocalizations directed to others. Such a risk profile might be quite specific to ASD. In addition, this symptom profile would suggest a critical need for early intervention is to address both the form and function of the infant's emerging speech, even if a definitive diagnosis of ASD is not yet possible. Our data suggest, however, that none of the vocalization variables we examined in this study is likely to be a very sensitive marker, in isolation, for later ASD symptom outcomes among at–risk infants. However, examining the quantity and quality of vocalizations may be clinically useful in predicting later ASD diagnosis when considered along with other behavioral risk markers.

Another possibility is that the dimensions of vocalizations examined in this study may be more useful in predicting later language outcomes than later ASD outcomes among these at–risk infants. As shown by the MSEL Receptive and Expressive Language scores in Table 3.2, the infants in all three groups were 1.5 to 2.0 standard deviations below the mean at 13–15 months. On average, language scores improved by 20–25 months, especially for the Spectrum and Non–ASD groups, and the variability generally increased as well. In a young sample of initially preverbal children diagnosed with ASD, Yoder, Watson, and Lambert (2015) found that consonant diversity in the vocalizations of the children at study entry predicted later expressive language outcomes. Thus, the dimensions of vocalizations we examined in this study may have utility for predicting language outcomes among at–risk infants, or perhaps would help to identify infants who will later have both poorer language outcomes and ASD, but may be insensitive to ASD–related diagnoses among infants with a better trajectory of language development.

IV.I. Limitations

The current study has several limitations, primarily related to our use of data collected for other purposes as a mechanism for looking at vocalizations in infants at–risk for ASD. Interpretation of the consistency of our results with prior research would have been enhanced by the inclusion of a comparison group drawn from among infants whose parents returned the FYIv2.0 for the same parent study, but whose infants scored at low risk for ASD. Second, although the data from the parent study indicated that the intervention tested did not have a main effect on ADOS results for these infants, there may be some unidentified moderators of the effect of the treatment on ASD symptoms that changed the association between vocalizations at 14 months and ASD symptoms at 23 months. Third, our reliability for coding atypical vocalizations was lower than for other vocalization categories, and thus measurement error may have masked between–group differences in atypical vocalizations in this study. Finally, grouping infants based on later definitive diagnostic outcomes may have yielded different findings and implications than our use of groups based on ADOS algorithm scores at 23 months; unfortunately, we did not have access to definitive diagnostic outcomes. Previous studies suggest that toddlers whose ADOS scores place them in the Autism or Non–ASD groups are likely to be given corresponding clinical diagnoses of ASD or non–ASD, respectively; however, for toddlers whose algorithm scores place them in the Spectrum group (or mild–to–moderate range of concerns on the ADOS–Toddler module), clinical diagnosis is more likely to diverge from the ADOS results (Charwarska, Klin, Paul, & Volkmar, 2007; Kim & Lord, 2009). False positives appear to be more likely among toddlers who are young in comparison to true positives, and, like true positives, show relatively low performance on nonverbal cognitive and adaptive measures, whereas false negatives appear to be more likely among toddlers who are young in comparison to true positives and have relatively higher scores on nonverbal cognitive and adaptive measures (Kim & Lord, 2009). As shown by the MSEL VR scores (Table 3.2), our participants had mean nonverbal cognitive scores in the average to low average range across all three ASD symptom groups. Due to both their young age and their relatively good nonverbal cognitive skills, clinical diagnoses, if available, may not have shown a high agreement with the ASD symptom groups.

IV.II. Conclusions and Future Directions

Despite its limitations, our study extends previous research on vocalizations of infants at–risk for ASD by examining these behaviors in a community–identified at–risk cohort shortly after their first birthdays. Previous studies of at–risk infants this age or younger largely have been confined to studies of infant siblings of children with ASD. Given that our entire cohort was at high risk for a later diagnosis of ASD and/or other DD, the significant association of some features of their vocalizations with ASD symptom group outcomes is a notable finding. Given the findings of Paul et al. (2011) that the inventory of consonants in infant siblings of children with ASD distinguished between the infants who went on to be diagnosed with ASD and those who did not, examining the predictive value of the consonant inventory in community screened infants at–risk for ASD would be a worthwhile future direction. In addition, research with this population offers exciting opportunities to understand the roles that vocalizations of infants at–risk for ASD may play in eliciting different input from caregivers of these infants (cf., Warlaumont, Richards, Gilkerson, & Oller, 2014), that could in turn impact opportunities for social engagement and language learning for the infants (cf., Woynaroski et al., 2016).

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PART III

AUTISM SPECTRUM DISORDER AND ITS IMPLICATIONS ON FAMILY QUALITY OF LIFE

CHAPTER 4

Chapter 4
Communicative and social–adaptive profile in children with ASD (level of support 1)

The content of this chapter has been published as Garrido, D., García–Fernández, M., Garcia-Retamero, R., & Carballo, G. (2017). Perfil comunicativo y de adaptación social en población infantil con trastorno del espectro autista: Nuevo enfoque a partir de los criterios del DSM–5. *Revista Neurología*, *65*, 49–56, doi: 10.33588/rn.6502.2017019

Perfil comunicativo y de adaptación social en población infantil con trastorno del espectro autista: Nuevo enfoque a partir de los criterios del DSM–5

Tras la nueva clasificación diagnóstica del Manual diagnóstico y estadístico de los trastornos mentales, quinta edición (DSM–5), el trastorno del espectro autista (TEA) ha pasado a considerarse una categoría dimensional que engloba una serie de trastornos que antes se consideraban como entidades diferentes. La bibliografía previa ha mostrado perfiles comunicativos y lingüísticos diferentes en personas con estos trastornos, por lo que podrían encontrarse resultados contradictorios en los individuos que en la actualidad reciben un diagnóstico de TEA. El objetivo es el de identificar los aspectos del lenguaje estructural (expresión y comprensión), interacción (pragmática) y adaptación social diferenciales en niños con diagnóstico de TEA de nivel 1 de apoyo y compararlos con niños con desarrollo típico. Diecisiete niños con síndrome de Asperger (según el DSM–IV–TR) y 20 niños con desarrollo típico de entre 7 y 12 años. Se ha equiparado el síndrome de Asperger del DSM–IV TR con el TEA con nivel 1 de apoyo del DSM–5. Se ha evaluado la inteligencia, la comunicación y la adaptación social con medidas directas estandarizadas y medidas indirectas paternas. Los resultados muestran que existen diferencias significativas en comprensión (comprensión de estructuras gramaticales; $p < .05$), interacción (cuestionario de capacidades y dificultades total; $p < .005$) y adaptación social (Children's Communication Checklist–2 total; $p < .005$) entre los grupos. Los participantes con TEA con nivel 1 muestran un nivel de inteligencia normal y buena expresividad estructural (sintaxis y semántica), lo que podría diferenciarlos sustancialmente, dada la gran heterogeneidad del trastorno, de otros niños que en la actualidad también reciben el diagnóstico de TEA. No obstante, nuestros participantes también presentan problemas de comprensión de estructuras gramaticales, interacción pragmática y adaptación social. Estas dificultades podrían relacionarse con posibles problemas emocionales y de socialización.

I. Introducción

El trastorno del espectro autista (TEA) es un trastorno del neurodesarrollo caracterizado por alteraciones en reciprocidad social y comunicación, y por patrones de conducta e intereses

repetitivos o restrictivos (APA, 2013). Dado que el grado de afectación de las dimensiones del espectro es diferente para cada persona, en la actualidad se especifica la gravedad del TEA en función del nivel de apoyo necesario para cada dominio: 1 (se requiere apoyo), 2 (apoyo sustancial) o 3 (apoyo muy sustancial).

Dentro de estos criterios dimensionales se engloban las categorías diagnósticas incluidas en el Manual diagnóstico y estadístico de los trastornos mentales, cuarta edición, texto revisado (DSM–IV–TR; APA, 2000). Aunque existe un acuerdo generalizado por el que las características claves del síndrome de Asperger son similares a las de otras categorías incluidas bajo el paraguas de los TEA, el síndrome de Asperger todavía se utiliza como término diagnóstico, y se considera por algunos autores como una entidad separada (Helles, Guillberg, Guillberg, & Billstedt, 2015; Tsai & Ghaziuddin, 2014). Por este motivo, el TEA presenta una gran heterogeneidad en cuanto a gravedad de síntomas y alteraciones asociadas a diferentes dominios de desarrollo, incluyendo la conducta adaptativa y las habilidades de lenguaje (Bennett et al., 2008). Algunos niños con TEA son no verbales, otros presentan déficits estructurales (comprensivos y expresivos), mientras que otros muestran un nivel lingüístico estructural similar o superior al de sus pares con desarrollo típico (Dodd, Franke, Grzesik, & Stoskopf, 2014). No obstante, todos ellos manifiestan déficits pragmáticos relacionados con las dificultades de interacción social (Ellis Weismer, 2013).

Los niños con TEA con nivel 1 de apoyo se definen como grupo sin problemas de lenguaje ni retraso cognitivo (Ghaziuddin & Mountain–Kimchi, 2004). Sin embargo, cuando se ha intentado identificar su perfil lingüístico estructural (Helland, Biringer, Helland, & Heiman, 2012; Martin–Borreguero, 2005; Saalasti et al., 2008), se ha mostrado que tienen distintas competencias y algunas dificultades que no muestran los niños con desarrollo típico. Por este motivo, se sugiere que el lenguaje y la comunicación en los niños con TEA con nivel 1 pueden ser superficialmente normales (Kaland, Mortensen, & Smith, 2011). Sus habilidades sintácticas y semánticas suelen estar preservadas: entienden y forman oraciones gramaticalmente correctas, hablan con elocuencia y su léxico se describe como similar al del adulto (Martín–Borreguero, 2005; Saalasti et al., 2008; Stothers & Cardy, 2012). Sin embargo, se han observado dificultades a la hora de hacer inferencias, utilizar conceptos abstractos, metáforas y dobles sentidos, y en la capacidad simbólica (Loukusa & Moilanen, 2009).

Los niños con diagnóstico de TEA presentan habilidades pragmáticas generalmente afectadas (Bishop & Baird, 2001; Helland, 2014; Saalasti et al., 2008) que tienen marcada relevancia en el desarrollo socioemocional (Jerome, Fujiki, Brinton, & James, 2002), por lo que se ha investigado su relación con la teoría de la mente (Hart, Fujiki, Brinton, & Hart, 2004). Sin embargo, la pragmática es por definición dependiente del contexto donde se desarrolla. Por ello, los informes paternos se han mostrado más eficaces que la evaluación directa cuando los niños con TEA tienen patrones de comunicación distorsionados o cuando los problemas comunicativos no son observables con pruebas estandarizadas (Bishop & McDonald, 2009).

Otros aspectos alterados en niños con TEA son la ausencia de reciprocidad en el discurso social, las dificultades para inferir las necesidades del interlocutor, para comprender aspectos comunicativos no verbales, el cambio de tema en la conversación y la tendencia a la derivación hacia detalles irrelevantes del discurso (Paul, Orlovski, Marcinko, & Volkmar, 2009).

Los problemas del habla descritos se relacionan con problemas en habilidades sociales y con la conciencia de los factores del entorno en los niños con TEA. El TEA se asocia con un riesgo elevado de deterioro en la adaptación social, incluyendo problemas de conducta, hiperactividad, dificultades con los compañeros y emocionales (Baron–Cohen & Wheelwright, 2003; Gilmour, Hill, Place, & Skuse, 2004; Meyer, Mundy, Van Hecke, & Durocher, 2006; Sturm, Fernell, & Gillberg, 2004). Las habilidades pragmáticas están implicadas en el establecimiento de relaciones interpersonales, dado que los niños con mayores dificultades lingüísticas suelen ser menos preferidos por sus compañeros (Farmer & Oliver, 2005; Fujiki, Brinton, Hart, & Fitzgerald, 1999). Por esto, es necesario saber si existe un patrón consistente de habla, lenguaje y habilidades pragmáticas en las personas con TEA (Sthothers & Cardy, 2012), así como determinar la posible relación entre habilidades comunicativas y problemas emocionales y conductuales, a fin de conocer el tipo de intervención más adecuada.

En este trabajo investigamos el perfil comunicativo y de adaptación social en niños con TEA en el nivel 1, previamente diagnosticados con síndrome de Asperger, y lo comparamos con el perfil en niños con desarrollo tópico. En línea con la investigación revisada arriba (Meyer et al., 2006; Stothers & Cardy, 2012; Sturm et al., 2004), partimos de

las siguientes hipótesis: las puntuaciones en cociente intelectual (H1), las habilidades estructurales en comprensión gramatical (H2) y las habilidades expresivas (H3) serán similares en los niños con TEA con nivel 1 y con desarrollo típico; en cambio, los niños con TEA con nivel 1 mostrarán un mayor deterioro en las habilidades pragmáticas (H4) y un mayor número de síntomas conductuales relacionados con la adaptación social –en especial, mayores dificultades con los compañeros– (H5).

II. Método

II.I. Participantes

La muestra está formada por 37 participantes, 17 niños con TEA nivel 1 y con 20 con desarrollo típico. Ambos grupos están igualados en edad cronológica (media: 9.9 ± 1.68 años y 10 ± 1.48 años, respectivamente). La proporción de filas y niños con TEA nivel 1 en el estudio se asemeja al ratio presente en la población general de personas con TEA (4 a 1) (Wing, 1981). En el estudio también han participado los padres de los niños. En la Tabla 4.1 se observan las características descriptivas de la muestra.

Tabla 4.1. Datos descriptivos de los participantes

	Desarrollo típico (n = 20)	Trastorno del espectro autista (n = 17)
Género de los padres		
Masculino	19	16
Femenino	1	1
Edad de los niños (años)	10 ± 1.48	9.9 ± 1.68
Género de los niños		
Masculino	19	16
Femenino	1	1
Edad de los padres (años)	40.65 ± 6.23	42.70 ± 5.01

Los criterios de inclusión del grupo con TEA fueron: escolares de entre 7 y 12 años, hispanohablantes y con diagnóstico de síndrome de Asperger (según el DSM–IV–TR; APA, 2000), y no mostrar otros trastornos. Se ha equiparado el síndrome de Asperger del DSM–IV–TR (APA, 2000) con el TEA con nivel 1 de apoyo del DSM–5 (APA, 2012). Por lo tanto, consideramos a este grupo de participantes como un subgroup dentro de los TEA con nivel 1 de apoyo. Los participantes del grupo con desarrollo típico cumplieron los mismos requisitos, exceptuando que no podían presentar ningún trastorno.

II.II. Medidas

El estudio fue aprobado y realizado de acuerdo con los requisitos éticos del Comité Ético de Investigación Biomédica Provincial de Granada. Todas las familias participantes firmaron el consentimiento informado antes de la evaluación.

Se han utilizado los siguientes instrumentos:

Pruebas estandarizadas directas

Escala de inteligencia de Wechsler para niños, IV edición (WISC-IV; Wechsler, 2012). Evalúa la capacidad cognitiva global y cuatro dominios específicos: comprensión verbal, razonamiento perceptivo, memoria de trabajo y velocidad de procesamiento.

Comprensión de estructuras gramaticales (CEG; Mendoza, Carballo, Fresneda, & Muñoz, 2005). Evalúa la comprensión de diferentes estructuras gramaticales de complejidad creciente.

Evaluación Clínica de los Fundamentos del Lenguaje (CELF–4; Semel, Wiig, & Secord, 2006). Proporciona una evaluación de las habilidades lingüísticas del niño y los puntos fuertes y débiles.

Pruebas estandarizadas indirectas

Children's Communication Checklist (CCC–2: Bishop, 2003). Cuestionario que mide diferentes aspectos comunicativos: habilidades estructurales del lenguaje, pragmáticas y de comunicación social. Permite extraer dos puntuaciones: compuesto de problemas de

interacción social (SIDC) para identificar a niños con dificultades pragmáticas y sociales, y compuesto general comunicativo como medida general de la competencia comunicativa.

Cuestionario de capacidades y dificultades (SDQ; Goodman, 1997). Evalúa problemas de conducta (síntomas emocionales, hiperactividad, problemas con los compañeros y conducta prosocial).

Pruebas no estandarizadas indirectas

Historia clínica. Cuestionario de elaboración propia sobre diferentes aspectos socioeconómicos y socioeducativos de los padres, y relativos al desarrollo y evolución del niño, para comprobar el nivel de apoyo que necesitaba cada participante.

La evaluación directa de cada participante se realizó entre dos y tres sesiones en función de la disponibilidad, la atención o los problemas conductuales de los niños. Para la evaluación indirecta, se entregaron los cuestionarios a los padres durante la primera sesión de evaluación.

II.III. Análisis de datos

Se han realizado análisis de varianza (ANOVA) para comprobar si los participantes en los grupos muestran diferencias en inteligencia y en las dos grandes áreas en las que se engloban las pruebas utilizadas: lenguaje (comprensión de estructuras gramaticales, lenguaje expresivo), interacción y adaptación social, utilizando el programa estadístico SPSS v.21.0.

III. Resultados

No se encontraron diferencias significativas entre el grupo con TEA nivel 1 (media: 101.82; error estándar de la media: 4.19) y desarrollo típico (media: 101,65; error estándar de la media: 3.28) en cociente intelectual (Tabla 4.2) ni en la comprensión de estructuras gramaticales (puntuación en bloques) (media: 12.29 ± 3.35 y 14.15 ± 2.87, respectivamente). Por el contrario, se han encontrado diferencias significativas en comprensión de estructuras gramaticales (nivel ítem; $p < .05$) (ver Figuras 4.1 y 4.2). Los participantes del grupo con TEA

Tabla 4.2. Puntuaciones y resultados en Inteligencia, Lenguaje e Interacción y adaptación social

Áreas evaluadas		Condición				Análisis		
		Grupo TEA nivel 1		Grupo DT		F	p	η^2 parcial
		M	EEM	M	EEM			
Inteligencia	WISC-IV	101.82	4.19	101.65	3.28	2.031	.163	.00
Lenguaje	CEG: bloque	12.29	.81	14.15	.64	3.296	.078	.08
	CEG: ítem	66.24	1.53	70.70	1.17	5.502	.025*	.14
	CELF-4: Recordando oraciones	10.06	.49	10.55	.41	.588	.448	.17
	CELF-4: Formulando oraciones	10.65	.47	11.70	.29	3.790	.060	.09
	CELF-4: Clases de palabras expresivas	11.64	.88	11.36	.42	.092	.764	.00
	CELF-4: Clases de palabras receptivas	20.55	1.35	10.36	.52	58.84	.001*	.72
	CELF-4: Clases de palabas total	11.36	.90	10.86	.50	.267	.610	.01
Interacción y adaptación social	CCC-2:Sintaxis	6.47	2.88	2.40	.98	2.022	.164	.05
	CCC-2: Semántica	10.47	3.28	3.60	1.77	3.670	.064	.09
	CCC-2: Coherencia	31.53	8.19	2.90	1.62	12.68	.001*	.28
	CCC-2: Iniciación inadecuada	83.59	5.47	4.75	2.07	204.4	.001*	.85
	CCC-2: Lenguaje estereotipado	42.94	7.34	.10	.10	40.21	.001*	.53
	CCC-2: Contexto	66.00	7.74	2.65	1.76	73.79	.001*	.68
	CCC-2:Comunicación no verbal	61.12	7.82	3.70	2.09	57.90	.001*	.62
	CCC-2: Relaciones sociales	46.35	7.86	.90	.41	39.38	.001*	.52
	CCC-2: Interés	91.76	3.85	8.50	3.48	257.36	.001*	.88
	SIDC	61.12	10.25	22.10	21.90	20.67	.001*	.37
	GCC	33.29	17.45	6.95	38.77	14.64	.001*	.29
	SDQ: Puntuación total	19.88	1.27	8.80	1.16	41.19	.001*	.54
	SDQ: Síntomas emocionales	5.24	.53	2.10	.32	26.59	.001*	.43
	SDQ: Problemas de conducta	2.29	.39	1.45	.31	2.920	.096	.07
	SDQ: Hiperactividad	6.71	.51	3.85	.60	12.48	.001*	.26
	SDQ: Problemas compañeros	5.65	.56	1.40	.35	43.07	.001*	.55
	SDQ: Conducta prosocial	5.65	.63	8.85	.31	22.53	.001*	.39

Nota: M=media; EEM=Error estándar de la media. Interpretación de η^2 parcial: ~.01 efecto menor; ~.06 efecto medio; ~.14 efecto grande (Cohen, 1988). *$p < .05$.

con nivel 1 también obtienen un percentil y puntuaciones similares a las del grupo con desarrollo típico en el nivel expresivo (Tabla 4.2), y se han encontrado diferencias significativas entre los grupos sólo en la subprueba de clases de palabras ($p < .005$).

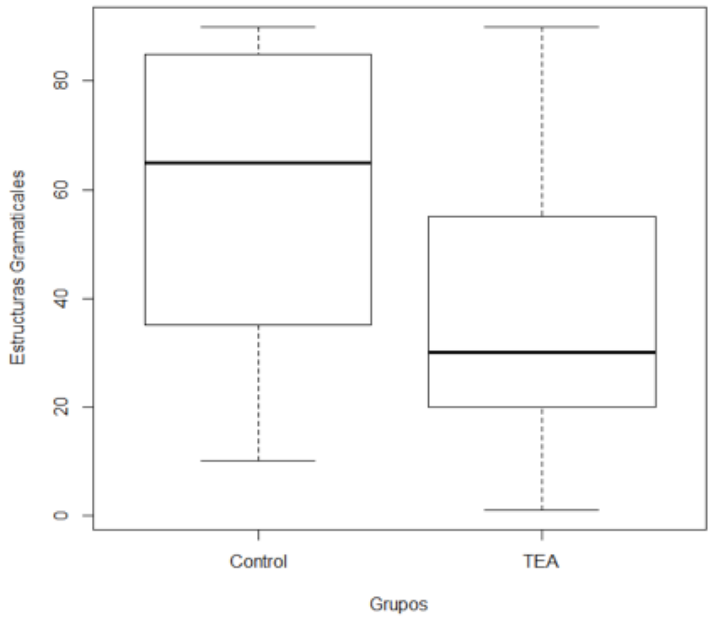

Figura 4.1. Percentil en la comprensión de estructuras gramaticales (CEG, nivel ítem) en los grupos de trastorno del espectro autista (TEA) y desarrollo típico (control).

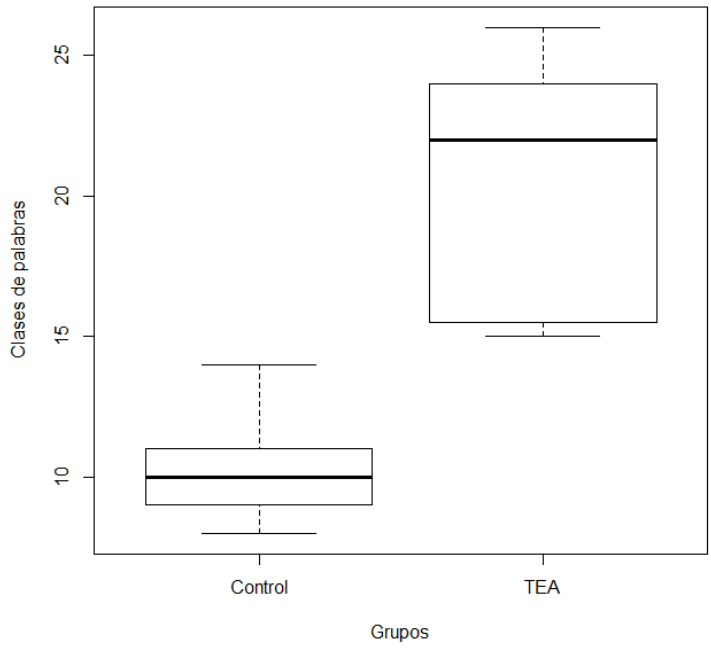

Figura 4.2. Número de errores en CELF–4 (clases de palabras) en los grupos de trastorno del espectro autista (TEA) y desarrollo típico (control)

En cuanto a la interacción, las puntuaciones en el CCC–2 muestran diferencias significativas en las variables de coherencia ($p < .005$), iniciación inadecuada ($p < .005$), lenguaje estereotipado ($p < .005$), contexto ($p < .005$), comunicación no verbal ($p < .005$), relaciones sociales ($p < .005$) e interés ($p < .005$). Sin embargo, no existen diferencias significativas entre los grupos en las variables de sintaxis (TEA con nivel 1, media: 6.47±11.89; DT, media: 2.4±4.41) y semántica (TEA con nivel 1, media: 10.47±13.56; DT, media: 3.6±7.93), ambas referidas a aspectos estructurales del lenguaje. También se han encontrado diferencias significativas entre los grupos en las puntuaciones compuestas del SIDC ($p < .005$) y compuesto general comunicativo ($p < .005$) (ver figuras 4.3 y 4.4). Los resultados obtenidos en adaptación social muestran diferencias significativas entre los grupos en el SDQ en la puntuación total ($p < .005$) (ver Figura 4.5) y en las variables de síntomas emocionales ($p < .005$), hiperactividad ($p < .005$), problemas con los compañeros ($p < .001$) y conducta prosocial ($p < .001$). Sin embargo, no existen diferencias significativas entre el grupo con TEA con nivel 1 (media: 2.29±1.61) y el grupo con DT (media: 1.45±1.39) en problemas de conducta.

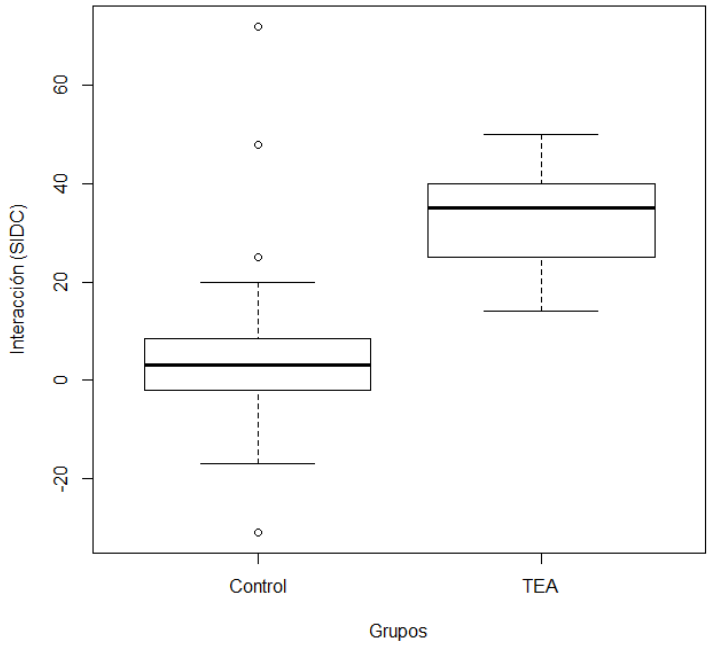

Figura 4.3. Puntuación en el compuesto de problemas de interacción social (SIDC) del CCC–2 en los grupos de trastorno del espectro autista (TEA) y desarrollo típico (control).

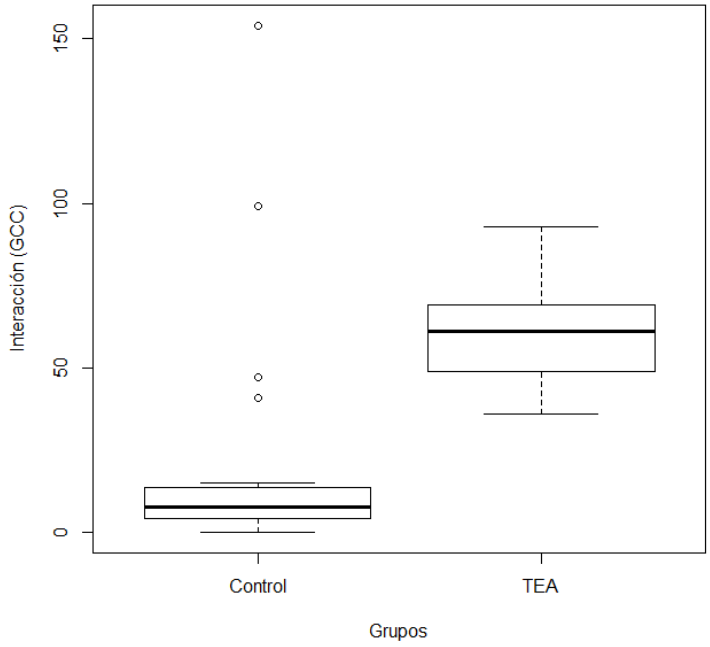

Figura 4.4. Puntuación en el compuesto general comunicativo (GCC) en los grupos de trastorno del espectro autista (TEA) y desarrollo típico (control).

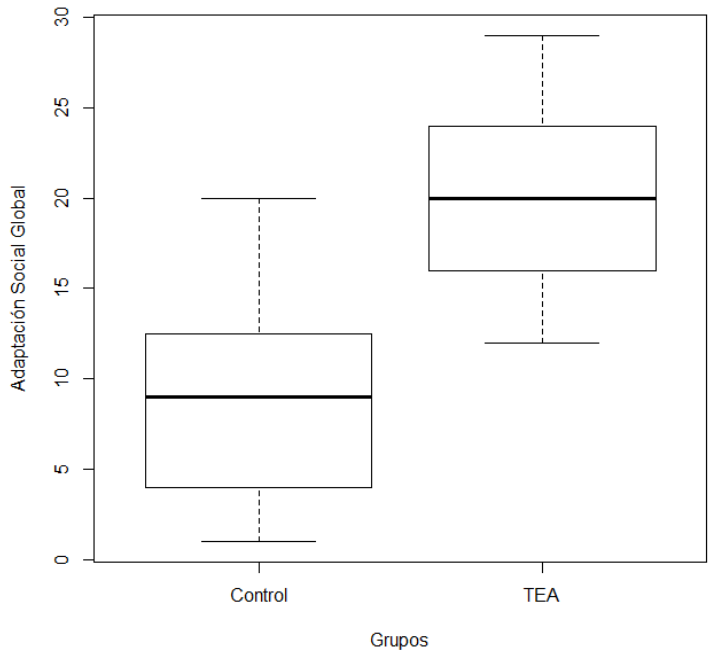

Figura 4.5. Puntuación total en adaptación social global (cuestionario de capacidades y dificultades) del SDQ en los grupos de trastorno del espectro autista (TEA) y desarrollo típico (control).

IV. Discusión

La finalidad de nuestro estudio ha sido identificar los aspectos diferenciales del lenguaje estructural (expresión y comprensión), interacción (pragmática) y adaptación social en niños con diagnóstico de Tea de nivel 1 de apoyo según el DSM–5 (APA, 2013) y compararlos con niños con desarrollo típico. Para ello, hemos utilizado instrumentos de evaluación directa e indirecta.

Rubin, Bukowski, Parker, and Bowker (1998) han puesto de manifiesto que las relaciones entre iguales son un contexto fundamental para desarrollar las habilidades sociales. Además, los déficits comunicativos influyen en dichas relaciones de manera fundamental (Leonard, Milich, & Lorch, 2011). Si la pragmática, como parte de la comunicación, tiene en cuenta aspectos sociales, emocionales y comunicativos (Adams, Baxendale, Llyod, & Aldred, 2005; Crespo–Eguilaz, Magallón, Sanchez-Carpintero, & Narbona, 2016), la evaluación del perfil lingüístico y pragmático de los niños con TEA con

nivel 1 en distintos niveles puede ayudarnos a comprender las dificultades que experimentan.

De manera general, los resultados de evaluación directa sugieren similitudes en los niños con TEA de nivel 1 y los de desarrollo típico en inteligencia, y comprensión de bloques de estructuras gramaticales y nivel expresivo. Estos datos apoyan nuestras hipótesis de partida (H1, H2 y H3) y se ven corroborados por la información de los padres, ya que no aparecen diferencias estructurales evaluadas a través del CCC–2 (sintáctico y semántico). El perfil cognitivo en ambos grupos es similar también, con un funcionamiento dentro del rango normal (Ghaziuddin et al., 2004; Kaland et al., 2011). Sin embargo, a pesar de que los niños con síndrome de Asperger se consideran gramaticalmente fluidos (Paynter & Peterson, 2010), hemos encontrado diferencias significativas entre los grupos de participantes en varias medidas. Concretamente, ambos grupos difieren en comprensión de estructuras gramaticales, dato coherente con los de otros estudios que indican existencia de dificultades gramaticales "sutiles" en la interpretación de pronombres reflexivos (Perovic, Modyanova, & Wexler, 2013) e independientemente del cociente intelectual de los niños (Eigsti, Bennetto, & Dadlani, 2007).

En los TEA, a menudo las dificultades de comprensión lingüística pueden ser difíciles de detectar cuando el lenguaje expresivo está aparentemente funcionando con normalidad, pero nuestros resultados muestran diferencias receptivas en la asociación de palabras semánticamente relacionadas. Estos problemas pueden evidenciarse a la hora de comprender frases, el discurso global o las preguntas que hacen referencias a conceptos abstractos (Leekam, 2007). Por tanto, nuestra hipótesis planteada sobre las similitudes en habilidades estructurales del lenguaje (H2) se ve apoyada sólo parcialmente debido a estas diferencias en comprensión gramatical.

En cuanto a la comunicación (H4), los resultados mostraron diferencias significativas entre los niños con TEA con nivel 1 y los de desarrollo típico en todas las variables excepto en sintaxis y semántica. Estos resultados apoyan nuestras hipótesis de partida y son similares a los encontrados por Helland (Helland, 2014). La competencia comunicativa de los niños con TEA con nivel 1 también se vio afectada, lo que puede deberse a que las habilidades pragmáticas están gravemente alteradas. Las diferencias más elevadas aparecen en interés, iniciación inadecuada y contexto, lo que coincide con los resultados encontrados por Geurts

et al. (2004), que muestran que una de las mayores diferencias entre los niños con TEA con nivel 1 y los de desarrollo típico se encuentran en la escala de intereses y en el contexto. En cambio, en el estudio de Geurts y Embrechts (2008) no ocurre así con las subescala de iniciación inadecuada.

Respecto a la adaptación social (H5), encontramos diferencias enter los niños con TEA con nivel 1 y los de desarrollo típico en todas las escalas: síntomas emocionales, hiperactividad, problemas con los compañeros y conducta prosocial. Aunque Russell, Rodgers, & Ford (2013) también encontramos puntuaciones más elevadas en la escala de problemas de conducta y de hiperactividad, en nuestro estudio los problemas de conducta no aparecen como una dificultad significativa, lo que puede indicar la inhibición social típica de estos niños con su entorno (Volden, & Phillips, 2010).

Los padres de niños con TEA nivel 1 informaron de que sus hijos tienen más dificultades pragmáticas y sociales que estructurales en el lenguaje, lo que apoya los resultados de Geurts y Embrechts (Geurts, & Embrechts, 2008). El uso inadecuado del lenguaje en situaciones sociales está presente frecuentemente en los TEA (Geurts, et al., 2004; Geurts & Embrechts, 2008; Volden & Phillips, 2010). De aquí se deduce la importancia de evaluar las dificultades de interacción para establecer si se producen modificaciones en el tiempo con el fin de ajustar la terapia a las necesidades de los niños (Geurts, & Embrechts, 2008).

En suma, un análisis global de nuestros resultados muestra que los niños con TEA de nivel 1 presentan un perfil con un nivel de cociente intelectual en valores normales y una buena expresividad general tanto semántica como sintáctica. No obstante, también presentan problemas de comprensión de estructuras gramaticales (déficits específicos) y pragmáticos que podrían estar relacionados con los problemas emocionales y la socialización. Siguiendo a Barkley (1997), este perfil podría entenderse bajo el hecho del carácter directivo de la planificación, organización y control de la conducta. Algunos autores sugieren que puede existir una mediación de la pragmática entre los problemas atenciones y de control inhibitorio y las relaciones sociales en la hiperactividad (Crespo–Eguilaz et al., 2016). Para este autor, los problemas de atención e impulsividad derivarían en conductas inadecuadas que, junto con el estrés de experiencias negativas previas, hacen que en nuevas situaciones se produzcan fallos pragmáticos, como las interrupciones, la iniciación

inadecuada y no mirar a los ojos durante las interacciones sociales. Futuros estudios deberían incluir medidas de control inhibitorio o atencionales.

La aparición del DSM–5 plantea el reto de considerar el TEA dentro de un continuo, lo cual supone englobar en una misma categoría una serie de trastornos considerados previamente como entidades diferentes. Pero, ¿supone esto una mejora respecto a la clasificación categorial previa? Nuestros datos se ven apoyados por otras investigaciones (Gibbs, Aldridge, Chandler, Witzlsperger, & Smith, 2012; Tsai, 2013) sugieren que quizá la perspectiva dimensional sea parcial o incompleta en eta población, debido a su diversidad y a las diferencias que se muestran en otros aspectos. Dado que se ha vuelto a la descripción que hizo Hans Asperger (1938) para describir el TEA en el DSM–5 (Artigas–Pallares, Paula–Perez, 2017), creemos que se deben tener en cuanta ciertos aspectos (por ejemplo, rasgos fenotípicos clínicos, perfiles cognitives y patrones de desarrollo (Lai, Lombardo, Chakrabarti, & Baron-Cohen, 2013) que pueden seguir diferenciando a grupos de personas que se engloban bajo un cierto diagnóstico. Este tipo de consideraciones contribuye a la especialización de los tratamientos e intervenciones que están destinados a mejorar las habilidades de esta población.

Observar las características comunes y diferenciales de las personas con TEA con nivel 1 dentro del espectro, en el contexto lingüístico–comunicativo, de interacción y adaptación social, es fundamental. Nuestro estudio, como otros anteriores (Geurts et al., 2004; Geurts, & Embrechts, 2008), refuerza la importancia de las habilidades pragmáticas en estas personas y de las dificultades lingüísticas añadidas que muestran. Desarrollar estas habilidades será fundamental en los contextos más importantes, familia, iguales y escuela, ya que, dependiendo de la utilización del lenguaje y la comunicación en los diferentes contextos, así será su éxito social y académico en las etapas iniciales de escolarización. Los participantes incluidos en el grupo con TEA con nivel 1 en nuestro estudio tienden a ser integrados en clases ordinarias, donde tienden a ser integrados en clases ordinarias, donde reciben las mismas instrucciones que otros niños con desarrollo típico. Del mismo modo, las dificultades en el lenguaje y en su uso podrían estas afectando a la calidad de las relaciones con sus compañeros. Por lo tanto, cobra importancia la intervención en su lenguaje y en la pragmática.

Deben considerarse algunas limitaciones metodológicas al interpretar los resultados de nuestro estudio. El rango de edad de los participantes es amplio, y sabemos que el desarrollo lingüístico se produce de una manera muy rápida en los niños. La valoración diagnóstica de los participantes con TEA se ha establecido siguiendo los criterios del DSM–IV–TR (APA, 2000). Asimismo, los profesores podrían proporcionar información indirecta adicional que muestre un panorama más amplio sobre los niños con TEA.

En investigaciones futuras sobre población con TEA deberían explicarse la posible relación entre las distintas dificultades lingüísticas y de comunicación y profundizar en las dificultades de comprensión gramatical oral y escrita, lo que podría implicar los resultados encontrados en nuestro trabajo.

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CHAPTER 5

Chapter 5
Emotional and behavioral functioning in children with ASD (level of support 1)

The content of this chapter has been adapted from Garrido, D., Carballo, G., Ortega, E., & Garcia-Retamero, R. (under review). Conducta adaptativa en niños con autismo y su efecto sobre la calidad de vida familiar.

Conducta adaptativa en niños con autismo y su efecto sobre la calidad de vida familiar

En la mayoría de los niños con trastorno del espectro autista (TEA) aparecen dificultades asociadas que, aunque no se incluyen en los criterios diagnósticos, podrían tener impacto sobre la calidad de vida familiar (CdVF). Este estudio examina el papel de la conducta adaptativa y su posible impacto sobre la CdVF en 49 familias de niños con TEA (nivel 1 de apoyo) y niños con desarrollo típico. Los resultados muestran diferencias significativas en las variables relacionadas con la conducta adaptativa, y en algunos de los componentes de la CdVF (interacción familiar, bienestar físico y emocional). En el modelo de regresión, la conducta prosocial ($\beta = .36$, $p < .05$) y el grupo ($\beta = –.36$, $p < .05$) aparecieron como los principales predictores de la satisfacción percibida en la CdVF. Por tanto, es importante atender a las dificultades en la conducta prosocial en TEA, dado su potencial efecto protector sobre la CdVF.

I. Introducción

El Trastorno del Espectro Autista (TEA) es un trastorno del neurodesarrollo que afecta a 1 de cada 59 niños en EEUU según el Centre for Disease Control and Prevention (Baio et al., 2018). Este trastorno se caracteriza por dos dominios: déficits en interacción social y comunicación, y patrones de conducta restringidos y repetitivos (APA, 2013). Sin embargo, Helland y Helland (2017) y Posserud, Hysing, Helland, Gillberg y Lundervold (2018) afirman que muy pocos niños con TEA presentan solo y exclusivamente características diagnósticas. Por ejemplo, otros rasgos distintivos del TEA aparecen de forma generalizada, como las dificultades conductuales, la falta de atención/hiperactividad, los problemas con los compañeros en la edad escolar o las dificultades emocionales (Hervás & Rueda, 2018; Russell, Rodgers, & Ford, 2013). Dado que algunas características centrales de TEA (p. ej., el lenguaje comprensivo) ejercen diversos efectos sobre la interacción y el funcionamiento familiar (Gardiner & Iarocci, 2012; Garrido, Carballo, Franco, & Garcia-Retamero, 2015), se hace imprescindible evaluar de manera exhaustiva el impacto que ejercen otras características no diagnósticas sobre el bienestar familiar.

El funcionamiento familiar y las necesidades de los diferentes miembros de una familia con un hijo con TEA pueden ser evaluados de diferentes maneras. Uno de los conceptos que mejor recoge este concepto es la calidad de vida familiar (CdVF); una medida del bienestar de la familia y que puede verse alterada por múltiples factores (tanto aspectos personales como ambientales) (Eapen & Guan, 2016).

El estudio de la CdVF en TEA se ha centrado especialmente en su caracterización y en su descripción como un componente diferencial respecto a las familias de niños con desarrollo típico (DT) o con otros trastornos del desarrollo, como el trastorno por déficit de atención/hiperactividad, o el síndrome de Down (Cohen, Holloway, Dominguez–Pareto, & Kuppermann, 2014; Vasilopoulou & Nisbet, 2016). En la literatura más reciente, este enfoque ha evolucionado y se ha profundizado en el estudio de diversas variables que pueden explicar la satisfacción de la CdVF en este trastorno (véase Eapen & Guan, 2016 para una revisión). Una de las áreas a la que se le ha prestado atención ha sido la conducta adaptativa (e.j., adaptación social, emocional y conductual), que son particularmente importantes en el TEA, debido a que forman parte de los indicadores críticos del progreso de una persona.

Entre los factores relacionados con la conducta adaptativa que se han evaluado y que ejercen un impacto sobre la CdVF, se encuentran algunos aspectos como: hiperactividad, nivel de retraso general del desarrollo (Baghdadli, Pry, Michelon, & Rattaz, 2014), problemas de oposición y conducta desafiante (McStay, Trembath, & Dissanayake, 2014; Suzumura, 2015), ansiedad y síntomas emocionales (Pozo, Sarria, & Brioso, 2014), y el deterioro en las actividades de la vida diaria (Eapen & Guan, 2016; Gardiner & Iarocci, 2012; Pozo et al. 2014). Sin embargo, los resultados son contradictorios. Por ejemplo, Simonoff et al. (2008) no encontraron factores de los padres relacionados con los problemas emocionales y conductuales de los niños con TEA. Por tanto, no existe precisión sobre si las posibles diferencias en la conducta adaptativa de los niños con TEA podrían influir sobre la CdVF.

Por otro lado, aunque las investigaciones citadas anteriormente se han llevado a cabo con familias de niños con TEA, la descripción de la población resulta incompleta a la luz de la reorganización actual de los criterios diagnósticos según el DSM–5 (APA, 2013), y por tanto los resultados no se pueden generalizar a personas con TEA en función de su nivel de severidad o de apoyo. Son escasos los trabajos en los que se describen algunas de las características de los participantes. Por ejemplo, en algunos estudios han participado niños

con un diagnóstico de TEA mínimamente verbales y/o con nivel de inteligencia en la media (Gardiner & Iarocci, 2012; Garrido et al., 2015). Sin embargo, otras investigaciones han incluido muestras de participantes en la que se incluyen diversos trastornos como el síndrome de Rett, TEA, y el síndrome de Asperger (SA) (McStay et al., 2014; Pozo et al., 2014). Dada la gran heterogeneidad que aparece a lo largo del trastorno, se hacen necesarias investigaciones que describan en profundidad las características diagnósticas de las muestras para poder ofrecerles intervenciones específicas a cada familia en función de las particularidades del TEA.

Por tanto, si bien las escasas investigaciones que relacionan la conducta adaptativa con el funcionamiento familiar indican que puede existir una relación entre ellas, no se ha estudiado en profundidad hasta la fecha, la influencia de la conducta adaptativa sobre la CdVF en TEA en poblaciones definidas por su nivel de apoyo. Por ello, el presente estudio tiene como objetivos (1) describir y evaluar las diferencias en la conducta adaptativa (síntomas emocionales, problemas de conducta, hiperactividad/falta de atención, problemas de relaciones entre compañeros y conducta prosocial) entre niños con TEA (nivel de apoyo 1) y niños con DT, (2) comprobar si existen diferencias en las percepciones sobre el grado de CdVF (a nivel de importancia y satisfacción) entre las familias que tienen un hijo con TEA o un hijo con DT, y (3) determinar la influencia de cada una de las variables referidas al funcionamiento emocional y conductual como posibles predictores en la satisfacción percibida en CdVF.

II. Método

II.I. Participantes

En este estudio han participado un total de 49 padres de niños con TEA y con DT con edades comprendidas entre los 28 y los 55 años (media de edad de 37.2 años). La mayoría de los participantes eran madres (92%). La edad media de los niños era de 9.98 años (rango de edad de 6−13 años). El ratio de niños y niñas con TEA era de 3:1. Este ratio es algo menor del ratio 4.5:1 que se suele informar (Baio et al., 2018).

Los hijos de los participantes del grupo TEA fueron diagnosticados con SA según el DSM–IV–TR (APA, 2000) o con TEA nivel de apoyo 1 según el DSM–5 (APA, 2013). Aquellos participantes que recibieron el diagnóstico a través del DSM–IV–TR (APA, 2000), debían tener un diagnóstico equiparado con TEA nivel 1 de apoyo según el DSM–5 (APA, 2013). Dado que algunos de los participantes estaban diagnosticados con SA, pero dicha categoría se encuentra en la actualidad incluida dentro de los TEA, este último término es el que se ha usado en este trabajo (ver Helland & Helland, 2017 para un procedimiento similar). Además, los padres de los participantes del grupo TEA completaron un cuestionario con información relativa al diagnóstico, edad del mismo, e historia de síntomas de TEA. El nivel socio–económico de la muestra era medio.

Los participantes de ambos grupos debían cumplir con los siguientes criterios de inclusión: (1) tener un/a hijo/a en edad escolar (entre 6 y 13 años), (2) no tener otro hijo/a con otra discapacidad o con algún trastorno del desarrollo, (3) tener como lengua materna el castellano, (4) tener un nivel de inteligencia en la media, y (5) no presentar comorbilidad con otros trastornos en el caso de los niños del grupo TEA. Además, el grupo DT no debía presentar ningún trastorno del desarrollo (ej., TEA, SA, síndrome de Down, X–Frágil, retraso en el desarrollo o problemas de lenguaje), así como tampoco tener una historia familiar previa de TEA. Las características sociodemográficas de los participantes se recogen en la Tabla 5.1.

Tabla 5.1. Variables sociodemográficas de los participantes del estudio

	Participantes					
	Grupo TEA (n = 24)			Grupo DT (n = 25)		
	M	SD	Rango	M	SD	Rango
Niños con TEA						
Edad	10.18	1.70	7.33–13.00	9.78	1.76	6.00–12.75
Género						
Masculino	19	–	–	17	–	–
Femenino	6	–	–	8	–	–
Padres						
Edad	39.17	7.69	29–54	35.32	5.79	28–55
Género						
Masculino	1	–	–	3	–	–
Femenino	23	–	–	22	–	–
Nivel educativo						
Sin estudios	0	–	–	0	–	–
Ed. primaria	2	–	–	1	–	–
Ed. secundaria	9	–	–	10	–	–
Formación profesional	3	–	–	5	–	–
Nivel universitario	10	–	–	9	–	–
Estado civil						
Soltero	0	–	–	0	–	–
Casado	19	–	–	21	–	–
Divorciado/Separado	5	–	–	4	–	–
Viudo	0	–	–	0	–	–
Situación laboral						
Desempleo	11	–	–	11	–	–
Tiempo parcial	9	–	–	7	–	–
Tiempo completo	4	–	–	7	–	–

II.II. Medidas

Se han realizado dos tipos de evaluaciones: directa e indirecta a través de los padres. En ambos casos, las pruebas que se han utilizado han sido estandarizadas.

Evaluación directa de los niños

T*est de vocabulario en imágenes Peabody* (PPVT–III; Dunn, Dunn, & Arribas, 2006). Tiene como finalidad evaluar el nivel de vocabulario comprensivo y sirve como screening de la aptitud verbal para personas entre 2.6 años y 90 años, con una duración, entre 10 y 20 minutos.

Escala de inteligencia de Wechsler para niños, IV edición (WISC–IV; Wechsler, 2012). Esta escala evalúa la capacidad cognitiva general y cuatro capacidades específicas: comprensión verbal, razonamiento perceptivo, memoria de trabajo, y velocidad de procesamiento.

Evaluación indirecta a los padres

Escala de calidad de vida familiar (CdVF; Verdugo, Rodriquez, & Sainz, 2009). Esta escala permite conocer el grado de satisfacción e importancia de la familia frente a cada uno de los cinco factores de la CdVF: interacción familiar, rol parental, bienestar emocional, bienestar físico y material, y recursos y apoyos. Esta escala ha mostrado una buena consistencia interna en nuestra muestra, con un Alfa de Cronbach de 0.82.

El Strengths and Difficulties Questionnaire (SDQ; Goodman, 1997). Este cuestionario consta de 25 ítems de detección de problemas conductuales desde los 3 a los 16 años de edad. Incluye cinco subescalas: síntomas emocionales, problemas de conducta, hiperactividad/falta de atención, problemas de relaciones entre compañeros y comportamiento prosocial. El SDQ ha demostrado ser una herramienta fiable en niños con TEA (Russell et al., 2013).

II.III. Procedimiento

El estudio fue aprobado por el comité ético de investigación biomédica de la Universidad de Granada (España). Todas las familias firmaron el consentimiento informado antes de la

evaluación. La evaluación directa de cada participante se realizó en una sesión de una hora y quince minutos aproximadamente, dependiendo del nivel de ejecución de cada uno de ellos. La evaluación indirecta se llevó a cabo a través de la entrega de los cuestionarios a los padres.

II.IV. Análisis de los datos

Se han realizado análisis de varianza (ANOVAs) con el paquete estadístico SPSS V.22.0 para comprobar si los participantes en los grupos TEA y DT muestran diferencias en las cuatro grandes áreas evaluadas: vocabulario receptivo, inteligencia, conducta adaptativa, y CdVF. El tamaño del efecto fue calculado a través de Eta cuadrado, considerando .04, .36 y >.36 como tamaño del efecto pequeño, medio y grande respectivamente. Posteriormente se realizaron análisis de covarianza (ANCOVAs) en los que se ha controlado la influencia del género, inteligencia y vocabulario receptivo.

Para determinar la influencia de los potenciales predictores, realizamos regresiones múltiples. Aquellas variables que eran asimétricas fueron transformadas logarítmicamente para los análisis. Además, se realizaron análisis adicionales con las variables control (género y edad) y los resultados se mantenían.

III. Resultados

Los resultados mostraron que no se encuentran diferencias significativas entre el grupo TEA y el grupo DT en términos de vocabulario a nivel receptivo e inteligencia (ver Tabla 5.2). Por el contrario, sí se encontraron diferencias significativas en las CdVF, y en la conducta adaptativa.

Tabla 5.2. Puntuaciones en las diferentes áreas evaluadas en función del grupo y análisis

		Participantes						Análisis		
		Grupo DT			Grupo TEA			*F*	*p*	η^2
		M	SD	EEM	M	SD	EEM			
Vocabulario		45.24	28.84	5.77	44.09	39.52	8.07	.01	.908	.00
Inteligencia		102.48	15.26	3.05	99.17	16.40	3.35	.54	.467	.01 .
CdVF	Importancia familiar (I)	28.40	1.19	.24	27.68	1.56	.33	3.20	.08	06
	Importancia familiar (S)	26.36	2.19	.44	21.41	4.09	.87	27.64*	<.001	.38
	Papel de padres (I)	25.12	5.14	1.03	26.23	2.54	.54	.84	.365	.02
	Papel de padres (S)	22.52	5.83	1.17	21.00	3.93	.84	1.06	.308	.02
	Bienestar emocional (I)	17.36	1.20	.40	15.50	3.22	.69	5.82*	.020	.11
	Bienestar emocional (S)	16.08	1.85	.37	9.50	3.08	.66	81.04*	<.001	.64
	Bienestar físico (I)	24.24	.52	.11	21.64	2.61	.56	23.85*	<.001	.35
	Bienestar físico (S)	23.32	.99	.20	18.32	3.91	.83	38.27*	<.001	.46
	Recursos (I)	16.32	4.54	.91	18.32	1.67	.36	3.79	.058	.08
	Recursos (S)	15.24	4.45	.89	15.14	1.75	.37	.01	.919	.00
SDQ	Síntomas emocionales	1.72	1.43	.29	5.04	2.76	.56	28.35*	<.001	.38
	Problemas de conducta	1.20	1.12	.22	2.46	1.64	.34	9.91*	.003	.17
	Hiperactividad	2.96	2.56	.51	5.96	2.84	.58	15.13*	<.001	.24
	Problemas compañeros	1.24	1.51	.30	5.50	2.81	.57	44.15*	<.001	.48
	Conducta prosocial	8.84	1.38	.28	5.50	2.69	.55	30.38*	<.001	.39

Nota: M = media; EEM = Error estándar de la media; (I) = Importancia; (S) = Satisfacción.
* = $p < .05$; Interpretación de η^2 = <.04 efecto pequeño; >.04 y <.36 efecto medio; >.36 efecto grande.

Calidad de vida familiar

Los resultados obtenidos en la CdVF mostraron diferencias significativas en satisfacción (ver Figura 5.1) en interacción familiar, bienestar emocional, y bienestar físico (todas con $p < .001$ y con un tamaño del efecto grande, $\eta^2 > .36$). Sin embargo, no se encontraron diferencias en satisfacción en papel de padres ($p = .31$) ni en recursos y apoyos ($p = .92$).

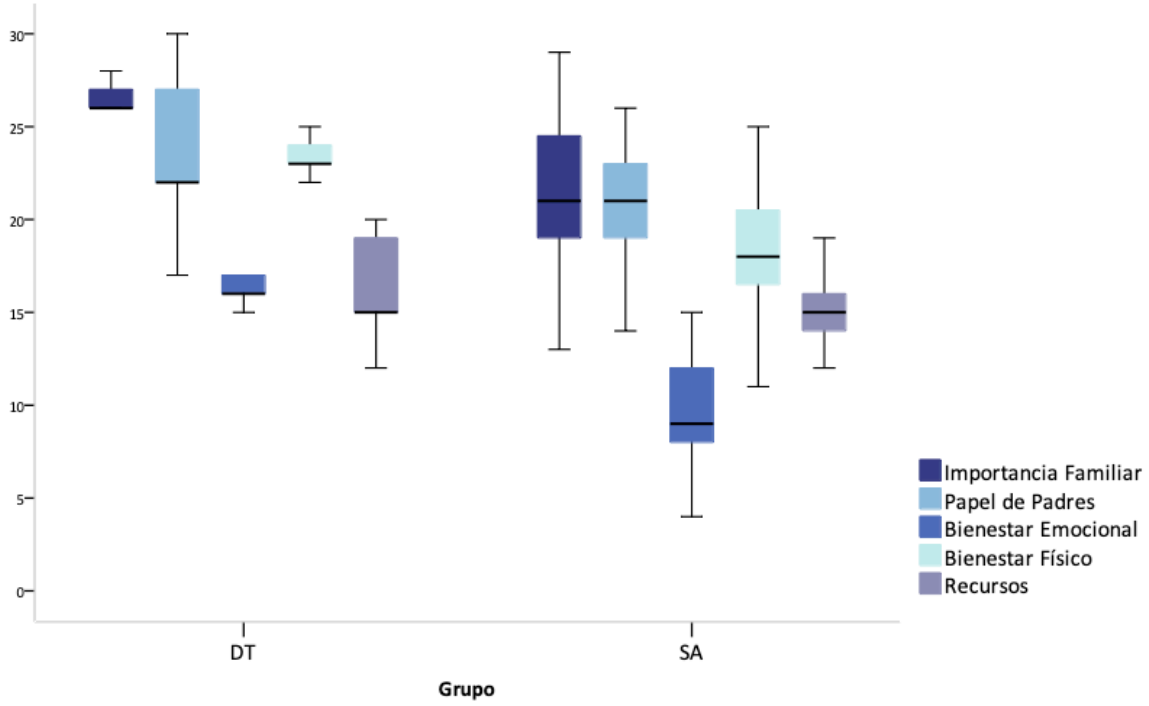


Figura 5.1. Box plots para las variables de satisfacción en CdVF en los dos grupos evaluados.

En relación a la importancia percibida (ver figura 5.2) de dichas áreas de CdVF, los resultados mostraron diferencias entre los grupos en bienestar emocional, y recursos y apoyos (ambas con $p < .05$ y con un tamaño del efecto medio, $\eta^2 = .11$ y $\eta^2 = .35$ respectivamente). Sin embargo, no se encontraron diferencias significativas en interacción familiar ($p = .08$), papel de padres ($p = .37$), ni recursos y apoyos ($p = .06$).

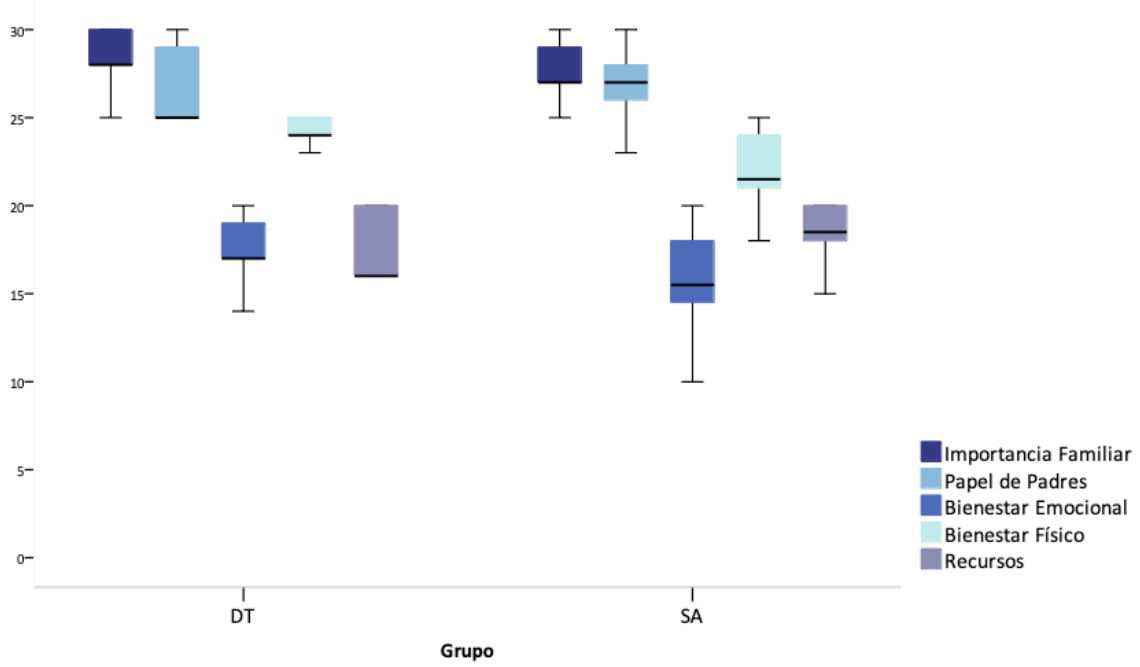


Figura 5.2. Box plots para las variables de importancia en CdVF en los dos grupos evaluados.

Conducta adaptativa

Los resultados mostraron que existen diferencias significativas con un efecto grande entre los grupos en la escala SDQ (ver Figura 5.3) en las variables de síntomas emocionales ($p < .001$, $\eta^2 = .38$), problemas con los compañeros ($p < .001$, $\eta^2 = .48$) y conducta prosocial ($p < .001$, $\eta^2 = .39$). También se muestran diferencias con un tamaño del efecto medio en las variables problemas de conducta ($p < .005$, $\eta^2 = .17$) e hiperactividad ($p < .001$, $\eta^2 = .24$).

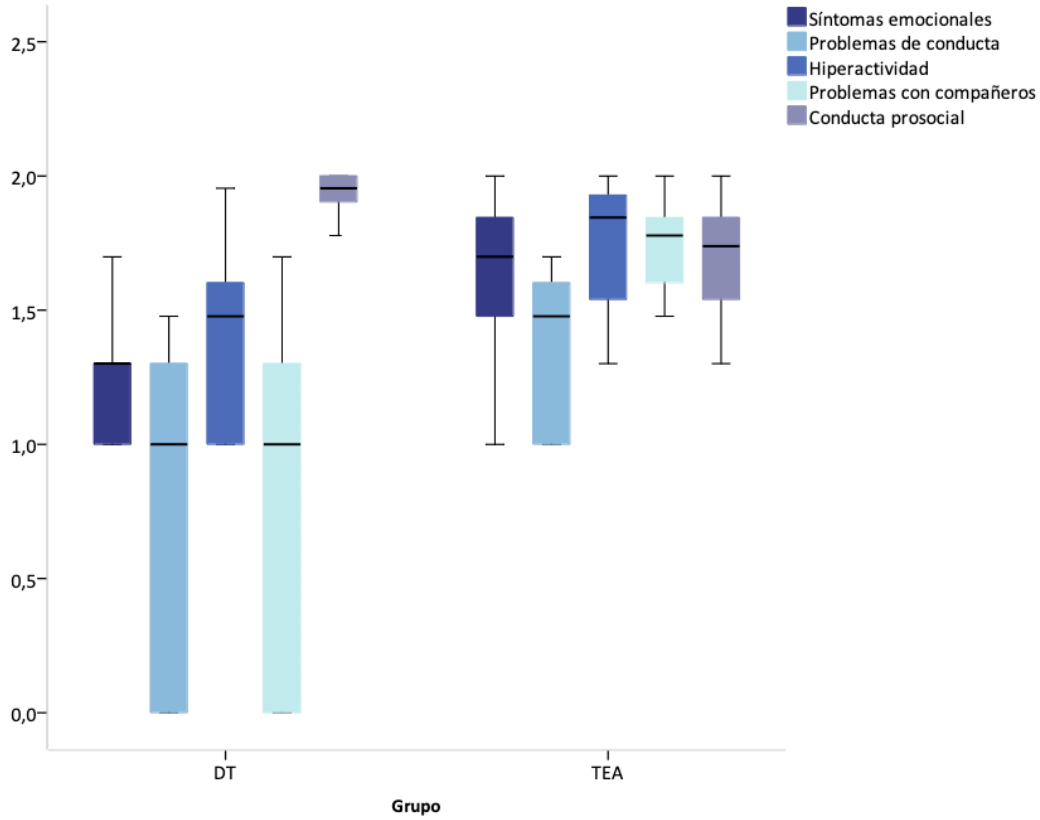
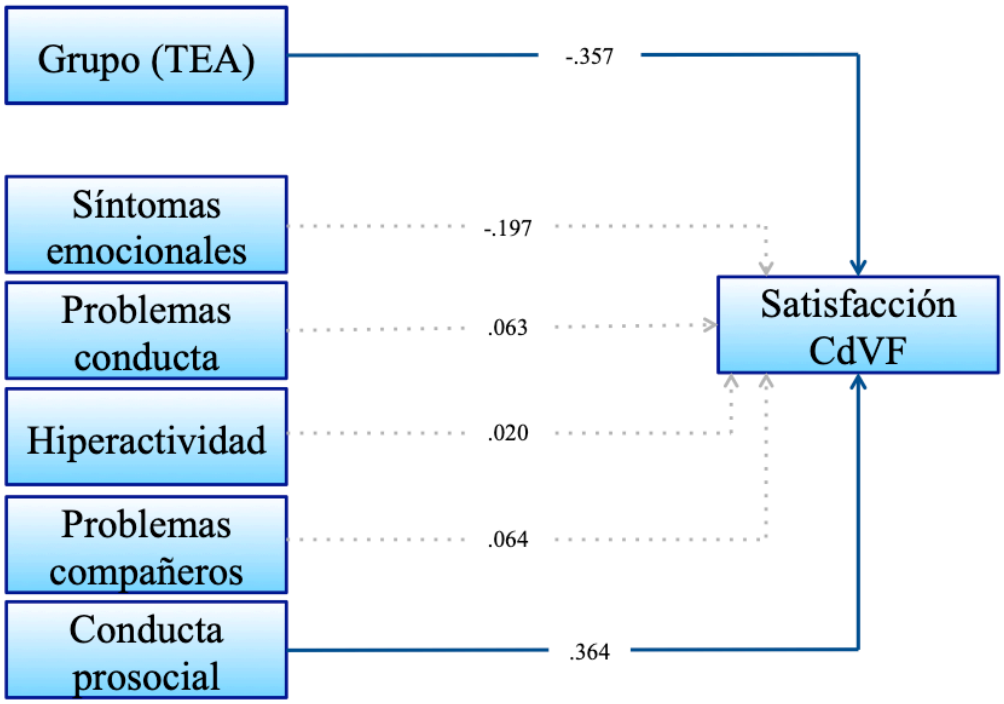


Figura 5.3. Box plots para las variables del SDQ en los dos grupos evaluados.

Hemos realizado un análisis de regresión múltiple con la satisfacción percibida en CdVF como variable dependiente. El resto de variables (grupo, síntomas emocionales, problemas de conducta, hiperactividad, problemas con los compañeros, y conducta prosocial) fueron incluidas como potenciales predictores.

El modelo (ver Figura 5.4) explica un 42% de la variabilidad total, F (6,42) = 6.849, $p < .001$. En este modelo, pertenecer al grupo de DT (β = .36, p = .047, R^2 = 38%) y presentar una mayor conducta prosocial (β = .36, p = .018, R^2 = 37%) predicen una mayor satisfacción percibida en CdVF. Los restantes potenciales predictores no contribuyen al modelo (síntomas emocionales $p = .25$, problemas de conducta $p = .65$, hiperactividad $p = .90$, y problemas con compañeros $p = .72$).



Nota: Las trayectorias significativas (p < .05) se muestran con líneas negras continuas. Los coeficientes representan las β estandarizadas.

Figura 5.4. Resultados del análisis de regresión

IV. Discusión

En este estudio hemos examinado el papel de la conducta adaptativa en niños con TEA y su relación con la satisfacción en CdVF. Nuestros resultados han mostrado que existen diferencias significativas entre los grupos en todas las variables relacionadas con la conducta adaptativa (síntomas emocionales, problemas de conducta, hiperactividad, problemas con los compañeros, y conducta prosocial) y en las variables de interacción familiar, bienestar emocional y bienestar físico de la CdVF. Estos resultados se mantenían controlando el género, la edad, el vocabulario receptivo y la inteligencia.

Nuestros resultados concuerdan con la literatura existente que defiende que aparecen diferencias en la satisfacción percibida en CdVF en las familias de niños con TEA (Cohen et al., 2014; Garrido et al., 2015; Vasilopoulou & Nisbet, 2016). Del mismo modo, los

resultados hallados en el área del funcionamiento emocional y conductual concuerdan parcialmente con los encontrados por otros autores (Russell et al., 2013). Por tanto, este estudio confirma la idea de que los niños con TEA suelen presentar problemas adicionales (emocionales y conductuales) a los que aparecen en los criterios diagnósticos tal y como se defiende en otros trabajos (Helland & Helland, 2017; Posserud et al., 2018).

Los resultados de nuestro estudio arrojan luz sobre la capacidad predictiva de la conducta adaptativa sobre la CdVF. Específicamente, la conducta prosocial predecía junto con el grupo la satisfacción percibida en la CdVF por los padres. Es decir, aquellos que mejor puntuaban en conducta prosocial presentaban una mejor satisfacción con su CdVF. En cuanto al grupo, pertenecer al grupo TEA se relacionaba con unos niveles inferiores de CdVF. Estos resultados concuerdan con otros trabajos que informan del potencial predictor que tienen los problemas emocionales y/o conductuales sobre la CdVF (Eapen & Guan, 2016; McStay et al., 2014).

Sin embargo, aunque existían diferencias entre los dos grupos en las variables relacionadas con los problemas conductuales, estas variables no contribuían en el modelo de regresión. Nuestros resultados no concuerdan con los encontrados por otros autores, donde se sugieren que los problemas conductuales explican los niveles de satisfacción de los padres de niños con TEA (Baghdadli et al., 2014).

Estos resultados se deben considerar bajo ciertas limitaciones. Primero, el número reducido de participantes de ambos grupos no nos permite generalizar los resultados y nos impide estadísticamente incluir otros potenciales predictores que serían de interés para el estudio de la CdVF. Segundo, los participantes fueron en su mayoría madres, por lo que quizás con un número similar de padres se podrían obtener resultados diferentes. Finalmente, dada la edad de los niños con TEA, ha sido imposible obtener medidas directas de la CdVF. Futuros estudios deberían considerar la propia percepción de las personas con TEA.

En suma, como se ha puesto de manifiesto en este trabajo, el TEA se asocia con un riesgo elevado de problemas de conducta, hiperactividad, dificultades con los compañeros y dificultades emocionales. Además, la conducta prosocial predice en parte la satisfacción en la CdVF percibida. Por tanto, las intervenciones que mejoren las habilidades prosociales

podrían tener un efecto directo tanto en el individuo como sobre el funcionamiento y la CdVF.

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CHAPTER 6

Chapter 6

Language comprehension in non–verbal children with ASD (level of support 3)

The content of this chapter has been published as Garrido, D., Carballo, G., Franco, V., & Garcia-Retamero, R. (2015). Dificultades de comprensión del lenguaje en niños no verbales con trastornos del espectro autista y sus implicaciones en la calidad de vida familiar. *Revista Neurología*, *60*, 207–14, doi: 10.33588/rn.6005.2014226

Dificultades de comprensión del lenguaje en niños no verbales con trastornos del espectro autista y sus implicaciones en la calidad de vida familiar

El nivel de comprensión del lenguaje en niños con trastornos del espectro autista (TEA) varía ampliamente. Sin embargo, la evidencia sugiere que estos niños comprenden el lenguaje peor que los de su misma edad con desarrollo típico (DT), y muestran retraso en el vocabulario receptivo. La investigación que relaciona calidad de vida y lenguaje es muy escasa. Los objetivos de este trabajo son profundizar en la comprensión de aspectos estructurales del lenguaje en niños con TEA y conocer la influencia de los déficits en comprensión del lenguaje en niños con TEA en las percepciones sobre calidad de vida en sus familias. En este trabajo se analiza la comprensión verbal en 26 niños no verbales con TEA (media: 9.8 años) y en 26 niños con DT (media: 3.9 años) igualados en edad de vocabulario, utilizando medidas estandarizadas de lenguaje receptivo. Los resultados muestran que el nivel de vocabulario receptivo, comprensión auditiva y comprensión gramatical en los niños con TEA está por debajo del que corresponde a su edad, y difiere significativamente de aquél en niños con DT. Asimismo, los padres de niños con TEA informan de graves problemas de comunicación en sus hijos y falta de apoyo social. La calidad de vida familiar se ve afectada por los problemas lingüísticos de los niños con TEA. Encontramos una importante relación entre las habilidades de lenguaje receptivo en los niños con TEA y las percepciones sobre la calidad de vida en sus familias. Estos resultados pueden tener importantes implicaciones en el diseño de intervenciones clínicas.

I. Introducción

La comunicación es uno de los aspectos más frecuentemente afectados y de los primeros indicadores en los trastornos del espectro autista (TEA) (Whitehouse, Barry, & Bishop, 2008). La investigación ha mostrado una gran variabilidad en los niveles lingüísticos estructurales en niños con TEA, incluyendo desde niños que presentan un vocabulario relativamente extenso, aunque con ecolalia y variaciones prosódicas y fonológicas, a los que muestran incapacidad total para el habla (Kjelgaard & Tager–Flusberg, 2001; Kostyuk et al. 2010; Moreno–Flagge, 2013).

De hecho, alrededor de una cuarta parte de la población con TEA son no verbales y muestra limitaciones comunicativas graves (Luyster, Kadlec, Carter, & Tager–Flusberg, 2008). Hasta la fecha, las descripciones lingüísticas de los niños verbales con TEA se han centrado, fundamentalmente, en enfatizar la ausencia de habilidades verbales, el retraso temprano del lenguaje y las habilidades pragmáticas alteradas (Eigsti, Bennetto, & Dadlani, 2007), sin profundizar en la comprensión de aspectos estructurales del lenguaje en esta población, ni en las consecuencias que dichas limitaciones tienen para ellos y sus familias.

Se ha comprobado que la comprensión es especialmente vulnerable en los niños con TEA y está más gravemente deteriorada que la expresión (Hudry et al., 2010; Rapin & Dunn, 2003; Watson, 2002; Ellis Weismer, Lord, & Esler, 2010). El vocabulario receptivo en niños pequeños de 2–3 años es significativamente menor que el expresivo (Ellis Weismer et al., 2010). Así, Eigsti et al. (2007) muestran que, a los 5 años, los niños con TEA podían decir formas complejas de palabras en comparación con formas simples, lo que puede indicar que no siempre comprenden las formas más complejas que producen, y que su lenguaje puede estar sobreestimado basándose en su superficialidad (Miniscalco, Fränberg, Schachinger–Lorentzon, & Gillberg, 2012), empleando medidas estandarizadas (Jarrold, Boucher, & Russell, 1997) y medidas de información paternas (Kjelgaard & Tager–Flusberg, 2001). Otros autores, sin embargo, no encuentran diferencias entre niveles de realización expresiva y receptiva, y concluyen que, al igual que los niños con desarrollo típico (DT), los niños con TEA comprenden el lenguaje (por ejemplo, las preguntas complejas tipo 'qu–') antes de producirlo (Goodwin, Fein, & Naigles, 2012).

El TEA puede influir considerablemente en la calidad de vida familiar (CdVF). La calidad de vida es una medida de bienestar de la persona que se utiliza con frecuencia en el estudio del desarrollo de las discapacidades (Lee, Harrington, Louie, & Newschaffer, 2008), y puede conceptualizarse como un constructo multidimensional e influido por agentes personales y ambientales. Es, a la vez, subjetiva y objetiva, y se ve reforzada por la autodeterminación, los recursos, el propósito en la vida y un sentido de pertenencia (Cummins, 2005).

La CdVF se ve alterada por múltiples factores unidos a la condición compleja del TEA: discapacidad, interacción social, problemas de comunicación, de conducta, con autolesiones, rituales complejos y rabietas, que pueden ser difíciles de manejar y que interfieren en la vida

cotidiana (Lee et al., 2008), por lo que es necesario un mayor apoyo social y asistencia con el fin de lograr y mantener el bienestar de toda la familia.

Las investigaciones que relacionan CdVF y lenguaje en niños con TEA son muy escasas (Lee et al., 2008; Allik, Larsson, & Smedje, 2006), y sugieren que el estrés emocional de los padres de niños con TEA está negativamente asociado con la habilidad de los niños para comunicarse funcionalmente (Ello & Donovan, 2005). Sin embargo, hasta la fecha, no se ha estudiado la influencia de las habilidades receptivas del lenguaje en niños con TEA sobre la CdVF.

En este trabajo perseguimos: (a) conocer los niveles de comprensión del lenguaje en niños no verbales con TEA en comparación con niños con DT, (b) profundizar en la comprensión de aspectos estructurales del lenguaje en niños con TEA, para comprender su funcionamiento; específicamente, pretendemos comprobar si existe un déficit de comprensión gramatical, (c) conocer el efecto que tienen los TEA en la calidad de vida familiar, y (d) comprobar la influencia que las habilidades receptivas del lenguaje tienen sobre la CdVF en niños con TEA y DT.

II. Método

II.I. Participantes

En el estudio participaron 52 niños distribuidos en dos grupos (n = 26) compuestos por niños con TEA y DT, respectivamente. Los del grupo con DT se igualaron a los del grupo con TEA en edad de vocabulario receptivo con el fin de partir de niveles similares de comprensión auditiva y para asegurarnos de que los niños obtuvieran una medida verbal sintáctica similar. Se verificó la pertenencia a cada grupo mediante el cuestionario de comunicación social (SCQ) y la escala de evaluación de personas con autismo de Gilliam (GARS). Los participantes en el grupo con TEA tenían una edad de 4.7–15.11 años (media: 9.7±3.1 años; 69% hombres), y en el grupo con DT, 2.0–10.3 años (media: 3.9±1.1 años; 69% hombres) (ver Figuras 6.1 y 6.2).

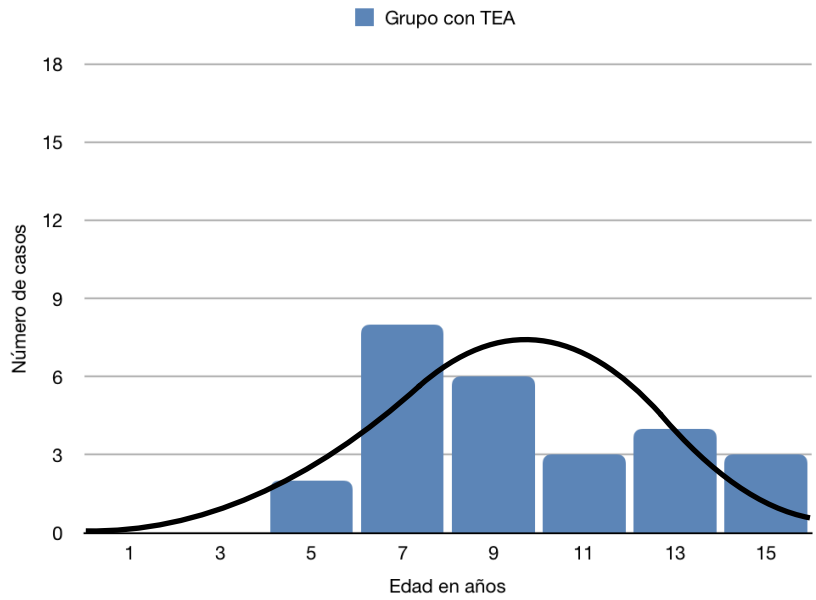

Figura 6.1. Distribución de los participantes en el grupo con trastorno del espectro autista (TEA) según la edad.

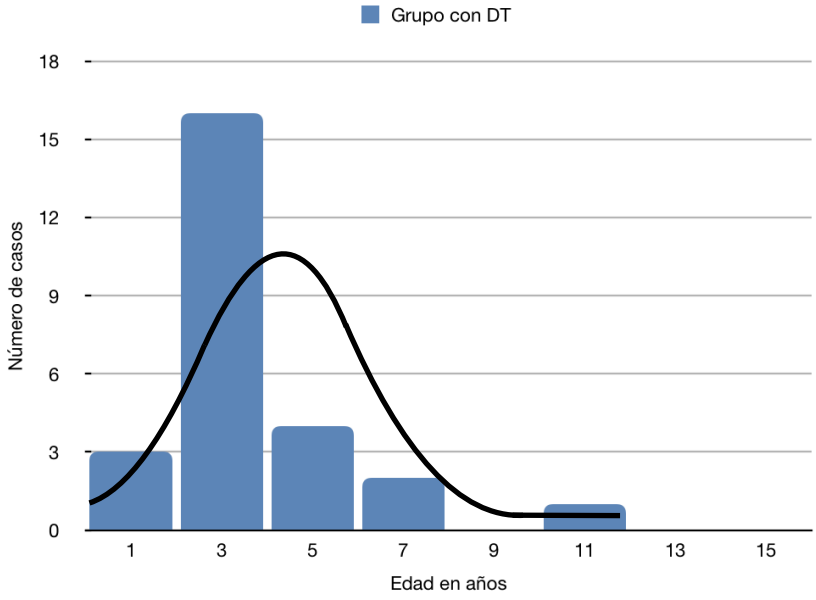

Figura 6.2. Distribución de los participantes en el grupo con desarrollo típico (DT) según la edad.

El rango de edad menor en el grupo con DT se debe a que obtienen mejores puntuaciones en el test de vocabulario receptivo a edades más tempranas (véase medidas), y a la igualación uno a uno de cada participante de ambos grupos en esta prueba para tener niveles equivales de vocabulario receptivo, aunque alcanzan el nivel a diferentes edades cronológicas (de forma similar a lo realizado por Eigsti et al. (2007). Las familias de los niños del grupo con TEA eran miembros de una asociación de apoyo a personas y familias con TEA, y de una asociación de autismo en Granada. Los niños con DT estaban escolarizados en escuelas infantiles o centros escolares de Granada. La selección de los participantes se llevó a cabo contactando con estas asociaciones y centros escolares.

Los criterios de inclusión para el grupo con TEA fueron: haber recibido el diagnóstico de TEA basado en los criterios del Manual diagnóstico y estadístico de los trastornos mentales, cuarta edición, texto revisado, y en la administración a los padres de la entrevista para el diagnóstico del autismo revisada (Rutter, Le Couteur, & Lord, 2010), no presentar ningún trastorno comórbido y no tener lenguaje verbal o que éste fuera no funcional. Para los participantes en el grupo con DT, fueron: no presentar ningún trastorno previo (p. ej., trastorno por déficit de atención e hiperactividad, síndrome de Down, parálisis cerebral…) y no tener historia familiar previa de TEA.

II.II. Medidas

Medidas de igualación y clasificación de los grupos

Test de vocabulario receptivo (PPVT–III Peabody; Dunn, Dunn, & Arribas, 2006). Permite la realización de un cribado de la aptitud verbal y la obtención de la edad equivalente de comprensión auditiva. Esta prueba se utilizó para partir de niveles similares de comprensión auditiva entre los niños con TEA y DT más pequeños, dado que no requiere una respuesta verbal.

Escala de Autistas de Guilliam (GARS; Guilliam, 2004). Permite cuantificar la gravedad del autismo y evalúa la comunicación no verbal.

Cuestionario de Comunicación Social (SCQ; Rutter, Bayley, & Lord, 2005). Procedimiento específico de diagnóstico de autismo, tipo cribado, que proporciona puntuaciones en interacción social, dificultades de comunicación, conducta restringida, repetitiva y estereotipada.

Subtest de cociente de inteligencia breve, memoria y atención sostenida de la escala manipulativa internacional de LEITER–R (Roid & Miller, 2011). Para niños entre 2 y 20 años.

Evaluación del lenguaje comprensivo y la comunicación

Test Token para niños (TTFC–2; McGhee, Ehrler, & DiSimoni, 2007). Evalúa la comprensión auditiva de órdenes.

Test de comprensión de estructuras gramaticales (CEG; Mendoza, Carballo, Fresneda, & Muñoz, 2005). Evalúa la comprensión de construcciones gramaticales de diferente complejidad en niños de 4–12 años. Hemos utilizado la versión CEG 2–4 para los niños más pequeños del grupo control (Calet, Mendoza, Carballo, Fresneda, & Muñoz, 2010).

Subescala de comunicación de la GARS (Guilliam, 2004). Evalúa conductas estereotipadas, comunicación, interacción social y alteraciones en el desarrollo.

Otras medidas

Cuestionario de apoyo social. De elaboración propia, permite conocer el número de amigos de los niños, la calidad de la relación con sus familiares y el número de contactos semanales que tienen con familiares y amigos.

Escala de calidad de vida familiar (ECVF; Verdugo, Rodriguez, & Sainz, 2009). Evalúa el grado de importancia y satisfacción en cada uno de los siguientes indicadores: interacción familiar, papel parental, bienestar emocional, bienestar físico y material, y recursos y apoyo para personas con discapacidad.

Cuestionario para familiares de personas con TEA. De elaboración propia, proporciona información demográfica, diagnóstica, de interacción social, comunicación, intereses, desarrollo cognitivo, conducta motora y postural, estimulación sensorial, alimentación y estado de ánimo.

II.III. Procedimiento

Una vez firmado el consentimiento informado por los padres o tutores de los niños, se administraron las pruebas descritas. Los niños completaron las escalas PPVT–III, LEITER–R, TTFC–2 y CEG. Los padres completaron las escalas GARS, SCQ, ECVF, apoyo social y cuestionarios para familiares. Los cuestionarios fueron completados por niños y padres simultáneamente en el domicilio familiar en 2–4 sesiones, según necesidades y tiempo disponible.

II.IV. Análisis de datos

En primer lugar, realizamos análisis para verificar la clasificación de los participantes en los grupos con TEA y DT, y si éstos muestran el mismo nivel de comprensión de vocabulario receptivo. En segundo lugar, analizamos las diferencias entre grupos en el nivel de lenguaje comprensivo (comprensión auditiva y estructuras gramaticales), comunicación no verbal y apoyo social en niños con TEA y DT, y calidad de vida en sus padres. Para ello, se realizan análisis de covarianza (ANCOVA), incluyendo la edad, el género y el cociente intelectual de los niños como covariados, y el grupo (TEA frente a DT) como factor.

Finalmente, realizamos un análisis mediacional para evaluar si las diferencias en la percepción sobre calidad de vida en los padres de niños con TEA y DT están mediadas por el nivel de lenguaje comprensivo de los niños. Dicho análisis va dirigido a identificar y evaluar el mecanismo o proceso que explica la relación entre dos variables (Baron & Kenny, 1986; Hayes, 2009). En nuestro modelo, el predictor o variable independiente es el grupo (TEA frente a DT), y el criterio o variable dependiente es el nivel de calidad de vida familiar. La variable mediadora es el nivel de lenguaje comprensivo en los niños. En lugar de asumir una relación directa entre el predictor y el criterio, en el modelo mediacional se asume que el predictor influye sobre la variable mediadora; y ésta, a su vez, influye sobre el criterio. La mediación, por tanto, tiene lugar cuando una tercera variable desempeña un papel importante en la relación entre otras dos variables (Sobel, 1982). En estos análisis se ha controlado el efecto del nivel de apoyo social en los niños, ya que, de otro modo, éste podría explicar la relación entre el grupo (TEA o DT) y las percepciones sobre la calidad de vida. Se ha utilizado Statistica v. 10 para la realización de los análisis.

III. Resultados

Igualación de los grupos en lenguaje comprensivo y clasificación de los participantes

Como se esperaba, no hay diferencias significativas entre los grupos de niños con TEA y DT en vocabulario receptivo ($p > .05$; Tabla 6.1), lo que permite concluir que ambos grupos se han igualado en cuanto a nivel de vocabulario comprensivo.

Tabla 6.1. Medias y EEM en las áreas evaluadas en los grupos con TEA y DT y resultados en los ANCOVAS

Áreas evaluadas		Grupo				Análisis		
		TEA		DT		*F*	*p*	η^2 parcial
		M	EEM	M	EEM			
Igualación de grupos y clasificación de participantes	Vocabulario receptivo (PPVT-III)	49.69	5.43	49.35	5.45	3.30	.120	.07
	Nivel de autismo (GARS)	90.08	3.42	51.46	4.03	24.14	.001*	.34
	Nivel de autismo (SCQ)	21.81	1.09	5.15	0.70	43.92	.001*	.48
Lenguaje comprensivo y comunicación no verbal	Comprensión auditiva (TTFC-2)	10.31	2.69	48.81	6.16	9.88	.003*	.17
	Estructuras gramaticales (CEG)	2.15	0.50	6.42	0.58	16.72	.001*	.26
	Subescala de comunicación (GARS)	9.00	0.58	3.88	0.64	34.81	.001*	.41
Apoyo social y calidad de vida	Apoyo social	6.18	0.53	8.24	0.40	7.04	.011*	.13
	Calidad de vida: Satisfacción (CdVF)	17.10	0.51	21.78	0.41	6.92	.011*	.13
	Calidad de vida: Importancia (CdVF)	22.21	0.57	22.98	0.43	1.08	.303	.02

Nota: M = media; EEM = Error estándar de la media. Interpretación de η^2 parcial = <.01 efecto pequeño; >.06 efecto medio; >.14 efecto grande (Cohen, 1988). * = $p < .05$.

También se encuentran diferencias marcadas entre los grupos en las puntuaciones en las escalas GARS ($p < .005$) y SCQ ($p < .005$), lo que permite verificar el diagnóstico en el grupo de niños con TEA y confirmar que todos los participantes del grupo control tienen un DT.

Evaluación del lenguaje comprensivo y la comunicación no verbal

Como se muestra en la Tabla 6.1, los niños en el grupo con TEA obtienen una puntuación típica y un percentil muy por debajo de su edad en comprensión auditiva ($p < .005$), y muestran puntuaciones más bajas en comprensión de estructuras gramaticales ($p < .005$) y comunicación no verbal ($p < .005$) que los niños con DT.

Evaluación del nivel de apoyo social y calidad de vida

Existen diferencias significativas entre grupos en el nivel de apoyo social recibido por los niños ($p < .05$) y en el grado de satisfacción mostrado por sus respectivas familias en calidad de vida ($p < .05$). Por el contrario, no hay diferencias significativas en la importancia concedida a los distintos aspectos medidos sobre calidad de vida, siendo todos ellos igualmente importantes para las familias de niños con TEA y DT ($p > .05$).

Análisis mediacional

En línea con los resultados previos, los participantes en el grupo con TEA muestran menores niveles en comprensión de estructuras gramaticales ($\beta = .69$; $t(47) = 3.73$; $p < .001$), comprensión auditiva ($\beta = .54$; $t(47) = 2.68$; $p < .05$) y comunicación no verbal ($\beta = -.65$; $t(47) = -3.18$; $p < .005$) que los participantes en el grupo con DT (ver Figura 6.3). Del mismo modo, los familiares de los participantes en el grupo con TEA muestran menor satisfacción con su calidad de vida que los del grupo con DT ($\beta = .46$; $t(47) = 2.31$; $p < .05$). Los familiares en ambos grupos, sin embargo, conceden la misma importancia a las diferentes dimensiones consideradas en la medida de calidad de vida ($\beta = -.37$; $t(47) = 1.38$; $p = .18$).

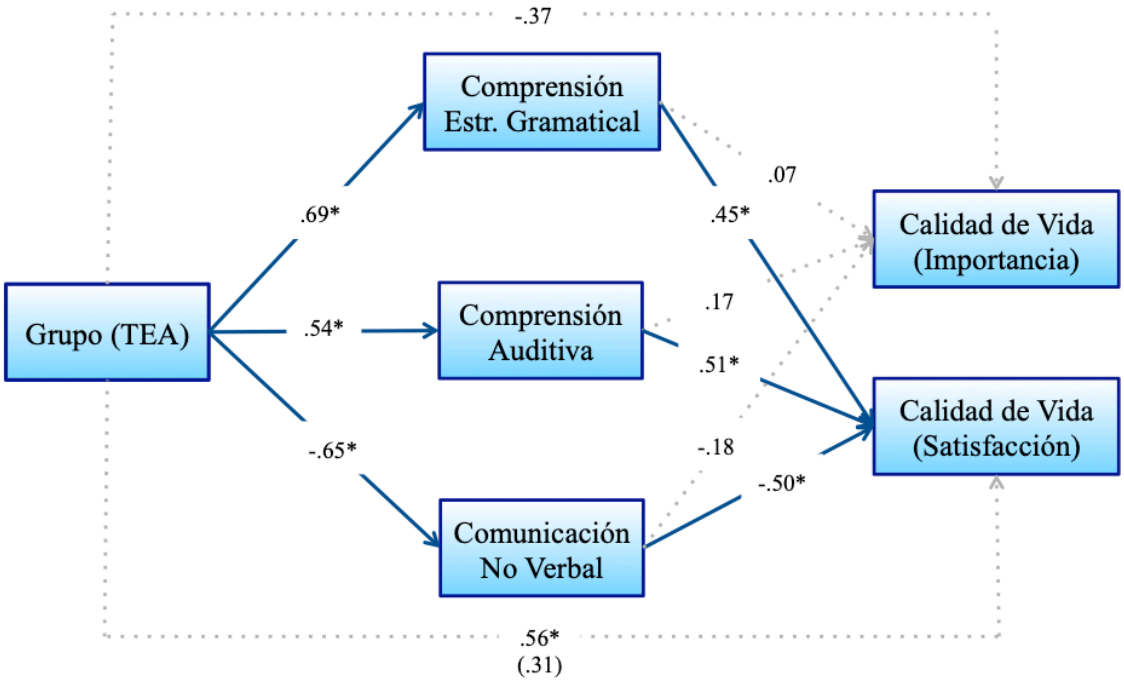

Figura 6.3. Influencia del trastorno del espectro autista sobre la calidad de vida y efecto mediador del nivel de lenguaje comprensivo.

Cuando se incluye el nivel de lenguaje comprensivo (comprensión auditiva y estructuras gramaticales) y de comunicación no verbal en los análisis, el efecto del grupo sobre el nivel de satisfacción en calidad de vida disminuye y deja de ser significativo ($\beta = .31$; $t(44) = .70$; $p < .05$). Este dato es consistente con los resultados en el test de Sobel ($z = 2.51$ y $p < .01$ para comprensión de estructuras gramaticales; $z = 2.28$ y $p < .05$ para comprensión auditiva; y $z = 2.46$ y $p < .01$ para comunicación no verbal), y pone de manifiesto que las diferencias en el nivel de satisfacción en los familiares en los grupos con TEA y DT están mediadas por las diferencias en el nivel de lenguaje comprensivo y comunicación de sus hijos.

IV. Discusión

Los resultados ponen de manifiesto que la comprensión del lenguaje parece ser especialmente vulnerable en los niños con TEA. No existen diferencias significativas entre estos niños y aquéllos con DT en el lenguaje comprensivo, aunque los niños con TEA muestran deterioro semántico, con un vocabulario por debajo de la edad esperada y semejante al de los niños más pequeños con DT. Sin embargo, en comprensión auditiva, los niños con TEA muestran realizaciones muy inferiores a las esperadas para su edad, deficiencias en el lenguaje receptivo que también aparecen en niños verbales con TEA (Condouris, Meyer, & Tager–Flusberg, 2003; Kjellmer, et al., 2012). Chan, Cheung, Leung, Cheung, y Cheung (2005) evalúan la comprensión de frases en niños con TEA de 5–6 años, y observan una realización considerablemente más baja que en DT, pero no estadísticamente significativa. En contraste, Äsberg (2010) no encuentra diferencias en el vocabulario receptivo o la gramática receptiva entre niños con TEA y DT, lo que confirma la heterogeneidad del lenguaje en esta población.

En la comprensión de estructuras gramaticales, también encontramos diferencias significativas entre ambos grupos, datos que concuerdan con otros trabajos realizados (Rapin & Dunn, 2003) y que confirman su contribución importante a los problemas comunicativos. Eigsti y Bennetto (2009) observan trastornos gramaticales en la mayoría de niños con TEA de alto funcionamiento, entre 9–17 años, en tareas de juicio gramatical. En nuestro estudio, las estructuras que resultan más difíciles para los niños con TEA son las que no siguen el orden estructural común, sujeto–verbo–objeto (SVO), como las oraciones coordinadas o las de relativo, en las que todos los niños fallan. Por el contrario, las estructuras más fáciles son las de SVO, con aciertos del 60%, seguidas de las atributivas (30%). Swensen, Kelley, Fein, y Naigles (2007) también afirman que los niños con TEA comprenden frases en este orden, SVO, antes de producir habla continua. Hasta la fecha, no disponemos de ningún trabajo sobre estructuras gramaticales del español en TEA. Los resultados, por tanto, muestran que la comprensión del lenguaje –y especialmente las estructuras gramaticales– es una de las áreas de debilidad en TEA que puede contribuir a explicar algunos de los déficits comunicativos encontrados, aunque existe muy poca evidencia sobre las alteraciones gramaticales (Boucher, 2012), por lo que se muestra como un área de investigación abierta.

También encontramos diferencias muy marcadas en comunicación entre los grupos de niños con TEA y DT, dato relevante, ya que la alteración en comunicación es uno de los rasgos que de forma típica describen el autismo y los TEA, y uno de los criterios principales que caracterizan el trastorno en el Manual diagnóstico y estadístico de los trastornos mentales, cuarta edición, con un consenso general tanto investigador como clínico.

Finalmente, encontramos diferencias marcadas en apoyo social en niños con TEA y DT y calidad de vida en sus familiares. Para Burgess y Gutstein (2007), la calidad –y no la cantidad– del apoyo social que reciben las personas es un predictor muy fiable de la percepción de calidad de vida en población con TEA. Las relaciones que carecen de intimidad, reciprocidad y enriquecimiento emocional provocan soledad intensa y frecuente, a pesar de la creencia común de que los niños con TEA prefieren estar solos (Bauminger & Kasari, 2000).

Las interrupciones en la vida familiar habitualmente presentadas en niños con TEA (demandas de cuidado y dependencia a largo plazo) tienen un efecto negativo en la calidad de vida de sus familias, que podría tener importantes consecuencias para la salud física y psicológica (Khanna et al., 2011). Nuestro estudio pone de manifiesto, por primera vez, que las diferencias en el grado de satisfacción con la calidad de vida percibida en los familiares de niños con TEA y DT se pueden explicar, en parte, por las diferencias en el nivel de lenguaje comprensivo y comunicación de sus hijos. Este dato podría tener importantes implicaciones clínicas, ya que las intervenciones dirigidas a mejorar el lenguaje comprensivo o las limitaciones en comunicación no verbal en niños con TEA podrían paliar o disminuir sustancialmente las percepciones de insatisfacción con la calidad de vida en sus familias.

En este sentido, Allik et al. (2006) también informan de que los padres de niños con TEA tienen una mayor carga familiar y se encuentran en mayor riesgo de sufrir malestar físico y psicológico que los padres de niños con DT, y encuentran relación entre bienestar materno y características conductuales del niño. Del mismo modo, Lee et al. (2008) observan que los padres de niños con autismo entre 3 y 17 años tienen un nivel más alto de preocupación por el bienestar de su hijo, mayores percepciones de sobrecarga de cuidado infantil y menor participación en las actividades comunitarias, por tanto, su calidad de vida es menor que la de los padres de niños con DT. Estos resultados contrastan con los trabajos de Park, Yelland, Taffe, y Gray (2012), en los que no se encuentra relación entre habilidades estructurales del

lenguaje y habilidades sociales y calidad de vida. Nuestro estudio, por tanto, podría resultar una aportación novedosa en este sentido, ya que puede clarificar la relación entre estos conceptos en la población con TEA.

En suma, como resultado de nuestro trabajo, podemos afirmar que, aunque los grupos se han igualado en vocabulario receptivo, la comprensión del lenguaje está ampliamente deteriorada en los niños no verbales con TEA, y que, a pesar de que el deterioro de la comprensión no es un criterio diagnóstico, se debe tener en cuenta en la intervención. El estrés emocional de los padres de los niños con TEA parece estar ligado a los problemas de comunicación de los niños. Tal y como afirman Ello y Donovan (2005), dicho estrés está negativamente asociado con la habilidad de los niños con TEA para comunicarse funcionalmente. Es decir, las diferencias en el nivel de lenguaje comprensivo y de comunicación de sus hijos en los grupos con TEA y DT miden (explican) las diferencias en el nivel de satisfacción en los familiares. No obstante, la imposibilidad de evaluar el lenguaje expresivo en los niños con TEA en nuestro estudio hace que los resultados no se puedan generalizar a los niños verbales con TEA. Sería de interés comprobar, en futuras investigaciones, si existen diferencias entre lenguaje expresivo y receptivo en esta población y la magnitud de tales diferencias, posiblemente utilizando otros grupos de comparación en los que existan alteraciones claras del lenguaje.

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PART IV

BEYOND CHILDREN WITH AUTISM SPECTRUM DISORDER: PARENTS AND SIBLINGS

CHAPTER 7

Chapter 7
Improving social–communication and family quality of life through a parent-mediated intervention

The content of this chapter has been published as Garrido, D., García-Retamero, R., & Carballo, G. (under review). Improving social–communication management and family quality of life through parent-mediated intervention in autism spectrum disorder.

Improving social–communication management and family quality of life through parent-mediated intervention in autism spectrum disorder

Research shows that interventions conducted in parents of children with autism spectrum disorders (ASD) effectively improve family quality of life (FQoL). However, these interventions tend to be long and require several sessions. In the current research we examined the effect of a brief intervention on parental behavior (i.e., strategies about communication, behavior, and parental stress) and FQoL. We compared results in the intervention group (N = 20) with those in a waitlist (control) group (N = 20). The intervention covered significant topics including knowledge of ASD, supporting attention and motivation, development of functional language, behavioral functioning, and stress control. Compared to those in the control group, parents who received the intervention reported improvements in FQoL (i.e., family interaction, and emotional well-being) and gains on language-communication management strategies and behavioral strategies. No differences were found on stress management strategies. Results showed a good treatment adherence. We conclude that brief intervention can improve FQoL, parental management of strategies related to language-communication development, and behavioral management.

I. Introduction

Autism spectrum disorder (ASD) is characterized by social-communication challenges and restricted and repetitive behaviors (APA, 2013). Because of these features, parents who have a child with ASD show higher levels of stress and anxiety, as well as lower levels of happiness if we compare them to those families that have children with typical development (Dabrowska & Pisula, 2010; Hastings, 2003; Hayes & Watson, 2013; for a systematic review, see Vasilopoulou & Nisbet, 2016). Thus, parents of children with ASD are at risk of experiencing low family quality of life (FQoL), and physical and psychological distress, which could be linked to the core of ASD symptoms such as difficulties of communication, and behavioral problems (Allik, Larsson, & Smedje, 2006; Ello & Donovan, 2005; Lee, Harrington, Louie, & Newschaffer, 2008). Consequently, there is an increasing need for developing

effective, evidence-based parent interventions that would help address core ASD symptoms and improve their FQoL.

I.I. Family Quality of Life in ASD

FQoL is a global, multidimensional and complex concept that allows us assess the dynamic sense of family well-being, which is consider as a global definition of the family-life situation (Mannan, Summers, Turnbull, & Poston, 2006). The features of a child with ASD have an impact over the family's experience and its FQoL. Members of these families tend to experience higher parental levels of depression or stress, and lower family support (Bundy & Kunce, 2009; Gardiner & Iarocci, 2012). In addition, both characteristics of children with ASD and their parents influence FQoL.

Parent's adjustment, social support and expectations about the development of their child are some of the parents' features that might have consequences for their FQoL (Pozo, Sarriá, & Brioso, 2014). Consequently, parents that are involved in direct intervention might enhance their adjustment, their social support, and their expectations, which could have a direct impact on FQoL. Additionally, research suggests that FQoL could be considered as a good measure of parents' adjustment and an important outcome for assessing the effectiveness of parental interventions for families of children with ASD (Cappe, Wolff, Bobet, & Adrien, 2011; Eapen, Črnčec, Walter, & Tay, 2014).

I.II. Language-communication, behavioral, and stress management in ASD

Parent training and educational interventions provide parents with information and teach them skills that reduce parental levels of stress and increase perceptions of self-efficacy and confidence, which is expected to have a direct impact on family emotional well-being and FQoL (Ayuda-Pascual, Llorente-Comi, Martos-Perez, Rodriguez-Bausa, & Olmo-Remesal, 2012; Feinberg et al., 2014).

For instance, Samadi, McConkey, & Kelly (2012) found that a direct intervention could have an impact on parental emotional well-being, stress, self-efficacy, problem-focused coping strategies, and family functioning. Indeed, parents reported that sharing their

concerns with others with similar experiences could help them create a supportive social network, which could have a positive impact over their FQoL.

To date, interventions aiming at promoting parental knowledge about ASD and family well-being have been limited (Dempsey & Keen, 2008; Samadi et al., 2012). For example, Iadarola et al. (2017) found that parents who were involved in an intervention showed an increase in competence, and a decrease in parental strain and stress levels. Most importantly, it seems that parental training could enhance parenting strategies and improve both parents and children's outcomes (Kasari, Gulsrud, Paparella, Hellemann, & Berry, 2015). Promising results have been found about parental skills improvements in different learning abilities, such as language and communication, behavioral, and stress management strategies. As an example, Matson, Mahan, and Matson (2009) reported that parents were able to develop skills to increase their children's language and communication skills, and to decrease their children's challenging behaviors.

In regard to parental management of language and communication abilities, some studies showed that Responsive Teaching (RT; Mahoney & McDonald, 2007), Pivotal Response Treatment (PRT; Koegel & Koegel, 2012), Social Communication, Emotional Regulation, and Transactional Support (SCERTS Model; Prizant, Wetherby, Rubin, & Laurent, 2003), the Denver Model (Rogers & DiLalla, 1991) or Son-RISE Program (SRP; Kaufman & Kaufman, 1976) were associated with increases in children's pivotal behavior, social imitation, child-initiated social engagement, and nonverbal dyadic orienting (Houghton, Schuchard, Lewis, & Thompson, 2013; Karaaslan & Mahoney, 2015; Koegel, Vernon, & Koegel, 2009). Certain studies have reported that RT is effective at enhancing the quality of parents' interactions (Karaaslan, Diken, & Mahoney, 2013; Karaaslan & Mahoney, 2013; Mahoney, Nam, & Perales, 2014; Mahoney & Perales, 2003, 2005; Mahoney, Wiggers, Nam, & Kralovic, 2014). This fact could have an impact on their sense of competence as parents. Minjarez, Williams, Mercier, and Hardan (2011) found that parents provided more opportunities for verbal responses, non-verbal requests, reinforcements of the child's verbal attempt, functional verbal utterances, improvements in adaptive behavior, early cognitive abilities, and the parent's confidence level after PRT.

Concerning behavioral management, several studies have shown that getting the parents involved in behavioral management intervention might reduce their level of stress,

and increase their skills, their sense of parenting competence, and their satisfaction (Dillenburger, Keenan, Gallagher, & McElhinney, 2004; Green et al., 2010; Kasari, Gulsrud, Wong, Kwon, & Locke, 2010; Kasari et al., 2015). For instance, intervention focused on Applied Behavior Analysis (ABA; Cooper, Heron, & Heward, 2007) shows how to reduce disruptive behaviors through environmental manipulations, differential reinforcement, and visual aids, and it shows positive results (Bears, Johnson, Handen, Smith, & Scahill, 2013; Tonge, Brereton, Kiomall, Mackinnon, & Rinehart, 2014). Indeed, Dillenburger et al. (2004) found decreased parenting stress and increased parenting self-efficacy after ABA intervention.

Addressing effectively parental stress is important not only for the increase of the child's well-being. It is also crucial for functioning within the family. Some studies have found a decreased parenting stress and an increased parenting self-efficacy after a professionally supported intervention (Keen, Couzens, Muspratt, & Rodger, 2010; Lindo, Kliemann, Combes, & Frank, 2016). Additionally, Feinberg et al. (2014) found that parents of children with ASD could decrease their parental stress symptoms after a brief cognitive behavioral intervention and problem-solving education. Likewise, Kasari et al. (2015) showed that after providing education and support to parents of young children with ASD, they reported lower parenting stress.

Even if the interventions have been placed on studies that reported positive outcomes, less is known about fidelity of implementation and treatment adherence of these types of interventions in parents of children with ASD (Hock, Kinsman, & Ortaglia, 2015; Schultz, Schmidt, & Stichter, 2011).

I.III. Fidelity of implementation and treatment adherence

In these family–centered practices, implementation fidelity refers to practices used to teach parents new skills (such as trainer experience or education, and providing written directions; Barton & Fettig, 2013). Moreover, some studies have described that parent treatment adherence is associated with demographics (such as age, education, marital status), and parent treatment attitudes and beliefs (Ajzen, 1991; Hock et al., 2015; Moore & Symons, 2011). In particular, empirical evidence supports theories such as the theory of planned

behavior, which suggests that attitudes toward a behavior predict the intentions to perform this behavior (Ajzen & Fishbein, 2005). This theory further suggests that behavioral intentions have a strong impact on actual behaviors. Some authors suggest that describing the potential impact of attitudes of parents on their actual performance could be useful for improving the quality of future interventions (e.g., Houghton et al., 2013).

Another factor that might indirectly affect adherence is the length of the intervention and parental involvement. Parent interventions are one of the approaches with larger cost-effectiveness advantage (Steiner, Koegel, Koegel, & Ence, 2012). However, most of these interventions include many sessions and they are demanding, time-consuming, and costly. This might add to parents' burden as they often have to cope with the demands associated with taking care of a child with ASD, such as intensive educational, behavioral, and health services (Roberts & Ridley, 2004). Therefore, brief interventions need to be further examined to identify possible paths to help parents improve their strategies and FQoL efficiently in fewer sessions.

I.IV. Significance of the study

Given the effects of different parent interventions (such as RT, PRT, ABA), it would be beneficial to identify whether a brief intervention that combines these different approaches can influence parental skills and behavior and FQoL. In addition, it would be informative to evaluate treatment adherence. In this paper we report the results of such intervention.

In particular, we conducted a 6-week intervention focused on four components: (1) ASD knowledge; (2) language-communication management; (3) behavioral management; and (4) parental stress management. The primary aims for this study were to: (1) examine parents' behavior and whether the intervention's effect remains after the intervention; (2) examine if there is any change in any dimension related to the FQoL after the brief intervention; and (3) examine the treatment adherence and its relationship to outcomes.

II. Methods

II.I. Participants

A total number of 45 families from Granada (Spain) with a child with ASD (ranging from 2.00 to 7.00 years old) were enrolled in this study. To be included in the study, the diagnosis for each child should be based on the DSM-TR-IV (APA, 2000) or DSM-5 (APA, 2013) and the Autism Diagnostic Interview-Revised (ADI-R; Le Couteur, Lord, & Rutter, 2003) or the Autism Diagnostic Observation Schedule (ADOS-G; Lord, Rutter, DiLavore, & Risi, 2002). Moreover, the diagnosis was confirmed by an independent tester with the Guilliam Autism Rating Scale (GARS-2; Guilliam, 2006). An additional inclusion criterion to be eligible was that children had to be minimally verbal, confirmed by parent report and child performance. This status was defined as using less than 20 intelligible words and productive syntax (Brignell et al., 2016; Kasari, Brady, Lord, & Tager-Flusberg, 2013). Finally, to be eligible for recruitment, parents had to report that they were not participating in another parent-mediated intervention at that point in time.

All families were recruited from several parents' associations of children with ASD. The intent-to-treat analysis included 23 families in the treatment group while 20 families were included as a waitlist group (see figure 7.1 for TREND Flow Diagram). From 23 parents that were enrolled in the intervention, but 3 did not complete the intervention (they shifted from psychotherapy to pharmacologic therapy). Data from these families were excluded from the analyses. All families that met our inclusion criteria were enrolled consecutively.

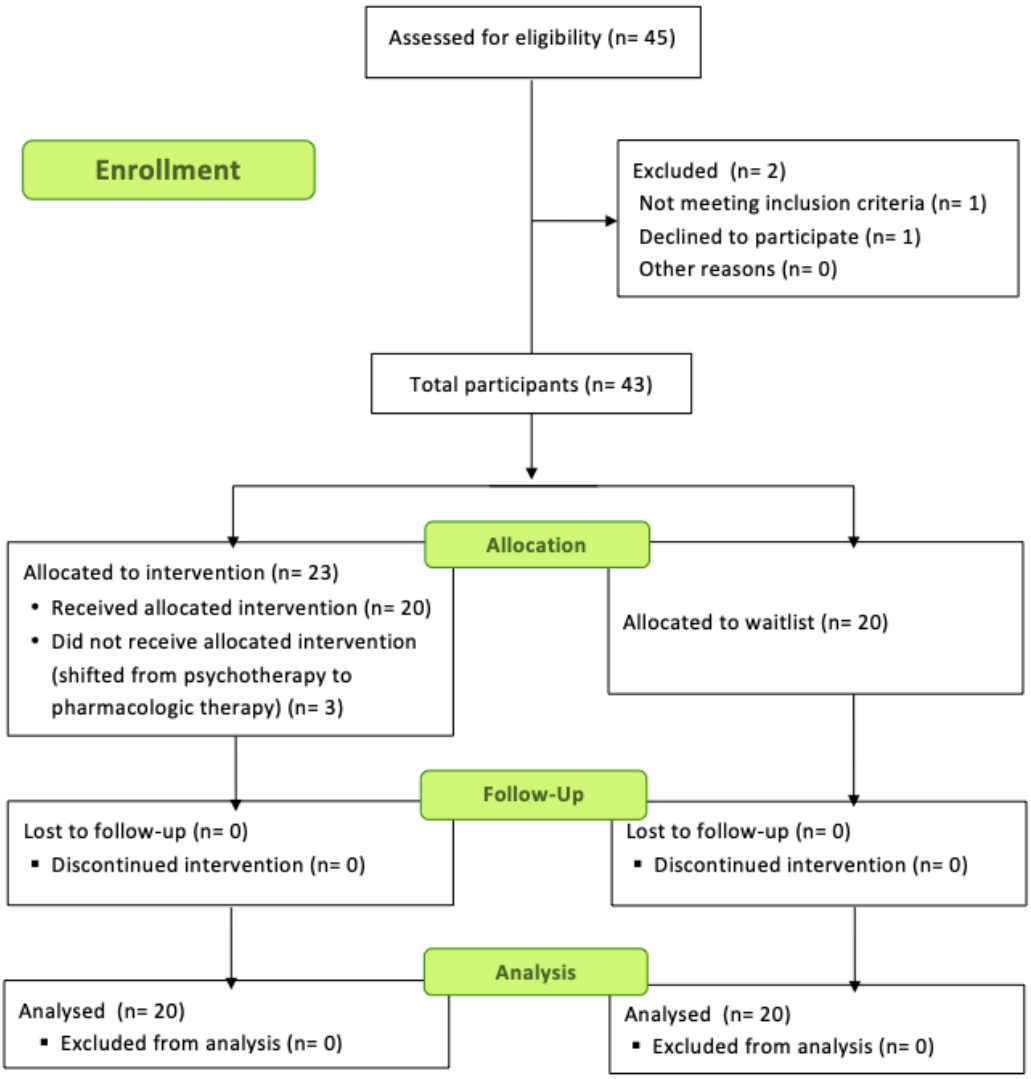

Figure 7.1. CONSORT flow diagram for participants in the intervention

II.II. Intervention

The intervention was based on mediated parent interventions approaches documented in the literature (Barlow & Lehman, 1996; Cooper et al., 2007; Koegel & Koegel, 2012; Mahoney & McDonald, 2007). Specifically, it aimed at increasing parent's satisfaction on their FQoL through building a trusting relationship by developing a stronger and more consistent communicative reciprocity, a supportive attention and motivation, a functional language, behavioral functioning, and stress control. The intervention uses a responsive and behavioral approach, incorporating adapted developmental strategies commonly employed across RT (Mahoney & McDonald, 2007), PRT (Koegel & Koegel, 2012), ABA-based behavioral

intervention (Attwood, 2003; Cooper et al., 2007), and cognitive behavior therapy (Barlow & Lehman, 1996). Specifically, the intervention included the following components: ASD knowledge (clarifying doubts and confronting some myths associated to ASD), language-communication management (e.g., responding to child's interaction attempts, appreciate what the child is doing, joining and following child's lead, imitating child's actions or vocalizations, and playing with child face to face), behavioral management (e.g., observing how the child behaves, responding immediately after the attempt/behavior, and comforting the child when is anxious, fussy, or angry), and parental stress management (e.g., reorganizing the negative emotions, and speaking with other about ASD). See Supplemental Material (S7.1) for a detailed description of these components.

Weekly 120-min sessions were provided for 6 weeks in several social centers. The therapist was a psychologist, specialized in ASD with more than 5 years of experience, who received in-depth training. A standardized format was used to conduct the sessions. The intervention was included in a short report and additional slides were provided as support material to the psychologist to ensure the implementation fidelity. The information related to each component, and written directions were available for parents (via email) after every session. In addition, we gave parents the option of discussing any questions or sharing their thoughts and improvements with the psychologist. Parent treatment adherence was measure through parent treatment attitudes and behavioral intention.

II.III. Measures

Measurement of FQoL. Participants completed the Family Quality of Life Survey (FQoLS; Verdugo, Rodriguez, & Sainz, 2009). We evaluated three factors related to FQoL (family interaction, emotional well–being, and the role as father/mother) in one domain: satisfaction. Moreover, we combined the three factors to come up with a global measure of FQoL. The scale showed a good internal consistency (Cronbach's = .78)

Knowledge about ASD. On seven–point scales ranging from 1 (strongly disagree) to 7 (strongly agree), parents evaluated their knowledge related to 4 components: ASD (e.g., "the ADS's origin is related to a vaccine"), language-communication management (e.g., "using visual aids [e.g., pictograms] facilitates and encourages communication with my child"),

behavioral management (e.g., “rewarding every achievement made by my child improves his/her behavior”), and parental stress management (e.g., “spending some time with my wife/husband and/or friends is a way to relieve stress”). This measure was included as an indicator of the knowledge that parents learnt after the intervention.

Self–report behavior. On a seven-point scale ranging from 1 (I never do it) to 7 (I always do it), parents evaluated their current behavior related to the following 3 components: language-communication management (e.g., “I use pictograms with my child with ASD to improve his/her communication”), behavioral management (e.g., “I reward every achievement that my child has manage to accomplish”), and parental stress management (e.g., “I spent some time on pleasurable activities”).

Treatment adherence. We measured parents’ attitudes towards the intervention (see also Hock et al., 2015, for a similar method) and their behavioral intention. In particular, they were asked about how important it was for them and their family to perform these behaviors, and how beneficial it was to perform them. Behavioral intentions were measured on a seven-point scale ranging from 1 (I have no intention to do it) to 7 (I am certain that I will do it). Using a seven-point scale, parents also indicated how likely it was that they would change their behavior.

II.IV. Procedure

All parents signed an informed consent form at the beginning of the study. The Ethics Committee of the University of Granada (Spain) approved the methodology of the study. The study had four phases (see Figure 7.2).

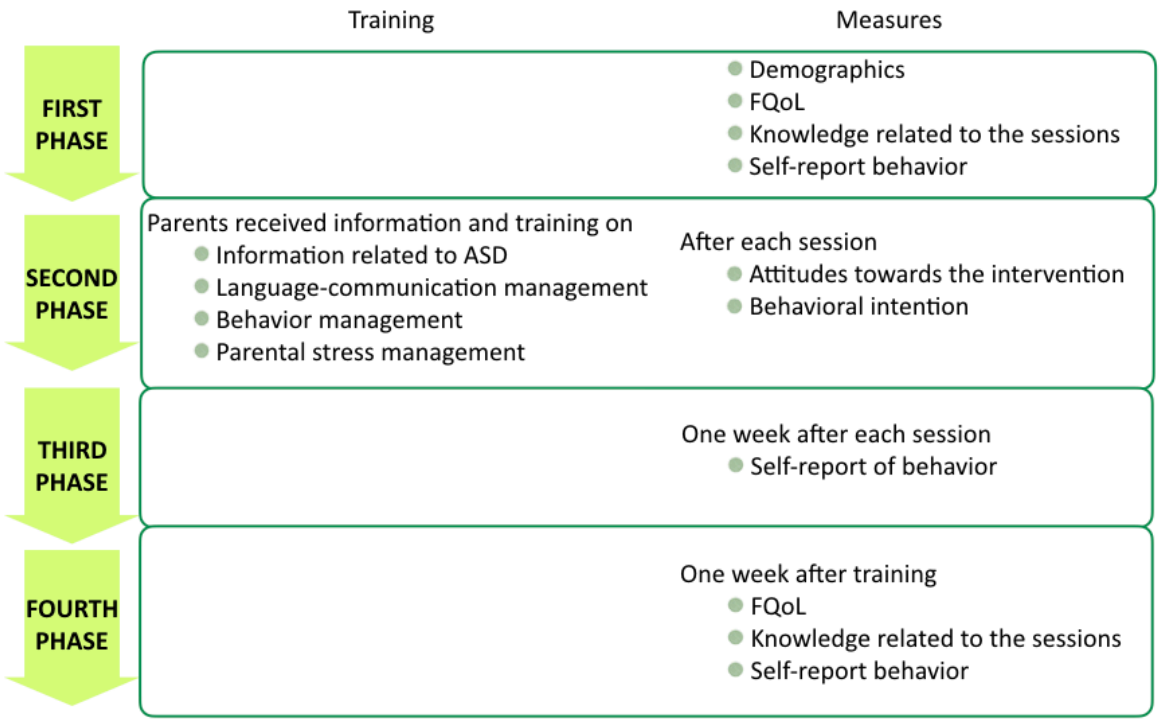

Figure 7.2. Design of the study showing the four phases and the variables measured in each phase.

In the first phase (i.e., baseline assessment, session 1), parents in the intervention and control groups completed several questionnaires, including standardized parent questionnaires related to child's autism traits (GARS-2; Guilliam, 2006), and additional scales that measured demographics, FQoL, the knowledge related to the sessions, and self-reported behavior. Parents completed the same questionnaire again in the third and fourth phase of the intervention (i.e., post-intervention and follow-up).

In the second phase (i.e., sessions 2, 3, 4, and 5), parents in the treatment group received information and a specific training about the following components: ASD, language-communication management, behavioral management, and parental stress management. These sessions were implemented in small groups (6 families on average). After each session, parents completed questionnaires evaluating the acquired knowledge, treatment adherence, and behavioral intentions.

In the third phase (i.e., post-intervention session) was conducted one week after the last session of the second phase session. In this session parents in the treatment group reported their behavior (self-reported behavior).

In the fourth phase (i.e., follow-up session), conducted one week after the last post-intervention assessment, parents in the treatment and control group completed the FQoL scale, and the measures of knowledge about ASD and self-report behavior. Afterwards parents in the control group were offered to participate in a full intervention to ensure that no potential benefits were denied.

II.V. Data Analyses

All statistical analyses were performed using SPSS 22.0 statistical software. To evaluate differences between groups, treatment versus control group effects were analyzed using general linear mixed models (GLMM). In our model, time and group, as well as its interaction were included as fixed effects. Random effects for the intercept and time were also considered and tested. The GLMMs were analyzed using restricted maximum likelihood estimation, which is widely used for longitudinal data (Brown & Prescott, 2006). To determine the optimal and most parsimonious model for the each dependent variable, we reduced the fixed effects once we determined which random effects were needed. Besides likelihood ratio chi-square test, Akaike Information Criterion (AIC), and Bayesian Information Criterion (BIC) were used to select the optimal models. If time by group interaction was significant, it was followed-up with simple slope comparison at post-intervention and the time at follow-up. Additionally, for all significant simple comparison Cohen's d were calculated as effect size.

III. Results

Forty-five families were evaluated to participate in this study. Two families were excluded because one did not meet the inclusion criteria (n = 1) or declined to participate (n = 1). All participants who finished the intervention (n = 20) returned for the follow-up session. The average age of the parents in this study was 35.03 years (SD = 4.10; range: 25-45). Overall, the sample was predominant comprised of mothers (n = 34; 85%), married (n = 25; 74%),

and highly educated (87.5% had completed college). Further, the average age of the children was 4.65 years (SD = 1.63; range: 2.00-7.00), 35 were male (87.5%), and the mean Autism Quotient (assessed using the GARS-2; Guilliam, 2006) was 92.13 (SD = 12.81; range: 80-145).

We compared demographics in the intervention and control groups at baseline. The analyses did not show significant differences in parents or children (ps > .05) (see Table 7.1). Descriptive statistics for the means and standard error means of FQoL, communication-language management, behavioral management, and parental stress management measures at baseline, post-intervention and follow-up are shown in Table 7.2. The results of the initial models were conducted using GLMMs are provide in S7.2, and the final models are provide in Table 7.3.

Table 7.1. Parents and children socio-demographic characteristics

Variables	Groups		*p*	Test statistic
	Treatment (n = 20)	Control (n = 20)		
Parent gender			.661	Fisher exact
Male	4 (20%)	2 (10%)	–	–
Female	16 (80%)	18 (90%)	–	–
Parent age	34.20 (4.29)	35.85 (3.83)	.492	*t* test = .48
Parent education			1	Fisher exact
College	18 (90%)	17 (85%)	–	–
Some college	2 (10%)	3 (15%)	–	–
High school	0 (0%)	0 (0%)	–	–
Marital status			1	Fisher exact
Married	15 (75%)	16 (80%)	–	–
Divorced	5 (25%)	4 (20%)	–	–
Single	0 (0%)	0 (0%)	–	–
Parent employment			.574	$\chi^2 = 1.11$, df = 2
Full time	1 (5%)	1 (5%)	–	–
Part time	4 (20%)	5 (25%)	–	–
No work outside home	15 (75%)	14 (70%)	–	–
Children's gender			1	Fisher exact
Male	18 (90%)	17 (85%)	–	–
Female	2 (10%)	3 (15%)	–	–
Children age	5.27 (.89)	4.85 (1.53)	.080	*t* test = 3.24
Children's IQ[a]	81.95 (12.16)	82.85 (11.85)	.814	*t* test = .23
Autism Quotient[b]	91.45 (15.54)	92.80 (9.71)	.362	*t* test = .85
Children's engaged in psychotherapy	18 (90%)	19 (95%)	.548	$\chi^2 = .36$, df = 1
Children's engaged in speech therapy	20 (100%)	20 (100%)	–	–

[a] Children's IQ was measure with the Leiter International Performance Scale-Revised (Leiter-R; Roid & Miller, 2011).

[b] Autism Quotient was measure with the Guilliam Autistic Rating Scale (GARS; Guilliam, 2006)

Table 7.2. Group comparisons of parents' variables between baseline, post-intervention, and follow-up.

	Intervention group						Waitlist group					
	Baseline		Post-Intervention		Follow-up		Baseline		Post-Intervention		Follow-up	
	Mean	SEM	Mean	SEM	Mean	SEM	Mean	SEM	Mean	SEM	Mean	SEM
Language-communication management												
Self-report behavior	4.57	.304	4.96	.307	5.50	.218	4.90	.198	4.89	.224	4.80	.189
Behavioral management												
Self-report behavior	5.35	.202	5.80	.088	5.53	.129	5.75	.106	5.73	.105	5.71	.112
Parental Stress management												
Self-report behavior	4.26	.238	5.68	.101	4.71	.209	4.63	.217	4.61	.220	4.61	.246
Family Quality of Life												
Family interaction	4.23	.093	4.52	.107	–	–	3.99	.101	3.92	.135	–	–
Emotional well-being	3.46	.157	3.68	.131	–	–	3.07	.172	3.00	.157	–	–
Parent's role	3.97	.136	4.54	.092	–	–	4.11	.104	4.14	.156	–	–

Table 7.3. Parameter Estimates for final model.

	Language-Communication management	Behavioral management	Parental Stress management	Family Quality of Life			
				Family interaction	Emotional well-being	Parent's role	Total FQoL
Fixed effects							
Intercept	4.837*	5.641*	2.563*	3.951*	3.101*	3.733*	3.667*
Time	-	-	2.120*	-	-	.231 [a]	-
Time Sq	-	-	-.506*	-	-	-	-
Group x Time	-.464	-	.704*	.283*	.269*	.144 [a]	.290*
Group x Time Sq	.241*	.002	-.169	-	-	-	-
Random effects							
Intercept	.297*	.184*	.414*	.115*	.120*	.281*	.077
Level 1 variance	.869*	.165*	.571*	.124*	.358*	.038	.086
Overall Model Fit							
AIC	295.274	201.396	312.564	109.149	142.968	145.328	79.046
BIC	300.798	206.938	318.053	113.862	147.681	150.015	83.759

Note: AIC = Akaike Information Criterion, BIC = Bayesian Information Criterion. Lower scores at AIC and BIC mean the model fits better.
[a] = marginally significant; * = <.05

III.I. Language-communication management

The intervention group showed a different pattern of results in language-communication responses through post-intervention and follow-up [F(1, 102) = 7.689, *p* = .007]. Simple slopes comparisons (see Table 7.4) revealed that although the differences between groups did not approach significance at post-intervention (*p* > .05), the intervention group showed more language-communication management responses than the control group at follow-up (*p* = .013, d = .83). Predicted values are shown in Figure 7.3.

III.II. Behavioral management

There was not a significant effect of group by time, indicating that the intervention group did not show a different pattern of results in the intervention and follow-up sessions. Predicted values are shown in Figure 7.3.

III.III. Parental stress management

There was an overall effect of group by time [F(1, 205) = 4.373, *p* = .038], indicating that there were no differences between groups at baseline, but the intervention and the control groups differed in parental stress management in the post-intervention and follow-up sessions. Simple slopes comparisons (see Table 7.4) revealed that parents in the intervention group showed significantly more adequate parental stress management than those in the control group at post-intervention (*p* = .039, d = .68). However, differences between groups were not significant at follow-up (*p* > .05). Predicted values are shown in Figure 7.3.

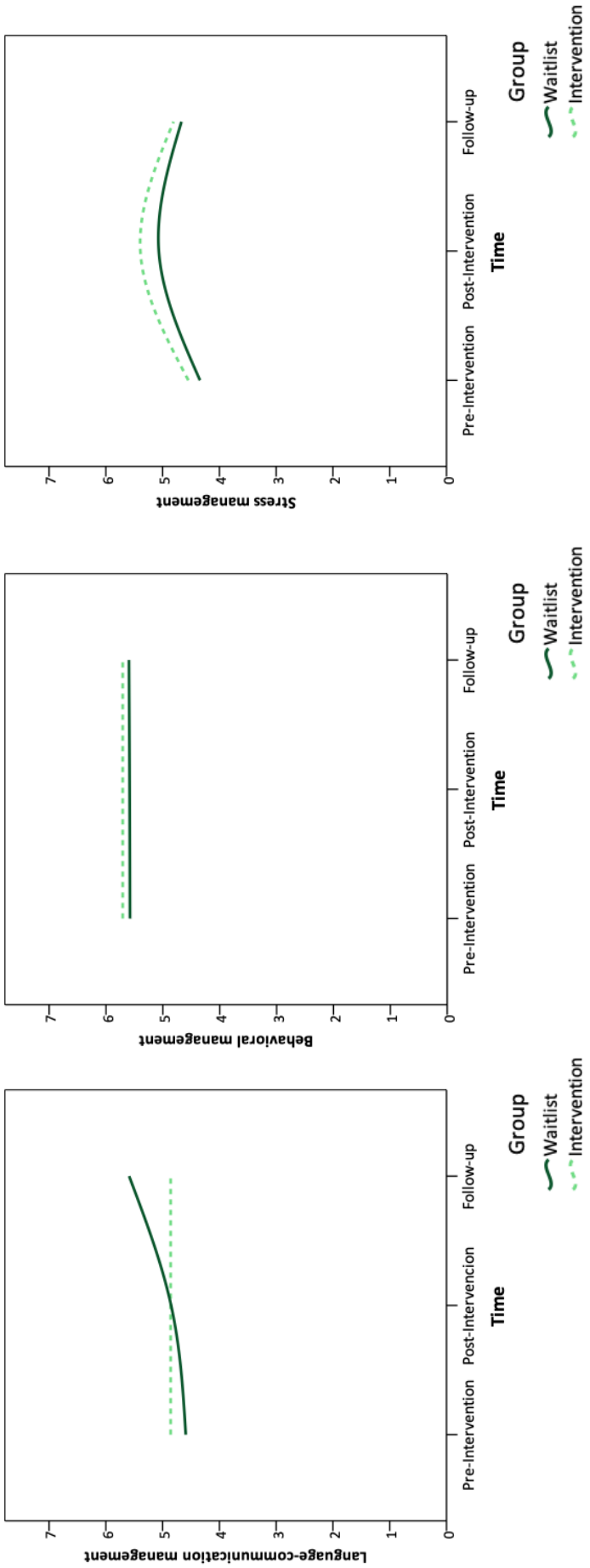

Figure 7.3. Differences on parental behavior (baseline, post-intervention and follow-up) on language-communication management, behavioral management, and parental stress.

III.IV. Family Quality of Life

There was an overall effect of group by time in family interaction [F(1, 75) = 16.881, $p < .001$], emotional well-being [F(1, 75) = 9.889, $p = .002$], and the global score of FQoL [F(1, 75) = 26.018, $p < .001$], indicating that there were no differences between groups at baseline, but parents in the intervention and control groups differed in their satisfaction on FQoL in the post-intervention session. There was a marginal effect of group by time in parent's role [F(1, 48) = 3.007, $p = .089$]. Simple slopes comparisons (see Table 7.4) revealed that parents in the intervention group showed significantly more family interactions ($p < .001$, d = 1.92), emotional well-being ($p = .001$, d = 1.17), parent's role ($p < .001$, d = 2.99), and global measure of FQoL ($p < .001$, d = 2.18) than those in the control group at post-intervention. Predicted values are shown in Figure 7.4.

Table 7.4. Simple comparisons between groups at posttest and follow-up.

Variable	Time	Estimat.	SE	t	*p*
Language-communication management	Post-Intervention	-.005	.280	-.029	.977
	Follow-up	.390	.280	2.611	.013
Behavioral management	Post-Intervention	-.178	.109	-1.112	.273
	Follow-up	-.161	.109	-1.007	.320
Parental stress management	Post-Intervention	.327	.210	2.136	.039
	Follow-up	.230	.210	1.454	.154
Family Quality of Life					
Family interaction	Post-Intervention	.701	.092	6.064	<.001
Emotional well-being	Post-Intervention	.515	.175	3.699	.001
Parent's role	Post-Intervention	.839	.029	9.492	<.001
Total FQoL	Post-Intervention	.746	.077	6.897	<.001

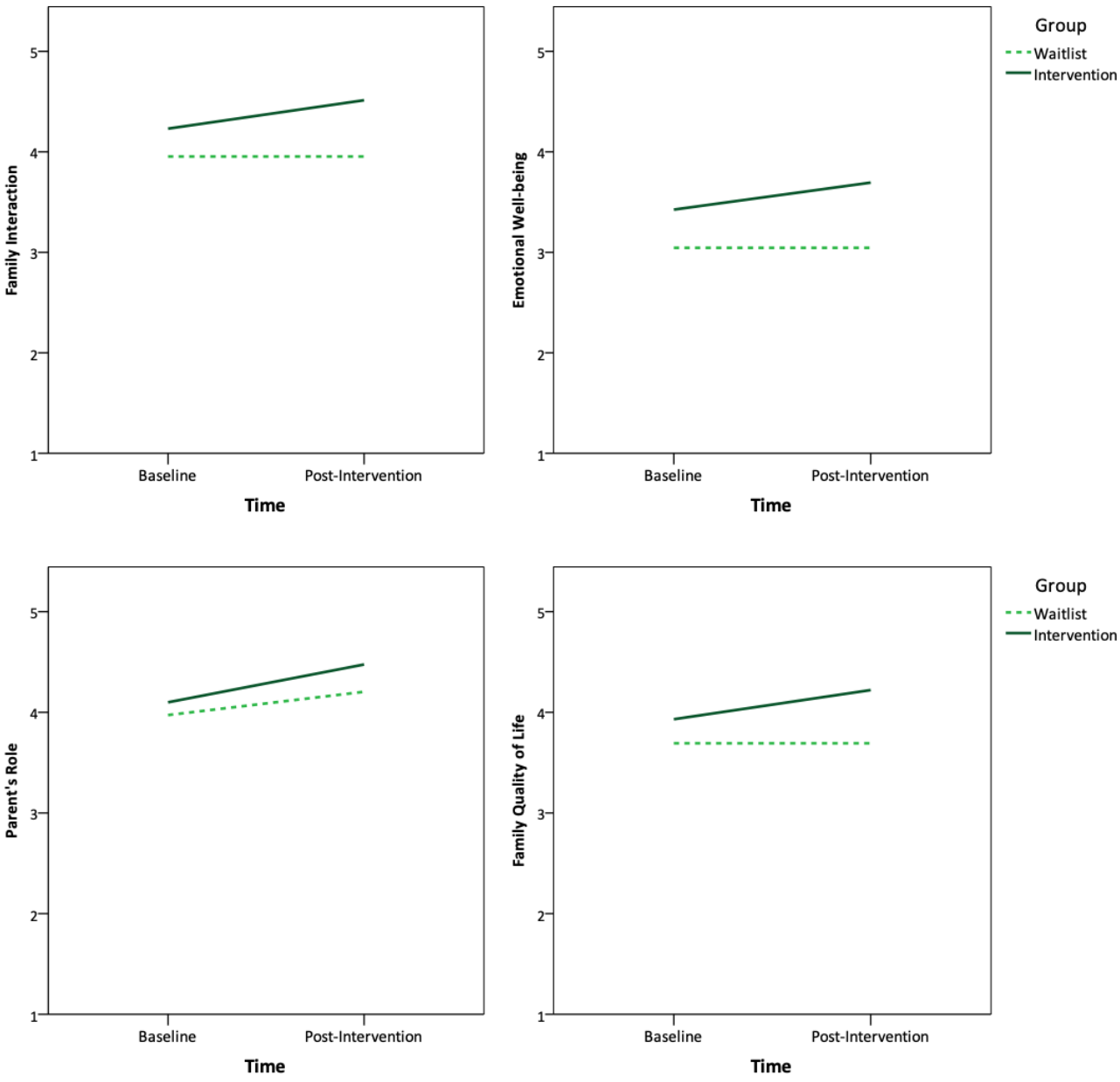

Figure 7.4. Differences between baseline and post-intervention on FQoL

III.V. Treatment adherence

We analyzed bivariate correlations between parents' attitudes and behavioral intentions and between behavioral intentions and the currently change in parental strategies. Results revealed a similar pattern of results in language-communication strategies and behavioral management. Specifically, parents with higher positive attitudes showed higher behavioral intentions (r = .720, p < .001 for language-communication, and r = .485, p = .030 for

behavioral management). Moreover, parents with higher behavioral intentions showed larger improvements in language-communication and behavioral management strategies (r = .819, p < .001 and r = .659, p = .002 respectively). Regarding stress management, results showed that parents with higher positive attitudes showed higher behavioral intentions (r = .690, p < .001). However, correlations were not significant between behavioral intentions and stress management (r = .141, p = .554).

IV. Discussion

This study investigated the effect of a brief parent intervention on parental management strategies related to language-communication, behavior, and parental stress, and FQoL. In contrast to parents in a control group, parents who received the intervention improved their language-communication, and stress management strategies, and their satisfaction on FQoL. However, the intervention did not influence behavioral management.

Improvements in parental language-communication strategies were described in the intervention group from baseline to follow-up. This improvement showed a large effect size, reflecting significant progress in one of the most critical aspects in ASD. Within the language-communication management, the mean number of strategies used by parents in the intervention group increased from baseline to follow-up. While the largest gain was observed between baseline and follow-up, a positive trend was also observed between baseline and post-intervention. This improvement was also consistent with prior research, which informed that parents were able to develop strategies related to language and communication skills (Matson et al., 2009). Significant findings in language-communication strategies are promising given the length of the intervention. Therefore, this study supports the idea that parents can benefit from a brief intervention and that they might be able to learn strategies to help their children.

Surprisingly, the intervention group did not show significant improvements in their behavioral management strategies after the intervention compared to the control group. These results could be explained because some strategies require more time to show differential changes (Iadarola et al., 2017). A potential explanation might be that although the ABA-based behavioral treatment has shown potential benefits, this approach often

requires a massive number of trials, which could interfere with families' daily routine (Dillenburger et al., 2004).

Regarding parental stress management, the intervention group showed an increased number of strategies intervention compared to the control group. However, the results of the model showed that the intervention effect was attenuated from a medium effect size at post-intervention to follow-up. This might be in part due to the parents' effort to become more conscious about the importance and need of stress control, although they could not manage it for longer, rather because of the daily burden they have to cope with (Roberts & Ridley, 2004).

Overall, parents in the intervention group showed improvements in their levels of satisfaction on FQoL. Specifically, intervention group showed gains on satisfaction on FQoL in two of the three different domains measured: family interaction, and emotional well-being, compared to the control group. Moreover, improvements in both domains showed large effect sizes. These improvements could be related to their parental competence in language-communication strategies, their self-efficacy, and confidence after the intervention (Ayuda-Pascual et al., 2012; Feinberg et al., 2014; Iadarola et al., 2017). On the other hand, improvements in intervention group related to parent's role were marginally significant compared to control group.

Fidelity of implementation and treatment adherence were addressed in this study. Particularly, fidelity of implementation was considered through the trainer experience and education. Moreover, written directions and information related to the sessions were available for parents. Treatment adherence was tested through a questionnaire, which evaluated parent intervention attitude and their behavioral intentions. Our results revealed that those parents who evaluated the information as more interesting and more important had the intention to change their behavior in language-communication and behavioral management. Moreover, parents who had the intention to change were those who showed bigger changes in their behavior in language-communication and behavioral management. Our results are consistent with the planned behavior theory (Ajzen & Fishbein, 2005). Surprisingly, this effect did not appear in parental stress. This fact could be explained by the non-significant differences between baseline and follow-up in this component. It seems that

intervention attitude and behavioral intentions could be used as an indicator of treatment adherence as other authors stated previously (see Hock et al., 2015).

These findings contribute to the growing evidence of brief parent interventions. In fact, results from our study suggest that parents of children with ASD are capable to acquire some strategies and some of them might be maintained. This is in line with previous research which supports that parents can learn different strategies and skills (Kasari et al., 2015; Matson et al., 2009). Moreover, including parents as part of the treatment of children with ASD can be highly beneficial. This argument rests on the fact that parents spend a significant amount of time with their children and therefore might be able to implement these strategies throughout the day.

The present findings should be considered in light of several limitations. The sample size was small and we did not include a long term follow-up. For instance, due to our sample size, we did not tested predictions in our analyses. Future investigations with a larger number of participants should take into consideration a long term follow-up, in order to determine the durability of the intervention. Moreover, although FQoL is roughly stable over the time, it was only assessed at baseline and at post-intervention. Future studies should evaluate the FQoL at multiple time points. Finally, the reported effects and outcomes ought to be read as preliminary findings and future research is needed to further determine the effects of this intervention.

This study reveals the potential of a brief and specific intervention that appears to be particularly effective with families of children with ASD. The practical applications of these findings are that parents could learn useful strategies through short-term interventions. This may have a positive transactional effect on the motivation of the parents involved in their children's interventions. Overall, the present study provides a step towards identifying brief interventions that may be especially promising for research on quality of life for families of children with ASD.

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Table S7.1. Intervention content overview

Topic	Key ideas	Content
ASD	• Oveview of program • Knowledge of autism • Myths of autism • Treatments of autism	• The origins of autism, biological and environmental causes • Myths (e.g., Is there any relationship between ASD and vaccines?; Is there any relationship between some symptoms related to ASD and some kinds of diet?) • ASD's features.
Language-communication management	• Functional language • Imitation/responses • Opportunities for language and interactions • Receptive language: Communication for understanding • Non-verbal communication	• Appreciate what your child is doing (attempts at interacting with objects and others) • Imitate your child's actions or vocalizations • Quick responses to your child's communication acts • Match your message with intonation and nonverbal language • Quiet standby, waiting for your child's response • Engage parallel play (play along side others without interacting) • Play with your child face to face • Reformulate approximations or unclear vocalizations.
Behavioral management	• Oveview of program • Applied behavior analysis • How to calm a child with autism	• Watch how your child behaves • Quick responses to your child's behavior • Differential reinforcement After any conduct, a consequence must appear (contingency) • Comfort your child when he or she is anxious, fussy, or angry.
Parental stress management	• Stress in families of children with autism • Reorganizing the negative emotions • Speaking with other about autism (family, professionals…)	• Experience your emotions vs allow emotions to rule your life • Recognize your negative emotions in order to combat them • The more number of negative thoughts, the more pain you feel • You are making all you can do it for your child • You should worry about something vs you should take care.

Table S7.2. Parameter estimates for initial model

	Language-Communication management	Behavioral management	Parental Stress management	Family Quality of Life			
				Family interaction	Emotional well-being	Parent's role	Total FQoL
Fixed effects							
Intercept	4.744*	4.995*	2.563*	4.002*	3.196*	3.733*	3.644*
Time	.192	.795*	2.120*	-.043	-.091	.231 [a]	.020
Time Sq	-.059	-.194*	-.506*	-	-	-	-
Group x Time	-.567	-.113	.704*	.301*	. 324*	.144 [a]	.281*
Group x Time Sq	.279	.0337	-.169	-	-	-	-
Random effects							
Intercept	.302*	.179*	.414*	.117*	.121*	.281*	.079*
Level 1 variance	.869*	.165*	.571*	.124*	.355*	.038	.085*
Overall Model Fit							
AIC	300.378	204.306	312.564	111.975	144.915	145.328	82.437
BIC	305.868	209.796	318.053	116.662	149.602	150.015	87.124

Note: AIC = Akaike Information Criterion, BIC = Bayesian Information Criterion. Lower scores at AIC and BIC mean the model fits better.
[a] = marginally significant; * = <.05

CHAPTER 8

Chapter 8

Parents' numeracy buffers the negative effect of ASD on family quality of life

The content of this chapter has been published as Garrido, D., Petrova, D., Cokely, E., Carballo, G., & Garcia-Retamero, R. (under review). Parental numeracy may be associated with higher quality of life in families with a child with autism spectrum disorder

Parental numeracy may be associated with higher quality of life in families with a child with autism spectrum disorder

Families of children with Autism Spectrum Disorder (ASD) are more likely to express relatively negative perceptions of their family quality of life (FQoL). To explore factors that may help explain these experiences, we conducted the first investigation of the relationship between FQoL and parental risk literacy (i.e., the ability to interpret and evaluate risk, as measured by statistical numeracy tests). The study utilized a case-control design involving sixty-one Spanish families (29 with a child with ASD) that completed a broad assessment of psychological and sociological factors (e.g., child and parental' ages, genders, levels of education, parental' employment status, marital status, severity of ASD, FQoL, social support, and objective and subjective numeracy). Statistical modeling of results indicated that, independent of all other assessed factors, objective—but not subjective—numeracy may generally be associated with relatively large differences in perceived FQoL among families with a child with ASD ($r = .52$). As compared to families with typically developing children, in families with a child with ASD, lower parental numeracy (< 33th percentile) was associated with about 3 times greater reduction in FQoL than was expressed by families with higher parental numeracy (≥66th percentile). The discovery of a relationship between FQoL and parental numeracy suggests that parental risk literacy may often be associated with factors that help buffer against negative outcomes related to perceived FQoL (e.g., risk identification, communication, and anticipation). Discussion includes considerations of study limitations and presents implications for future research on causes, consequences, and potential interventions.

I. Introduction

Autism Spectrum Disorder (ASD) is generally classified as a complex neurodevelopmental disorder characterized by social-communication challenges and restricted and repetitive behaviors (APA, 2013). Perhaps because of these unique characteristics and associated

challenges, parents of children with this disorder tend to show higher levels of anxiety and stress compared to parents of children with typical development (TD) (see Vasilopoulou & Nisbet, 2016 for a recent systematic review). Families with children with ASD are also at a higher risk of experiencing physical and psychological distress, and these families also tend to report significantly lower levels of family quality of life on standardized psychometric assessments (FQoL) (Allik, Larsson, & Smedje, 2006; Garrido, Carballo, Franco & Garcia-Retamero, 2015; Mannan, Summers, Turnbull, & Poston, 2006). To date, most studies investigating factors that affect FQoL in children with disabilities such as ASD have focused on negative experiences and perceptions (Hastings & Taunt, 2002), although some researchers have more recently begun to examine protective factors (Vasilopoulou & Nisbet, 2016). One such line of inquiry has revealed that social support may improve FQoL in families of children with ASD (Khanna et al., 2011; McStay, Trembath, & Dissanayake, 2014). Theoretically, social support may promote better mental health and more positive perceptions for many reasons, including enhanced capacity to evaluate and cope with complex decision making (Khanna et al., 2011; Kuhlthau et al., 2014). Might other factors that improve decision quality, such as parental levels of risk literacy (i.e., one's ability to interpret and evaluate risk), also generally benefit families with a child with ASD?

Previous research suggests that individual differences in cognitive skills, particularly those related to risk literacy (i.e., acquired skills for accurate interpretation and evaluation of risk; see RiskLiteracy.org) often help people overcome some of the demands associated with complex decision making in a health context. Although it stands to reason that the information overload experienced by families with a child with ASD would likely be less among those who are more fluent in the language of risk, currently we know of no study that has investigated the role of parental risk literacy among families with a child with ASD. It is well-established that ASD includes heterogeneous symptoms, and as such families of children with ASD often receive a wide-range of information and must make many decisions based on complex information available from physicians, websites, support groups, professional societies, research articles, and clinical recommendations, to name a few (Romanczyk & Gillis, 2005). In addition, parents often have to process and integrate this information to make health-relevant decisions for their children or themselves, and often need to assess potential risks and consequences of even routine policies and choices

(e.g., risks of changing a bedtime routine, breakfast cereal, or after-school routine). Unfortunately, this information can be overwhelming, confusing, and contradictory (Grant, Rodger, & Hoffmann, 2015; Webster, Cumming, & Rowland, 2016), which is likely to be confusing and may complicate decisions regardless of the availability of other protective factors (e.g., social support).

Numeracy, or people's ability to accurately understand and use information about risk, is one of the most predictive individual differences in decision making about health, it is essential for risk literacy (Cokely et al., 2018; Cokely, Ghazal, & Garcia-Retamero, 2014; Peters, 2012; Reyna, Nelson, Han, & Dieckmann, 2009), and it is also a reliable predictor of relevant health outcomes (e.g., whether people suffer several serious diseases and seek for medical attention when they need it) (Garcia-Retamero, Andrade, Sharit, & Ruiz, 2015; Petrova et al., 2017; Petrova, Kostopoulou, Delaney, Cokely, & Garcia-Retamero, 2018). In the current research, we investigated to what extent parents' levels of numerical skills in families with a child with ASD predict their FQoL. We also compared perceptions of FQoL in these families with those in families with a child with TD; we investigated the effect of both objective numeracy (i.e., actual individual differences) and subjective (or self-reported) numeracy; and we compared the predictive power of these skills with that of social support.

II. Method

II.I. Participants

Participants were recruited at two schools (controls) and two associations of parents of children with ASD (cases) in Granada, Spain. Sixty-five parents were approached to participate in our study. All parents signed an informed written consent before participation. The inclusion criteria for participants in the ASD group were to have a child with ASD according to (1) the DSM-TR-IV (APA, 2000) or DSM-5 (APA, 2013) and (2) ADI-R (Le Couteur, Lord, & Rutter, 2003) or ADOS-G (Lord, Rutter, DiLavore, & Risi, 2002). The inclusion criteria for participants in the TD group were that their children did not have any disorder (e.g., attention deficit hyperactivity disorder, Down syndrome, or cerebral palsy)

or a previous family history of ASD. The Ethics Committee of the University of Granada approved the methodology of the study.

II.II. Measures

Participants completed a demographic survey developed for the current study, which included child and parents' age, gender, level of education, employment status, and family composition. Additional specific measures related to severity of ASD, social support, numeracy skills, and FQoL were also collected, as described below.

Severity of ASD. Parents completed the Guilliam Autism Rating Scale (GARS; Guilliam, 2004). This scale is a norm referenced screening instrument that helps professionals identify ASD. This scale gathers information about specific characteristics typically noted in ASD (stereotyped behaviors, communication, and social interaction). We used this measure as severity of ASD.

Social support. We evaluated social support with the Structural Social Support Questionnaire (Berkman & Syme, 1979), including parents' number of friends, perceptions of quality of their relationship with relatives, and number of weekly contacts that parents have with their relatives and friends. We computed the average of parents' answers in these items. This questionnaire showed good internal consistency, with a Cronbach's alpha score of 0.71.

Objective Numeracy. Objective numeracy was measured with a scale of 13 items, consisting of 9 items developed by Lipkus, Samsa, and Rimer (2001) as well as the 4 items of the Berlin Numeracy Test (Cokely, Galesic, Schulz, Ghazal, & Garcia-Retamero, 2012). The scale assesses the ability to compare risk magnitudes, convert percentages to proportions, convert proportions to percentages, convert probabilities to proportions, and compute probabilities. The scale showed adequate internal consistency, with Cronbach's alpha score of 0.85. Tests of criterion validity have indicated that scores in the questionnaire are highly correlated with correct answers to ecologically valid probabilistic medical decisions (Cokely et al., 2018; Garcia-Retamero, Cokely, Ghazal, & Joeris, 2016).

Subjective numeracy. It was measured with a scale developed by Fagerlin et al., (2007), which is an 8-item self-report measure of the perceived ability to perform various mathematical tasks and preferences for use of numerical versus prose information. The scale demonstrates good reliability (Cronbach's alpha = 0.82) and is significantly correlated with the objective numeracy scale (r = 0.63–0.6856; Galesic & Garcia-Retamero, 2010).

Family quality of life. Participants completed the Family Quality of Life of People Survey (FQoLS; Verdugo, Rodriguez, & Sainz, 2009). This instrument evaluates FQoL in two domains (importance and satisfaction), and it includes two global scores (i.e., importance of FQoL and satisfaction with FQoL) derived from five factors related to FQoL: emotional wellbeing, family interaction, financial resources, the role as father/mother, and physical wellbeing.

II.III. Data Analysis

All statistical analyses were performed using IBM SPSS statistics version 22.0, and in the R statistical environment. We computed descriptive statistics to characterize the sample of participants. To test whether group (family with a child with ASD vs. TD), levels of objective and subjective numeracy, and social support predict FQoL, we fitted a linear regression model, controlling for the effect of two variables that could have a significant impact on FQoL (i.e., severity of ASD and level of education). To investigate whether the relationships of numeracy and social support with FQoL varied as a function of group, we tested the moderation effect of numeracy and social support and their interaction with group using the PROCESS macro, embedded and operated in SPSS (Hayes, 2013). For the current analysis, we selected PROCESS Model 1 for moderation. Given the limited sample size, and to prevent violation of normal distribution assumptions, 5.000 bootstrap samples were drawn to provide a robust estimation of direct and indirect effects (Erceg-Hurn & Mirosevich, 2008).

To further understand the moderation effects, we estimated effects at the sample mean, and plus/minus one standard deviation of the value of the moderator. A sensitivity analysis (using G*Power calculator; Faul, Erdfelder, Lang, & Buchner, 2007) showed that given seven predictors (6 main effects and one interaction), power=.80, and alpha=.05, the

minimum effect size that could be detected with the obtained sample size of n = 61 is *f*=.104 (Cohen's d=.25).

III. Results

From all parents (n = 65) that were invited, 61 (94%) agreed to participate in the study. The final sample included 29 parents of a child with ASD and 32 parents of a child with TD. Most participants were mothers (93%) with an average age of 41.5 years (ranging from 31 to 58 years). We conducted preliminary analyses to examine socio-demographic characteristics and equivalences in our variables of interest in the groups of parent with a child with ASD and TD, respectively. Independent t-test and chi square analyses were computed for all measures, potential moderators, and socio-demographic characteristics (see Table 8.1). Analyses revealed only two significant differences between groups, namely the levels of satisfaction with their FQoL ($p < .001$), and severity of ASD symptoms ($p < .001$), suggesting that groups were otherwise similar by level of education, objective numeracy, and subjective numeracy, or availability of social support (i.e., number of friends, and number of relatives with whom participants frequently contacted).

Table 8.1. Descriptive analysis of ASD and TD groups.

		Group		Analysis		
		ASD (n = 29)	TD (n = 32)	Coeff	*p*	Effect size
Age		43.00 (6.53)	40.28 (4.74)	3.51	.066	.06
Gender				1.29	.255	.29
Male		3	1	–	–	–
Female		26	31	–	–	–
Level of education				4.25	.374	.23
Basic level		8	7	–	–	–
Upper secondary level		10	17	–	–	–
Bachelor's degree		10	8	–	–	–
Doctorate degree		1	0	–	–	–
Marital status				6.56	.87	.45
Single		1	3	–	–	–
Married		23	29	–	–	–
Divorced		5	0	–	–	–
Widowed		0	0	–	–	–
Employment status				7.61	.055	.51
Full-time		11	7	–	–	–
Part-time		9	6	–	–	–
Unemployed		9	19	–	–	–
Severity of ASD		33.59 (8.36)	11.66 (8.53)	102.53*	<.001	.64
Social support		6.44 (1.41)	7.47 (2.19)	3.09	.084	.05
Numeracy	Subjective	7.10 (3.74)	7.84 (2.84)	.58	.449	.01
	Objective	25.07 (7.82)	23.41 (9.09)	.77	.385	.01
FQoL	Importance	18.69 (2.04)	19.34 (1.29)	2.29	.135	.04
	Satisfaction	13.45 (3.49)	18.34 (2.19)	443.87*	<.001	.43

Note: Cohen's d and eta squared were calculated as measures of effect size (categorical and continuous variables respectively).

* = $p < .001$

We conducted a Pearson's correlation to examine the relation between subjective and objective numeracy. Results showed that these two variables were highly correlated (r = .38). To avoid multicollinearity in multiple regression analyses, we ran two separate regressions to identify which of these variables was a more reliable predictor of satisfaction with FQoL. Results showed that in our data, objective numeracy had a predictive value for FQoL ($F(1, 59) = 6.67$, $p = .01$, $R^2 = .10$, R^2 adjusted = .09) but subjective numeracy did not ($F(1, 59) = .20$, $p = .660$, $R^2 = .003$, R^2 adjusted = -.01).

The multiple linear regression analysis showed that only objective numeracy. ($\beta = .24$, $p < .05$) and group ($\beta = -.67$, $p < .001$) had a significant main effect on FQoL. We tested whether the relationship between objective numeracy and FQoL varied as a function of group by adding their interaction. The test of moderation including the interaction between objective numeracy and group are presented in Table 8.2, including standardized regression coefficients (βs) for each predictor. Results showed that the interaction between group and objective numeracy was significant ($\beta = .59$, $p < .05$). The complete model indicated that in the context of all variables, these two factors explained the majority of differences observed in FQoL (54% of the total variance, $F(7, 53) = 8.958$, $p < .001$).

Table 8.2. Linear regression model. Dependent variables: Satisfaction with FQoL.

	β	t	*p*	95% CI	
				LLCI	ULCI
R^2 = .49, MSE = 2.75					
Group	-.63	-6.60	<.001	-6.11	-3.27
Objective numeracy	.25	2.62	.110	.07	.50
R^2 = .53, MSE = 2.64					
Group	-1.14	-4.85	<.001	15.75	21.33
Objective numeracy	-.02	-.15	.881	-12.04	-5.00
Group x Objective Numeracy	.59	2.37	.210	.08	.94
R^2 = .54, MSE = 2.72					
Group	–1.22	–4.28	<.001	-13.54	-4.87
Level of education	.04	.45	.656	-.49	.77
Social support	.07	.66	.514	-.27	.54
Severity of ASD	.05	.32	.752	-.07	.10
Objective numeracy	-.08	-.49	.627	-.48	.29
Subjective numeracy	.06	.56	.577	-.07	.12
Group x Objective Numeracy	.65	2.39	.200	.09	1.03

Despite the lack of significant effects of subjective numeracy and social support in the multiple regression model, we checked whether there were similar interaction between group and these variables on FQoL. Thus, we also tested additional models that included the interactions between group and subjective numeracy and between group and social support. Results showed that these models failed to improve predictive power, and did not reach levels required to indicate statistically significant associations (β = −.49, p = .154, and β = .35, p = .411 respectively).

Plots of interactions comparing the effect of objective numeracy on FQoL are shown in Figure 8.1. Results indicate that FQoL was particularly low in families with a child with ASD whose parents had relatively low objective numeracy (about 3 times larger among low versus high parental numeracy groups). In contrast, FQoL was higher in families with a child with

ASD whose parents have relatively high objective numeracy and in families with a child with TD, regardless of their parents' levels of objective numeracy. In addition, there was a main effect of group such that FQoL was lower on average for families with a child with ASD, regardless of their level of objective numeracy. In families of children with ASD, parents with lower parental numeracy (< 33th percentile) showed 3 times greater reduction in FQoL than parents with high numeracy (> 65th percentile)–a result that is in contrast with that in families of children with TD, who did not show differences in FQoL as a function of numeracy (see figure 8.1)–. To further understand these results, we compared differences between groups in FQoL as a function of level of numeracy.

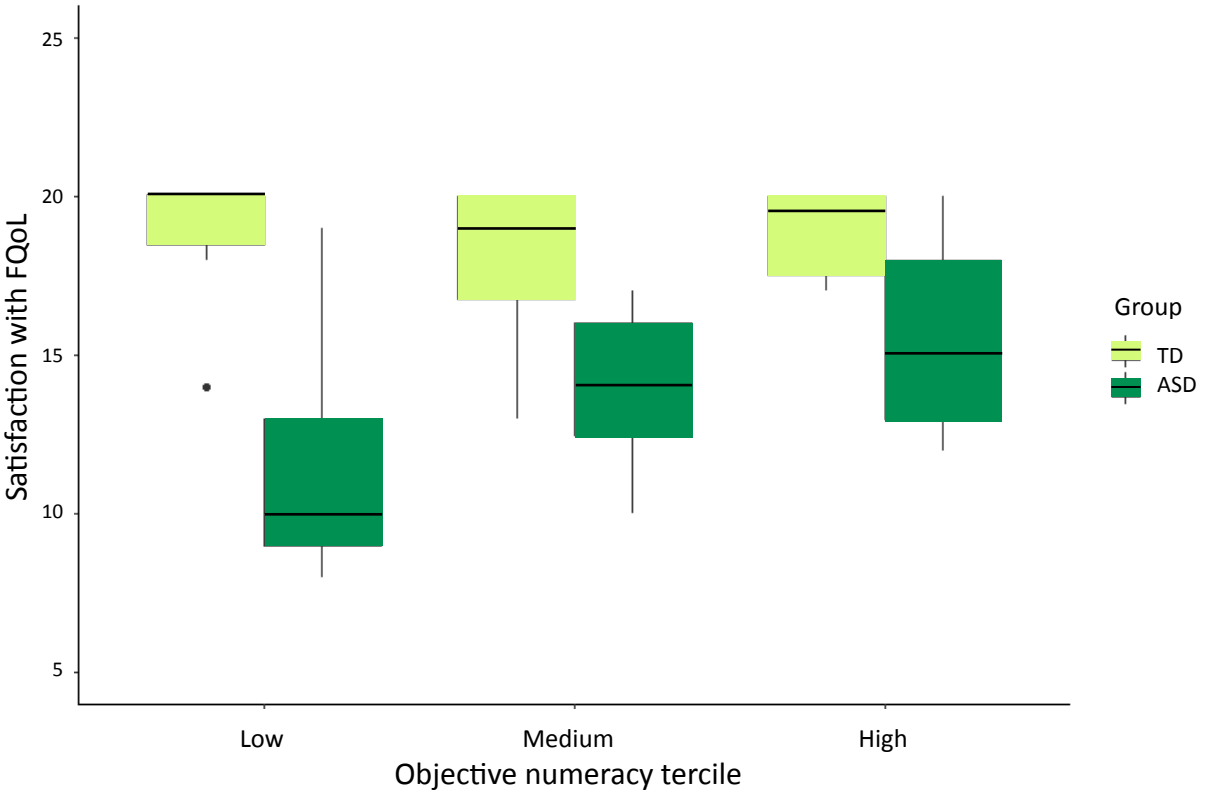

Figure 8.1. A visual representation of the moderation effect of numeracy (based on terciles) on satisfaction with FQoL by group.

Calculation of simple slopes (see Table 8.3) indicated that the effect of group on FQoL was statistically different both at the mean [F(1, 57) = 47.696, $p < .001$], and minus one standard deviation from the mean [F(1, 57) = 41.542, $p < .001$] on objective numeracy (a medium effect size of d = .66, and d = .68, respectively). Of note, although the effects of group on FQoL was statistically different at plus one standard deviation from the mean [F(1, 57) = 9.825, $p = .003$], the effect size was nominally smaller (d = .46). To the extent this finding generalizes, the results suggests the difference in FQoL for low versus high parental numeracy among families with a child with ASD is larger than the average negative effect on FQoL of having a child with ASD as compared to families with TD children.

Table 8.3. Conditional effect of group on FQoL at different values (mean, and plus/minus one standard deviation from mean) of objective numeracy.

					95% CI	
Objective numeracy value	Coeff	SE	t	*p*	LLCI	ULCI
4.202	−6.39	.99	−6.45	<.001	−8.37	−4.40
7.492	−4.72	.68	−6.91	<.001	−6.08	−3.35
10.781	−3.05	.97	−3.14	.003	−4.99	−1.10

IV. Discussion

To our knowledge, this is the first study investigating whether objective and subjective numeracy predicts FQoL in families with a child with ASD vs. TD. Our results suggest that objective numeracy but not subjective numeracy may act as a positive factor protecting the negative effect of having a child with ASD on perceptions of quality of life. This effect held after controlling for several factors that could influence FQoL including severity of ASD, level of education, and social support.

These results are in line with previous research showing that risk literacy often help people when they make informed decisions in daily life (Cokely et al., 2012). Specifically,

we tested one of the most predictive individual differences in decision making about health (i.e., numeracy; Lipkus, Peters, Kimmick, Liotcheva, & Marcom, 2010; Reyna et al., 2009). Unfortunately, many people lack basic numeracy, which limits their ability to accurately interpret risks (i.e., risk literacy; Cokely et al., 2012). Our research adds to this literature showing that parents of children with ASD and numerical skill deficiencies are also vulnerable and experience lower levels of quality of life. These families might have more difficulty to understand and use the large amount of conflicting information that they receive about their children's condition. For instance, these people might have more difficulty understanding information about the nature and causes of their children's condition, including the probabilistic role of genetics and the environment (Lea, Kaphingst, Bowen, Lipkus, & Hadley, 2011); they might be more vulnerable to information overload when exposed to higher levels of decision complexity; they might have more unrealistic expectations about the effectiveness and risks of novel medical treatments; and they might have more difficulty interpreting the risks that their children suffer side effects. Other potential explanations of our results include suboptimal decisions about treatment, lower adherence to treatment, more negative interaction with health care professionals, and worse general adjustment to living with ASD, which can ultimately affect FQoL.

In contrast, families of children with ASD with high number might accurately understand and use the information that they receive about their children's symptoms and should be more likely to manage their condition and organize their family environment, which would improve their FQoL. Overall these results suggest that recent skill assessments (e.g., brief numeracy tests that assess levels of risk literacy such as the Berlin Numeracy Test; Cokely et al., 2012, 2018) can help identify vulnerable individuals, who would specially benefit from clinical interventions. In addition, educational efforts and simple interventions designed to improve risk understanding and decision-making skills (e.g., transparent visual aids) would crucial elements of potential long-term solutions and can have substantial benefits at minimal costs, particularly when designed to serve vulnerable populations with limited numeracy (Garcia-Retamero & Cokely, 2013, 2017).

In contrast to other studies (e.g., Khanna et al., 2011; McStay et al., 2014), social support did not predict FQoL in our study; the group of participants with a child with ASD did not have lower levels of social support either. In addition, our results showed that

individual differences in objective numeracy were more reliable predictors of FQoL than social support. One explanation of this result is that our participants in our study were enrolled in a respite care program, which could have provided them with the necessary social support. This support might have improve their mental health has previous research suggests (e.g., Yantzi, Rosenberg, & McKeever, 2007), but it did not reduce or eliminate the negative effect of numerical skill deficiencies on perceptions of quality of life. Thus our research suggests that low levels of numeracy make people vulnerable regardless of other protective factors (e.g., social support).

Our research contributes to the growing literature on parental characteristics that make parents resilient and help families with children with ASD thrive as much as families with children with TD do. Nevertheless, as with any research, our study has several limitations. For instance, our sensitivity analysis indicates that with the obtained sample size, a moderate or large effect size is likely to be detected. However, the current sample size did not allow us to include other potential important control variables in our analyses (e.g., parents' age, employment status, or family composition). Moreover, 93% of the sample consisted of mothers. Thus, fathers as a group have been unrepresented in this study and results cannot be generalized to all parents. Future studies with a larger number of participants investigating the effect of all these factors are encouraged. Moreover, future studies should considered causes and differential consequences of risk illiteracy in these families, and potential interventions that might help parents to improve their decision making in a health context (e.g., decision aids). Nevertheless, our research shows that adequate levels of objective numeracy can help families of children with ASD improve their FQoL, possibly by reducing the burden of information overload and decision vulnerability and helping them managing their children's condition.

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CHAPTER 9

Chapter 9

Language and motor skills in siblings of children with ASD younger than 3 years

The content of this chapter has been published as Garrido, D., Petrova, D., Watson, L. R., Garcia-Retamero, R., & Carballo, G. (2017). Language and motor skills in siblings of children with autism spectrum disorder: A meta–analytic review. *Autism Research*, *10*(11), 1737–1750, doi:10.1002/aur.1829

Language and motor skills in siblings of children with autism spectrum disorder: A meta–analytic review

Children with autism spectrum disorder (ASD) show significant linguistic and motor impairments compared to children with typical development (TD). Findings from studies of siblings of children with ASD show similarities to conclusions from studies of children with ASD. The current meta–analysis reviewed studies reporting linguistic and/or motor skills in siblings of children with ASD compared to siblings of children with TD. Thirty–four studies published between 1994 and 2016 met all inclusion criteria. We compared three different age groups (12 months or younger, 13 to 24 months, and 25 to 36 months). At 12 months, compared to siblings of children with TD, siblings of children with ASD had worse receptive language (d = –.43, 95% CI [–.53 –.33]) and expressive language skills (d = –.40, 95% CI [–.57,–.23]), and these effects were sustained at 24 and 36 months. Similar, albeit smaller differences in fine motor skills were detected at 12 months (d = –.22, 95% CI [–.39,–.04]), and these differences were larger at 36 months (d = –.36, 95% CI [–.54,–.17]). There were differences in gross motor skills at 12 months (d = –.22, 95% CI [–.40,–.04]), but only a few studies were available at later ages. Compared to siblings of children with TD, infants who have siblings with ASD have worse linguistic and motor skills. These differences are detectable as early as when infants are 12 months old and seem to be sustained until they are 3 years old. Differences in language skills are larger than those in motor skills, especially during the first year.

I. Introduction

Autism spectrum disorder (ASD) is a complex neurodevelopmental disorder characterized by symptoms in two broad domains, i.e., notable deficits in communication and social interaction, and the presence of restricted and/or repetitive interests and behaviors (APA, 2013). Early signs of ASD can be detected in some children during the first year of life

(Bolton, Golding, Emond, & Steer, 2012), with diagnoses often possible before age 3 (Ozonoff et al., 2015).

Impairments in language and communication are central components of ASD, even though the specific nature and extent of the impairments in children with ASD is variable (Bishop, 2010). Impairment in the social use of language and communication is required for a diagnosis of ASD, but impairments in the development of linguistic structure and vocabulary are not. Despite this, children with ASD often show notable delays in the development of expressive language (e.g., syntax, expressive vocabulary) (Hudry et al., 2010), receptive language (Kamio, Robins, Kelley, Swainson, & Fein, 2007), and phonology (Rapin, Dunn, Allen, Stevens, & Fein, 2009).

Besides language and communication problems, atypical or delayed motor skills are other potential symptoms associated with ASD. A meta–analysis showed that individuals with ASD demonstrate significant and generalized alterations in motor performance (Fournier, Hass, Naik, Lodha, & Cauraugh, 2010) and about 80–90% of children with ASD show some degree of motor difficulties (Hilton, Zhang, Whilte, Klohr, & Constantino, 2012). Such motor difficulties could include atypical fine and gross motor skills (Barbeau, Meilleur, Zeffiro, & Mottron, 2015; Landa & Garret–Mayer, 2006).

I.I. Siblings of children with ASD: language and motor skills

Research confirms that siblings of children with ASD (often referred to as high risk children; HR) have an increased likelihood of developing ASD themselves, or of developing sub–clinical symptoms of ASD, compared to siblings of children with typical developmental (TD) (also referred to as low risk children; LR) (Messinger et al., 2013). For instance, studies estimate that about 2–19% of HR children receive an ASD diagnosis, compared to 0.6% of LR children (Newschaffer, Fallin, & Lee, 2002; Muhle, Trentacoste, & Rapin, 2004; Levy, Mandel, & Schultz, 2009; Ozonoff et al., 2011). In addition, one study found that about 19% of HR children not later diagnosed with ASD nevertheless showed significantly elevated autistic traits by 12 months of age, (e.g. reduced eye contact, orienting to name, and social smiling; Georgiades et al., 2013). This suggests that even if HR children do not show the pattern or

severity of symptoms that warrant an ASD diagnosis, they can have different or less severe developmental problems such as language delays (Johnson, Myers, & American Academy of Pediatrics Council on Children with Disabilities, 2007; Gamliel, Yirmiya, Jaffe, Manor, & Sigman, 2009; Paul, Fuerst, Ramsay, Chawarska, & Klin, 2011).

Indeed, HR children are more likely to show developmental impairments in language and communication (Drumm & Brian, 2013), and language difficulties are among the main indicators of the Broader Autism Phenotype (BAP, the presence of autistic–like traits) during preschool years (Elsabbagh & Johnson, 2007; Toth, Dawson, Meltzoff, Greenson, & Fein, 2007). Such difficulties can range from lack of fluency (Ozonoff, Rogers, Farnham, & Pennington, 1993) to severe pragmatic difficulties (Ben–Yizhak et al., 2011; Levy & Bar–Yuda, 2011). Some studies have shown that as a group, compared to LR children, HR children demonstrate lower receptive and expressive language skills (i.e., fewer canonical syllables and use of words) already during the first three years of life (Toth et al., 2007; Paul et al., 2011).

HR children are also at increased risk for motor difficulties, although the literature does not provide the same level of evidence related to motor skills in this population as is available for language skills. Interestingly, whereas most HR children have motor skills falling within the typical developmental range, they may nevertheless use less sophisticated functional movements than expected (Mulligan & White, 2012). It has also been suggested that HR children may show relatively high scores in gross motor skills but low scores in fine motor tasks due to difficulties in motor imitation (Klin, Saulnier, Tsatsanis, & Volkmar, 2005). For instance, one study detected some movement anomalies in HR children compared to LR children as early as 6 months of age (i.e., difficulties with postural control; Flanagan, Landa, Bhat, & Bauman, 2012).

The results reported above suggest that studies of HR infants can provide valuable information about early markers of the BAP (Paul et al., 2011; Pisula & Ziegart–Sadowska, 2015). Further research on the development of HR children could also help us attend early on to potential intervention needs that these children may have. However, the results of the existing studies are mixed and it is not clear (a) to what extent there are developmental differences in language and motor skills between HR and LR children, (b) when these differences can be detected, and (c) whether the differences increase or decrease with age.

To help clarify these issues, and to provide a synthesized overview of the differences between siblings of children with ASD and siblings of children with TD in the areas of language and motor development, we systematically reviewed the available literature using meta–analytic methods and compared language and motor skills in HR and LR children.

Our focus on impairments in language and motor skills is motivated by theoretical models positing mechanistic links between the two domains (e.g., Alcock & Krawczyk, 2010; Leary & Hill, 1996) along with empirical evidence that they are strongly related. For instance, some evidence supports an assumption that motor skills (i.e. locomotor experiences) facilitate social interaction and social communication (Bhat, Landa, & Galloway, 2011; Karasik, Tamis–LeMonda, & Adolph, 2011). Empirically, receptive and expressive language skills correlate with motor skills in children with ASD (Luyster, Kadlec, Carter, & Tager–Flusberg, 2008), and poor motor skills may predict small gains for children with ASD in interventions targeting oral expressive language (Belmonte, Saxena–Chandhok, Cherian, Muneer, George, & Karanth, 2013). Oral –and manual– motor skills are predictors of speech fluency (Stone & Yoder, 2001; Thurm, Lord, Lee & Newschaffer, 2007; Gernsbacher, Sauer, Geye, Schweigert, & Hill Goldsmith, 2008), and gesture use is one of the best predictors of receptive and expressive language skills (Luyster et al., 2008). However, it is not clear to what extent deficits in language and motor skills in HR children are related (e.g., what deficits are observed earlier or more strongly).

Consequently, the purpose of this meta–analysis is to estimate the size and consistency of the differences in both language and motor skills between HR and LR children, and to describe how these differences compare to each other (e.g. Are differences in language skills detected earlier? Are they bigger?). In particular, we considered both receptive and expressive language skills, as well as both fine and gross motor skills at different key developmental ages (from 1 to 3 years old). We elected to compare siblings of children with TD (LR) to siblings of children with ASD (HR), regardless of any subsequent diagnosis. Because our interest was studying development in HR children in the period before definitive diagnoses are typically provided, we focused on studies assessing children 3 years old or younger.

II. Methods

II.I. Data collection, inclusion criteria and identification of studies

We conducted a systematic review of empirical articles examining development in siblings of children diagnosed with ASD. We selected those articles that compared siblings of children with ASD (HR children) to siblings of children with TD (LR children) on linguistic and/or motor skills. Our search was limited to articles written in English and published between 1994 (publication date of the DSM–IV establishing diagnostic criteria for ASD) and 2016. We searched the following databases: Web of Science, PubMed, PsycINFO, ERIC, and Medline, using combinations of the following keywords: Autism Spectrum Disorder, ASD, autis[1], siblings, at risk, high risk, low risk, unaffected, affected, language, linguistics, and motor. We identified additional studies from the reference lists of the articles already selected and searched the grey literature (e.g. unpublished studies and congress abstracts). The initial search returned more than 4000 publications. After review of title and abstract 809 articles remained.

From these, we excluded studies (k = 677) according to the following criteria: (a) high risk group did not comprise children with a sibling with ASD, (b) study focused on genetics in HR children, (c) study focused on children at high risk for non–ASD disorders, (d) article was published outside of the specified dates, (e) study reported neuroimaging measures exclusively, (f) study focused on treatment.

Studies were further excluded based on the following exclusion criteria (k=98): (a) LR group was absent, (b) the LR group did not include siblings of children with TD, (c) HR and LR groups were selected based on mixed criteria, (d) children in the LR and HR samples had an average age over 36 months, (e) HR and LR children were not matched on chronological age, (f) the study did not include monolingual children, (g) the study did not report outcomes for both language and/or motor skills, (h) linguistic and/or motor skills were not evaluated with objective scales, (i) the ASD diagnosis of affected siblings was not based on Autism Diagnostic Interview–Revised (ADI–R; Le Couteur, Lord, & Rutter, 2003) or Autism Diagnostic Observation Schedules (ADOS–G; Lord, Rutter, DiLavore, & Risi, 2002), (j) the necessary values required for coding could not be obtained even after contacting the authors, and (k)

[1] An asterisk stands for any character that shares the same root (e.g. autis*: autism, autistic).

article was a duplicate of an already included article, or reported data on a sample that was already included from another publication.

Our final sample included 33 articles, reporting 34 studies. Figure 9.1 offers an overview of the search process. Two authors independently extracted data from the 34 studies and any disagreement was resolved with discussion.

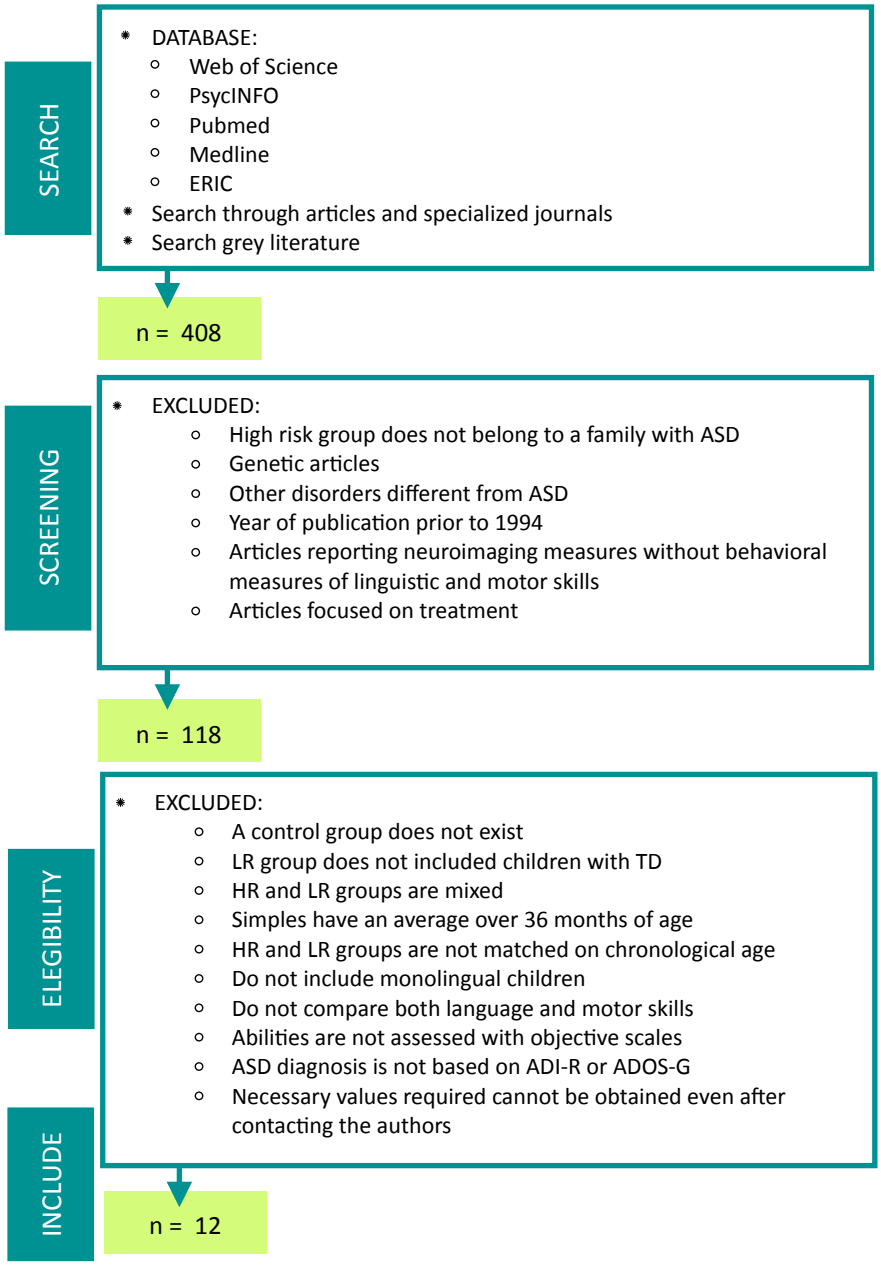


Figure 9.1. Searching process and articles selected.

We recorded the following information for each study: publication year, group sample size, age of children (mean, range and standard deviation for HR and LR groups), and the tests used to measure linguistic and motor skills. We recorded means and standard deviations for the following dependent variables: (a) expressive language, (b) receptive language, (c) fine motor skills, and/or (d) gross motor skills, in the age ranges of (a) up to 12 months, (b) between 13 and 24 months, and (c) between 25 and 36 months. When a study contained measurement data from multiple timepoints within the selected age intervals, we chose the measurement that was closest to the upper value within the interval (i.e. 12, 24 or 36 months). When a study used more than one test to assess language or motor skills, we chose the test that was more frequently used across the sample of studies.

II.II. Meta–analytic procedure

To conduct the meta–analysis we used the metafor package R (Viechtbauer, 2010). Where data from multiple groups had to be combined (e.g., when the HR group was divided in subgroups depending on the later presence of an ASD diagnosis), we followed procedures recommended in the Cochrane handbook for systematic reviews (Higgins, 2008). As a measure of effect size, we calculated the standardized mean difference and followed Cohen (1988) to interpret the sizes of the obtained effects (.2, .5, and .8 for small, medium, and large, respectively). The analyses were based on unadjusted means because none of the included studies provided means adjusted for covariates.

Studies were weighted using the standard “inverse variance” method. In particular, we fitted random effects models, in which studies were weighted by the inverse of the sum of the sampling variances and the residual heterogeneity (Viechtbauer, 2010). We further fitted mixed–effects models considering the following potential moderators: exact age in months, type of test used, and publication year. Because of the small overall number of studies, each potential moderator was examined separately. The models were fitted with restricted maximum likelihood estimation.

To estimate the amount of heterogeneity between studies we conducted statistical tests for heterogeneity and consulted the I^2 statistic. This statistic estimates (in percentages) how much of the total variability of the effect size estimates can be attributed to

heterogeneity among the true effects, such that 30–60%, 50–90%, and 75–100% are considered to reflect moderate, substantial, and considerable heterogeneity, respectively (Higgins, 2008; Viechtbauer, 2010). To examine publication bias in the data, we generated funnel plots. When the number of studies permitted it, we conducted statistical tests for funnel plot asymmetry (Higgins, 2008). In addition, we examined the data visually, using contour–enhanced funnel plots which permit easy identification of asymmetry due to publication bias (Peters, Sutton, Jones, Abrams, & Rushton, 2008).

III. Results

Table 9.1 shows basic information for all studies included in the meta–analysis. The studies were published between 2005 and 2016, and included a total of 2376 children (64% HR and 36% LR) at 12 months, 3764 children (66% HR and 34% LR) at 24 months, and 3422 children (63% HR and 37% LR) at 36 months. The HR and LR groups were not matched a priori on demographic characteristics in any study, and only thirteen studies tested for demographic differences between the groups. In one study (Young et al., 2011), the HR group (n = 157) contained 3 LR children who had received an ASD diagnosis. Given the large simple size of the study and the small number of misplaced LR children, we decided not to exclude this study from the meta–analysis. Fifteen studies included information regarding subsequent ASD and other diagnoses; the remaining 19 did not include such information.

Table 9.1. Basic information of each study used in the present meta–analysis

Authors	HR Siblings		LR Siblings		Scales	
	N	Age: months	N	Age: months	Language	Motor
Zwaigenbaum et al. (2005)	65	12	23	12	MSEL	
Mitchell et al. (2006)	74	12	37	12	MSEL	
	95	24	46	24		
Gamliel et al. (2007)	38	24	38	24	RDLS	
	39	36	39	36	CELF–P	
Presmanes et al. (2007)	46	15	35	15	MSEL	MSEL
Stone et al. (2007)	64	16	42	16	MSEL MCDI	MSEL
Toth et al. (2007)	42	20	20	22	MSEL	MSEL
Yirmiya et al. (2007)	30	24	30	24	RDLS	
	30	36	30	36	CELF–P	
Young et al. (2009)	33	24	25	24	MSEL	MSEL
Young et al. (2011)	157	12	75	12	MSEL	MSEL
	157	24	75	24		
	157	36	75	36		
Paul et al. (2011)	38	12	31	12	MSEL	MSEL
	24	24	21	24		
Key and Stone (2012)	15	9	20	9	VABS	
Macari et al. (2012)	50	12	34	12	MSEL	MSEL
	50	24	34	24		
Mulligan et al. (2012)	13	12	12	12	MSEL	MSEL
Chawarska et al. (2013)	49	6	35	6	MSEL	MSEL
Curtin and Vouloumanos. (2013)	31	12	31	12	MCDI	MSEL
	25	18	26	18		
Droucker et al. (2013)	14	12	20	12	MCDI	
	11	18	21	18		
Ference and Curtin (2013)	20	12	23	12	MCDI	
Ibañez et al. (2013)	26	36	13	36	MSEL	
Schwichtenberg et al. (2013)	104	36	76	36	MSEL	MSEL
Elison et al. (2014)	105	12	53	12		MSEL
Hudry et al. (2014)	54	7	50	7	MSEL	
	52	24	47	24		
Klerk et al. (2014)	44	36	40	36	MSEL	MSEL
Libertus et al. (2014)	23	6	19	6	MSEL	MSEL
Libertus et al. (2014)	107	6	22	6	MSEL	MSEL

Ozonoff et al (2014)	294	12	116	12	MSEL	MSEL
	294	24	116	24		
	294	36	116	36		
Gangi et al. (2014)	43	24	13	24	MSEL	
	39	36	20	36		
Herlihy et al. (2014)	21	25	27	25	MSEL	
Leonard et al. (2015)	53	7	48	7	VABS	MSEL
	52	24	47	24		
	53	36	48	36		
Messinger et al. (2015)	1241	24	583	24	MSEL	MSEL
	1241	36	583	36		
Miller et al. (2015)	119	36	188	36	MSEL	MSEL
Talbott et al. (2015)	47	18	27	18	MCDI	
Ekberg et al. (2016)	29	10	16	10	MSEL	
StJohn et al. (2016)	124	12	50	12		MSEL
	125	24	49	24		
Lazenby et al. (2016)	213	12	133	12	MSEL	

Note: BSID–II (Bayley´s Scales of Infant Development; Bayley, 1993), CELF–P (Clinical Evaluation of Language Fundamentals–Preschool; Wiig, Secord, & Semel, 2004), RDLS (Reynell Developmental Language Scales; Reynell and Grubber, 1990), MSEL (Mullen Scales of Early Learning; Mullen, 1995), MCDI (MacArthur–Bates Communicative Development Inventories; Fenson et al., 1993), y VABS (Vineland Adaptative Behaviour Scales–2nd Edition; Sparrow, Cicchetti, & Balla, 2005).

III.I. Language and motor skills tests used

The following tests were used to evaluate language and/or motor skills: Bayley Scales of Infant Development (BSID–II; Bayley, 1993), Clinical Evaluation of Language Fundamentals–Preschool (CELF–P; Wiig, Secord, & Semel, 2004), MacArthur–Bates Communicative Development Inventories (MCDI; Fenson et al., 1993) Mullen Scales of Early Learning (MSEL; Mullen, 1995), Reynell Developmental Language Scales (RDLS; Reynell & Grubber, 1990), and Vineland Adaptive Behavior Scales–2nd Edition (VABS; Sparrow, Cicchetti, & Balla, 2005).

Given the assumption of independence in meta–analysis, we could only select one dependent measure for each ability and sample at each point. We selected the measure most widely used across the included studies in order to decrease variance between studies. The majority of articles used the MSEL (71% for language and 100% for motor skills), so whenever multiple instruments were used, we selected scores from the MSEL. The MSEL is

an extensive standardized assessment of expressive language, receptive language, fine motor, and gross motor skills and provides age equivalent and standard scores from birth to 68 months old. The CELF–P Scale evaluates expressive and receptive language in children aged between 36 and 72 months. The VABS Scale is a parent report measure of communication, daily living, and motor and social skills, used from birth to adulthood. The MCDI Scale is also a parent questionnaire measure of language development used for children aged between 8 and 37 months.

III.II. Differences in language skills

Detailed results from the three meta–analyses on expressive language skills are shown in Figures S9.1, S9.2, and S9.3. Relative to LR children, HR children showed worse expressive language skills at all ages. The size of the effect was moderate at 12 months (SMD = −.40, 95% CI [−.57, −.23], n = 2044, k = 18), at 24 months (SMD = −.34, 95% CI [−.45, −.23], n = 3590, k = 18) and at 36 months (SMD = −.44, 95% CI [−.58, −.30], n = 3422, k = 12).

Detailed results from the three meta–analyses on receptive language skills are found in Figures S9.4, S9.5, and S9.6. Similar to the results on expressive language, relative to LR children, HR children showed worse receptive language skills at all ages. The size of the effect was moderate at 12 months (SMD = −.44, 95% CI [−.53, −.34], n = 1694, k = 15) at 24 months (SMD = −.52, 95% CI [−.68, −.37], n = 3243, k = 15) and at 36 months (SMD = −.48, 95% CI [−.60, −.36], n = 3422, k = 12).

III.III. Differences in motor skills

Figures S9.7, S9.8, and S9.9 provide detailed results from the three meta–analyses on fine motor skills. Relative to LR children, HR children showed significantly worse fine motor skills. The size of the effect was small at 12 months (SMD = −.21, 95% CI [−.39, −.04], n = 1542, k = 12), and small–to–moderate at 24 months (SMD = −.35, 95% CI [−.46, −.24], n = 3177, k = 11), and at 36 months (SMD = −.36, 95% CI [−.54, −.17], n = 2906, k = 6).

Only one study assessed differences in gross motor skills between HR and LR children at 36 months, so we could only conduct meta–analyses at 12 and 24 months. Detailed results of these are shown in Figures S9.10 and S9.11. Relative to LR children, HR children showed significantly worse gross motor skills at 12 months, with a small effect size (SMD = −.22, 95% CI [−.40, −.04], n = 738 , k = 7). Only four studies assessed gross motor skills at 24 months. On average, HR children tended to show worse gross motor skills at 24 months; however, this effect was not statistically significant (SMD = −.57, 95% CI [−1.20, .05], n = 377, k = 4). The one study that assessed differences in gross motor skills between HR and LR children at 36 months showed significant differences between the groups (SMD = −.44, 95% CI [−.83, −.04], n = 101, k = 1).

III.IV. Moderators

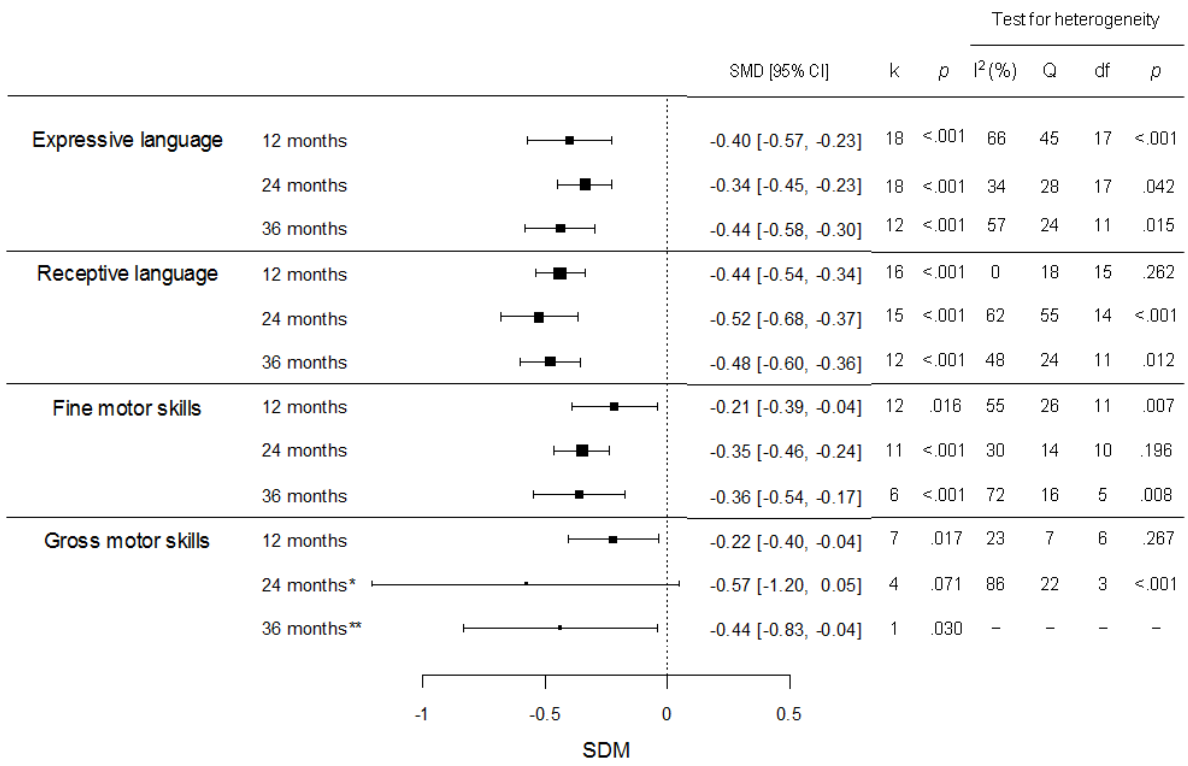
We investigated if the year of publication, the average age of participants in months, and the type of test used (i.e., parent–report or not) influenced the effect size by fitting mixed–effects models and testing for moderation where applicable. For instance, due to the relationship between motor and language skills, it is possible that children with poor language skills may show poor motor skills partially due to failure to understand motor task instructions. We did not find effects of clinician– vs. parent–report measures on language skill differences ($p > .05$). Because motor skills were measured with the MSEL in all studies, we were not able to compare clinician– vs. parent–report measures of motor skills, leaving us unable to fully address the question of whether parents would report different levels of motor skills based on observations outside of a testing context.

Moderator tests indicated that for the interval 25–36 months, smaller effects were observed in younger versus older children for expressive language ($QM(1) = 9$, $p = .003$) and receptive language ($QM(1) = 12$, $p = .001$). These differences were driven by the study by Herlihy and colleagues (2014). This was the only study in the sample in which the assessment was performed at 25 and not at 36 months and it found no significant effects (see Figures S3 and S6). There was another effect of age on fine motor skills in the 3–12 months interval, such that larger differences were found in younger children ($QM(1) = 7$, $p = .008$, see Figure

S9.7). Finally, studies published later found larger differences in expressive language at 24 months (QM(1)=5, p = .026, see Figure S9.2).

III.V. Comparisons of effects

Figure 9.2 gives an overview of the estimated effect sizes and results from the tests for heterogeneity for each dependent variable (expressive language, receptive language, fine motor, and gross motor skill). In this figure we can compare the effect sizes by age, and observe that differences in both language and motor skills are reliably detected as early as 12 months of age. The figure further suggests that the differences in language are somewhat larger compared to differences in motor skills. For instance, at 12 months differences in language are about twice as large as differences in motor skills. Finally, there is a tendency such that larger differences in fine motor skills are detected at a later age

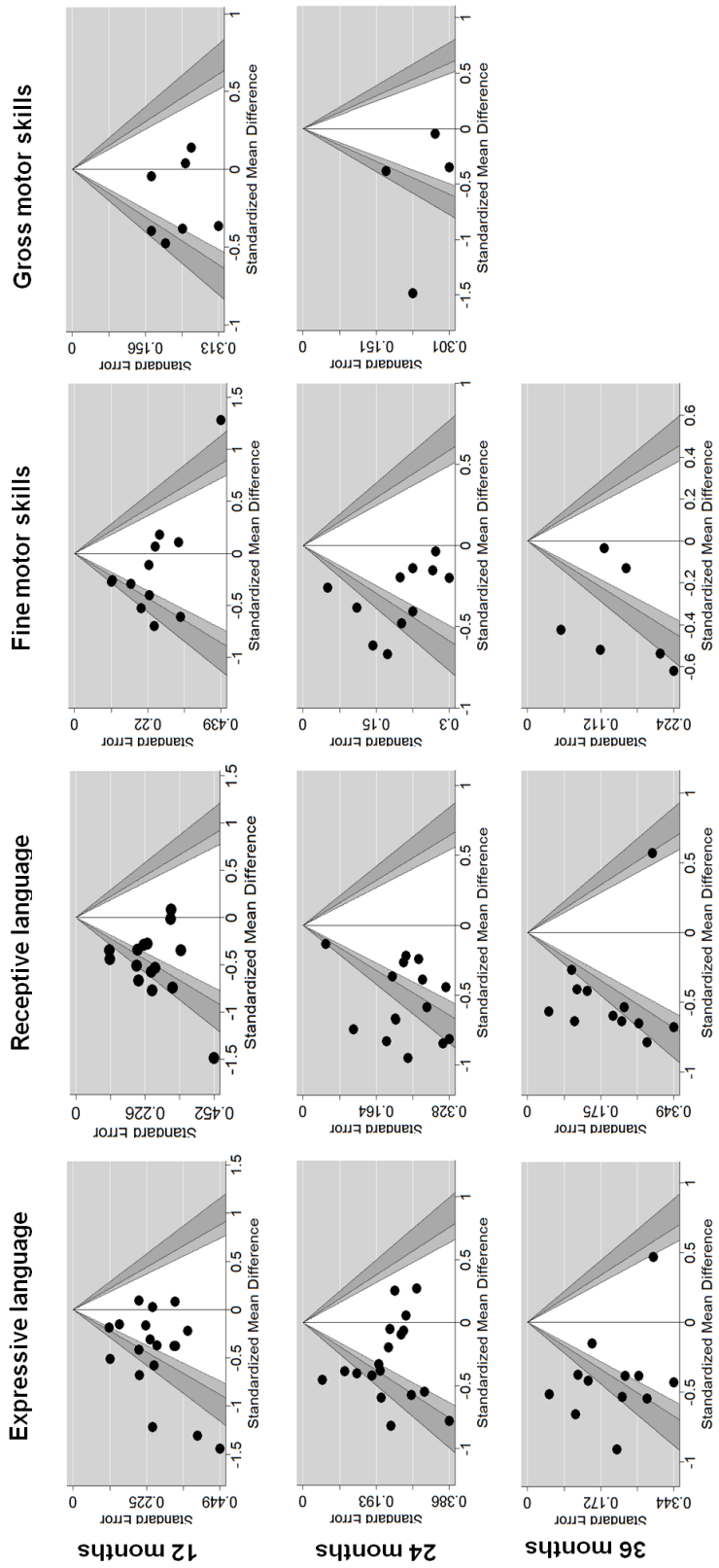


Note. SDM=standardized mean difference: A negative value indicates lower scores for the HR vs LR group. The observed effects are drawn proportional to the precision of the estimates. LLCI/UPLL=Lower/Upper level 95% confidence intervals. Confidence intervals not crossing 0 (i.e., the reference line) indicate a significant effect. I^2=Estimated % of the total variability in effect size estimates that can be attributed to variability among the true effects.

Figure 9.2. Estimated effects sizes and tests for heterogeneity from random effects models for each of 11 mini meta–analyses

III.VI. Publication bias

Figure 9.3 shows contour–enhanced funnel plots for the eleven mini meta–analyses, in which publication bias is signaled if studies appear to be missing in the white regions of statistical non–significance. Generally we observed no signs of publication bias, with the exception of receptive language at 36 months where studies appear to be missing in the regions of non–significance, despite a non–significant asymmetry test ($p > .05$).



Note. The unshaded (i.e., white) region in the middle corresponds to p–value>.10, the gray–shaded region to p–values between .10 and .05, the dark gray–shaded region to p–values between .05 and .01, and the region outside of the funnel corresponds to p–values<.01. If studies appear to be missing in areas of statistical non–significance (i.e., white areas), publication bias is likely (Peters et al., 2008).

Figure 9.3. Contour–enhanced funnel plots

IV. Discussion

After a systematic search and review, we examined the 34 eligible studies providing data on linguistic and/or motor skills in siblings of children with ASD (HR children) and siblings of children with TD (LR children). Our goal was to estimate to what extent HR children show differences in these skills relative to LR children in the first three years of life, and to compare the performance of HR children on motor versus language development. The collection of mini meta–analyses presented here demonstrates that, compared to children who have older siblings with TD, children who have older siblings with ASD have worse linguistic and fine motor skills, and these differences are detectable during the first three years of life. Our results accord with those from other authors (e.g. Ozonoff et al., 2010), who report that the first ASD symptoms can already be seen at 12 or 24 months in infants who are later diagnosed with ASD. Our results show that infants at heightened genetic risk for ASD as a group show patterns of lower performance similar to those found in HR infants later diagnosed with ASD. Consistent with our target population being at risk for ASD, but not necessarily developing ASD, we found small to moderate differences between HR and LR children. Importantly, this meta–analysis demonstrated that on average, detectable differences in language and motor skills based only on genetic risk can be expected already during the first three years of life. Differences in language were about twice as big as differences in motor skills as early as 12 months of age. This suggests that at an early age language assessment might be more useful than motor skills assessment at detecting risk of subsequent development delays.

IV.I. Expressive and receptive language in siblings of children with ASD

Our analyses show significant differences in language skills between siblings of children with ASD and siblings of children with TD. For both receptive and expressive language skills effect sizes are moderate already at 12 months and remain so at 36 months. These results are interpreted as additional support for previous findings (e.g. Zwaigenbaum et al., 2005; Landa & Garrett-Mayer, 2006) showing significant differences in expressive and receptive language in HR compared to LR children. They suggest that potential deficits in HR children can be reliably identified already during the first year of age. In the current data, we do not see

evidence that these differences in language increase or decrease from the first to the third year.

If we compare both language aspects, we find somewhat stronger differences in receptive language than expressive language at 12, 24, and 36 months. Assuming that language deficits increase with age, these results suggest that differences in comprehension between HR and LR children are detectable earlier than differences in expression, similar to what is found in children with ASD (e.g. Landa & Garrett-Mayer, 2006; Snyder, 2007; Goodwin, Fein, & Naigles, 2012). This finding is in line with the idea that language development in children with ASD follows a similar pattern to that of TD children. In other words, infants (both with ASD and TD) understand words before they begin saying them.

IV.II. Fine and gross motor skills in siblings of children with ASD

Our analyses show that there are significant differences between HR and LR infants in fine motor skills at 12, 24 and 36 months. Looking at the longitudinal trajectory, we see some evidence that differences in fine motor development between HR and LR children become larger from the first to the third year. This finding is consistent with Ozonoff and colleagues (2015), who found that HR children could show additional symptoms warranting an ASD diagnosis at three years despite not meeting criteria for the diagnosis at earlier ages. Additionally, our results are in line with those of Leonard et al. (2015), who found much larger differences in gross motor skills between HR and LR children at 36 compared to 7 months of age (see Figures S9.10 and S9.11).

Overall, compared to linguistic skills, differences in fine motor skills are smaller, especially during the first year of life. This suggests that, contrary to what some previous studies have suggested (see Iverson, 2010), instead of difficulties in movement contributing to language delays, language delays at a very early age could be contributing to fine movement differences that become evident or more pronounced later. For instance, language delays and lack of (successful) communication attempts could be limiting the experiences of some HR children that would support the normal trajectory of development of fine motor skills. It is also possible that there is a bidirectional process, such that language

and motor skills influence each other. Longitudinal studies assessing both abilities could clarify the developmental trajectory (e.g., Leonard et al. 2015).

The finding that HR children show less proficient fine and gross motor skills than LR children supports outcomes from studies that illustrated differences and/or deficiencies in movement between siblings of children with ASD and siblings of children with TD (e.g. John et al., 2016; Landa & Garrett-Mayer, 2006; Ozonoff et al., 2010; Flanagan et al., 2012). The current review also highlights the need for studies comparing gross motor skills in HR and LR children, especially in two and three–year old infants. Such studies can give us further insight into the developmental trajectory of children at high risk.

It is also important to keep in mind that all studies that met the inclusion criteria used the MSEL to assess motor skills. This means that the extent of the detected differences between HR and LR children is limited by the sensitivity of this particular test. For instance, HR children may be experiencing motor difficulties that are not reliably detected by the MSEL, in which case differences in motor skills may actually be larger than the results reported here suggest.

IV.III. Limitations

This meta–analysis was based on mean differences unadjusted for important demographic covariates, as these were not reported in the selected studies. None of the studies compared groups that were matched based on important demographic characteristics. This limitation potentially introduces heterogeneity and reduces the precision of the effect sizes estimated here, as adjusting for covariates can either increase or decrease the obtained effect size, depending on the relationship of the covariate to the outcome of interest (Voils, Crandell, Chang, Leeman, & Sandelowski, 2011). Future studies should take into account factors like gender, severity of the sibling's ASD diagnosis, and diagnostic outcome, as these variables could be potential moderators. We did not find any important moderators that had consistent influence on effect sizes across all mini meta–analyses. However, given that there was small variability between studies on some of the parameters tested (e.g., only a few studies used parent–based measures), more studies are needed to draw definitive conclusions regarding possible moderators.

IV.IV. Implications for future research and practice

We would like to encourage further research into language and motor skills development in populations at high risk for ASD identified on the basis of familial (genetic) risk. Knowing the specific language and motor difficulties those children at risk for ASD may experience, at what age these start to appear, and what tests are best at detecting them, can help health professionals intervene in families with children at high risk. For instance, further research along these lines would help us identify at a very early age the risk of specific later diagnoses (e.g., ASD vs. TD vs. language delay) and would increase the possibility for early intervention not only for HR children who will later be diagnosed with ASD but also for those who will later manifest other developmental problems.

Future research on HR children should focus on answering the following questions: what happens when HR children grow up? It seems that differences in motor and language development would continue to be heterogeneous and not only apparent at earlier ages (e.g. Gamliel et al. 2009). Do differences between HR and LR children increase incrementally with age into adulthood, or do differences dissipate over time? If the HR children who go on to be diagnosed with ASD are removed from the comparisons, what are the effect sizes for differences in linguistic and motor skills of HR children not ever diagnosed with ASD compared to LR children? Answers to these questions could provide a better understanding of the potential value of implementing interventions for HR infants before the time a definitive diagnosis of ASD can be made. That is, if in the natural course of development, differences dissipate with time in any of these areas, then prodromal interventions may not be cost–effective, given that only about 20% of infant siblings of children with ASD will eventually receive a diagnosis of ASD themselves (Ozonoff et al., 2011). On the other hand, if differences between HR and LR children increase over time, and especially if this pattern is evident for the 11–38% of HR children who will not ever meet criteria for an ASD diagnosis but who will meet criteria for other diagnoses and/or exhibit cognitive or language delays (Charman et al., 2016; Elsabbagh & Johnson, 2010; Miller et al., 2016; Zwaigenbaum et al., 2005), this would offer stronger support for early intervention with all infant siblings of children with ASD.

The results of our meta–analysis reflect the heterogeneity of the available studies of children with ASD and related populations. As mentioned before, there are mixed results concerning specific deficits observed in younger siblings of children with ASD (e.g., see Gamliel, Yirmiya, & Sigman, 2007; Hudry et al., 2014 for findings on language skills and Hilton et al., 2012; Mulligan & White, 2012 for findings on motor skills). The findings of our meta–analyses support the idea that whereas there is not a stable or homogeneous pattern of altered linguistic and motor skills in siblings of children with ASD, there certainly are atypical aspects of language and motor skills among HR children that yield differences in group comparisons between HR and LR children already during the first three years of life (Georgiades et al., 2013; Ozonoff et al., 2014 Gammer et al., 2015).

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Note. References marked with an asterisk indicate studies included in the meta–analysis.

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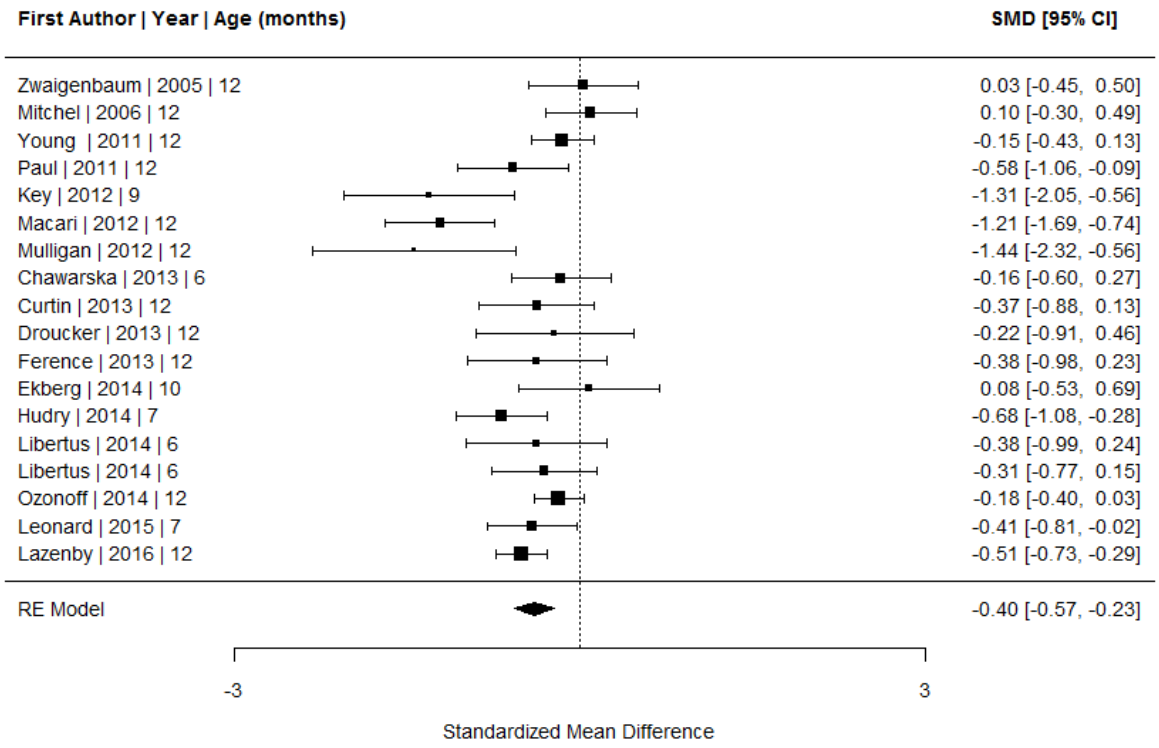
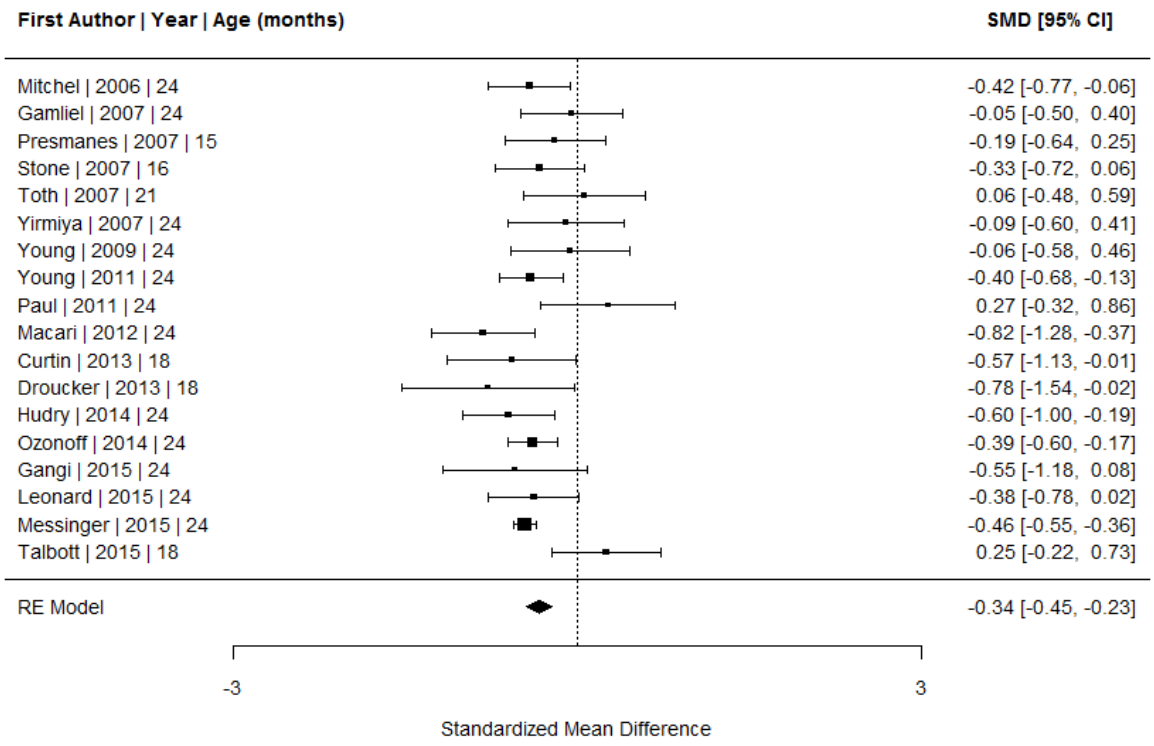
Supporting information



Note. SMD= Standardized mean differences and 95% confidence intervals. A negative value indicates lower scores for the high risk vs. low risk group. The observed effects are drawn proportional to the precision of the estimates.

Figure S9.1. Forest plot for expressive language abilities at 12 months



Note. SMD= Standardized mean differences and 95% confidence intervals. A negative value indicates lower scores for the high risk vs. low risk group. The observed effects are drawn proportional to the precision of the estimates.

Figure S9.2. Forest plot for expressive language abilities at 24 months

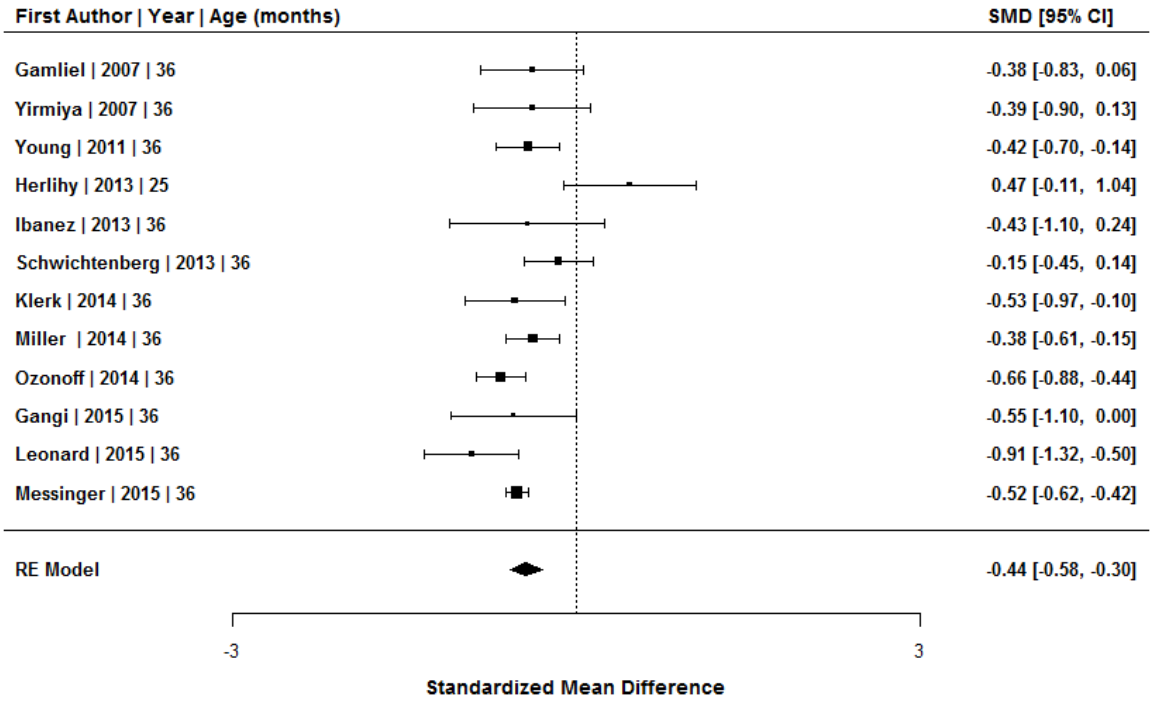


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Figure S9.3. Forest plot for expressive language abilities at 36 months

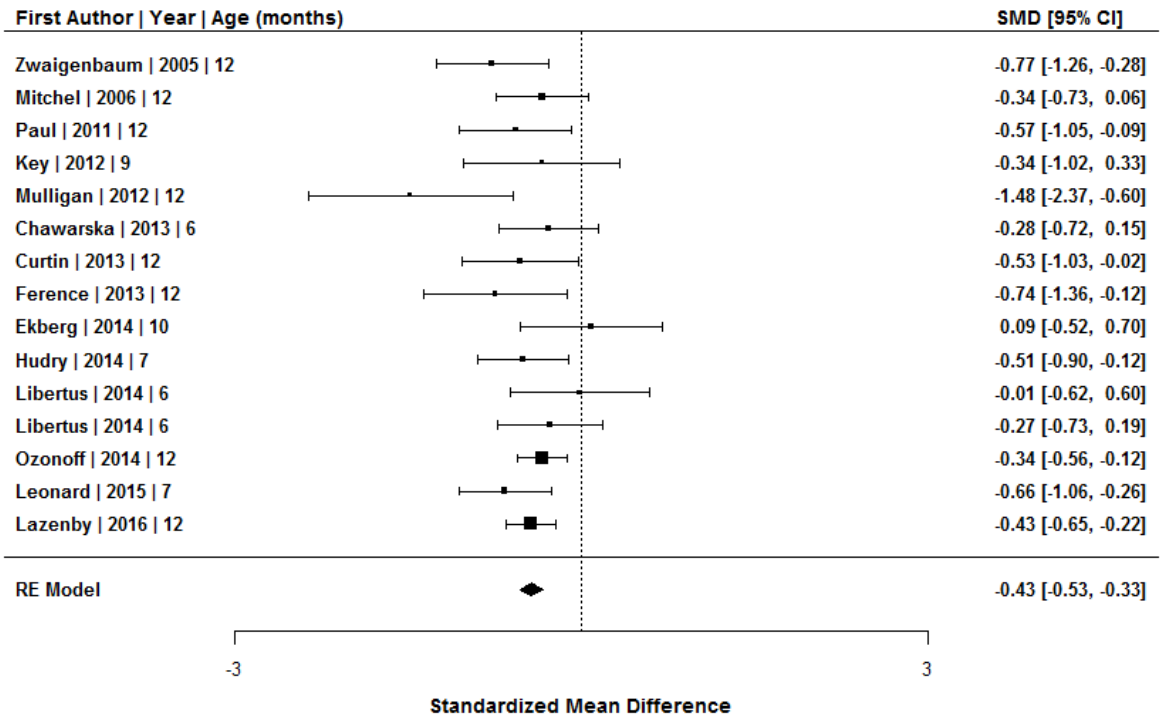

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Figure S9.4. Forest plot for receptive language abilities at 12 months

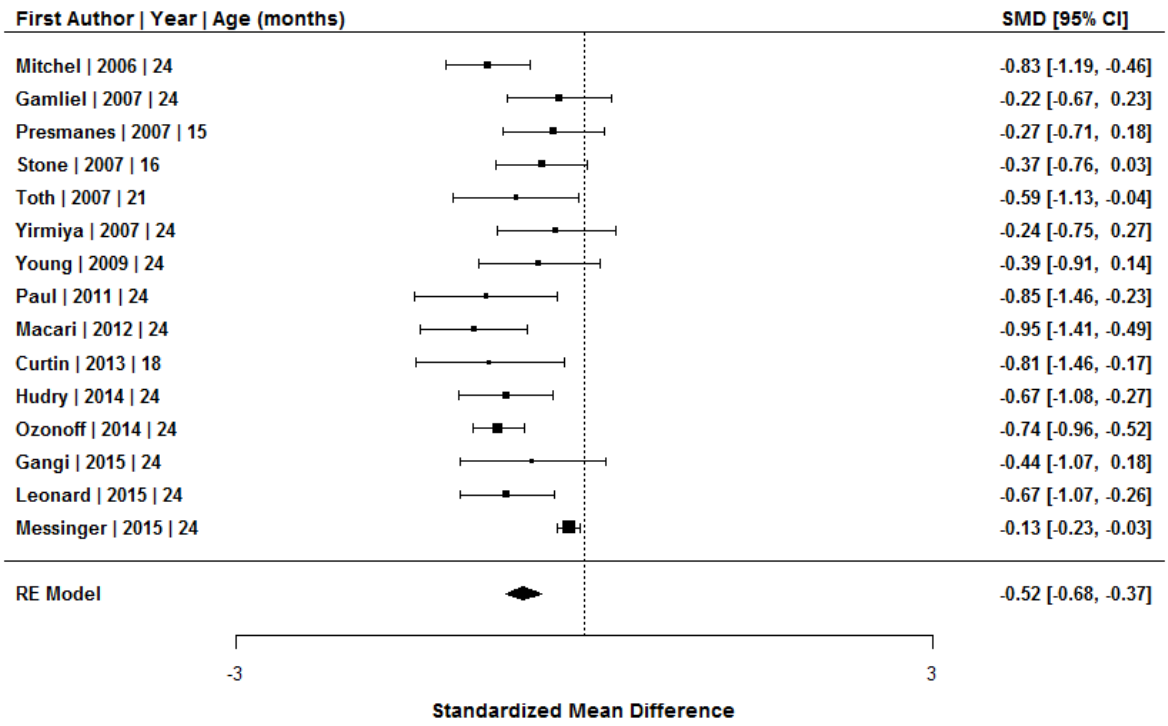

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Figure S9.5. Forest plot for receptive language abilities at 24 months

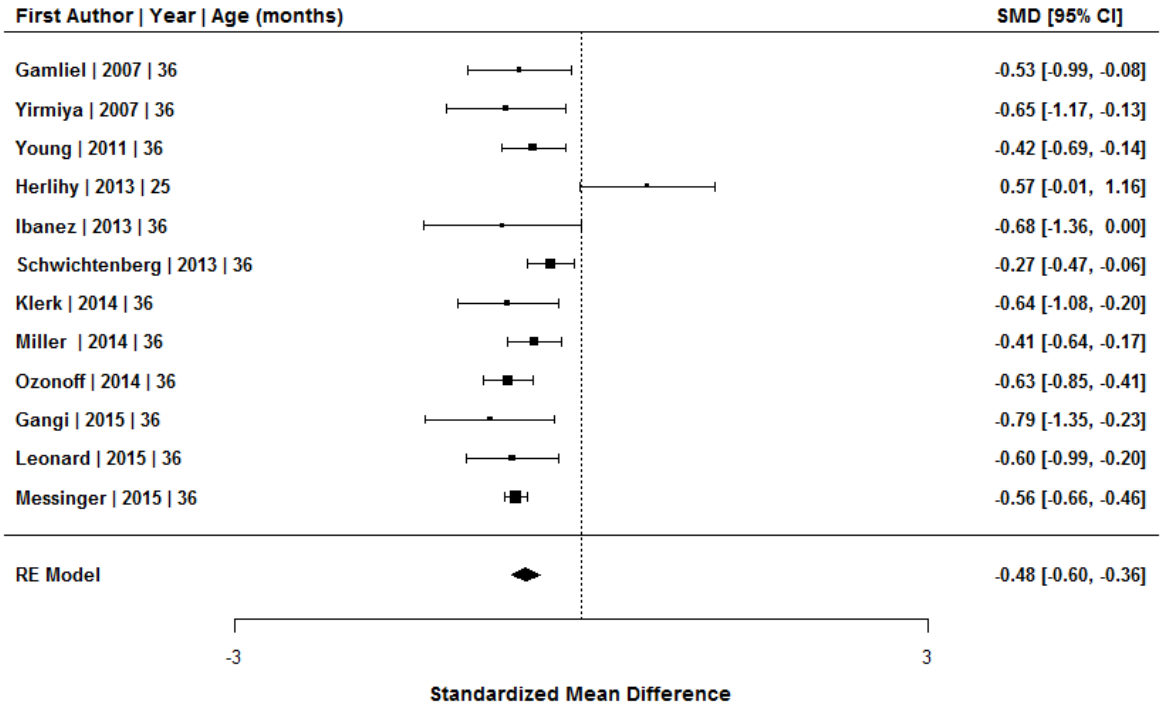

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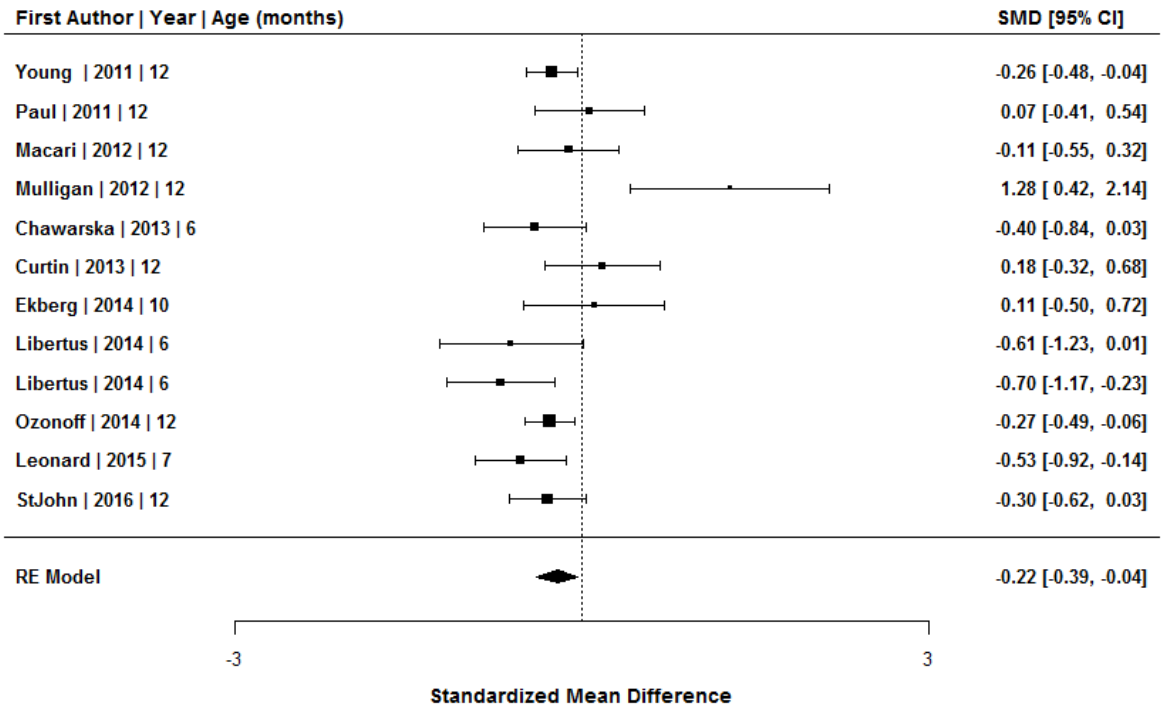
Figure S9.6. Forest plot for receptive language abilities at 36 months



Note. SMD= Standardized mean differences and 95% confidence intervals. A negative value indicates lower scores for the high risk vs. low risk group. The observed effects are drawn proportional to the precision of the estimates.

Figure S9.7. Forest plot for fine motor skills at 12 months

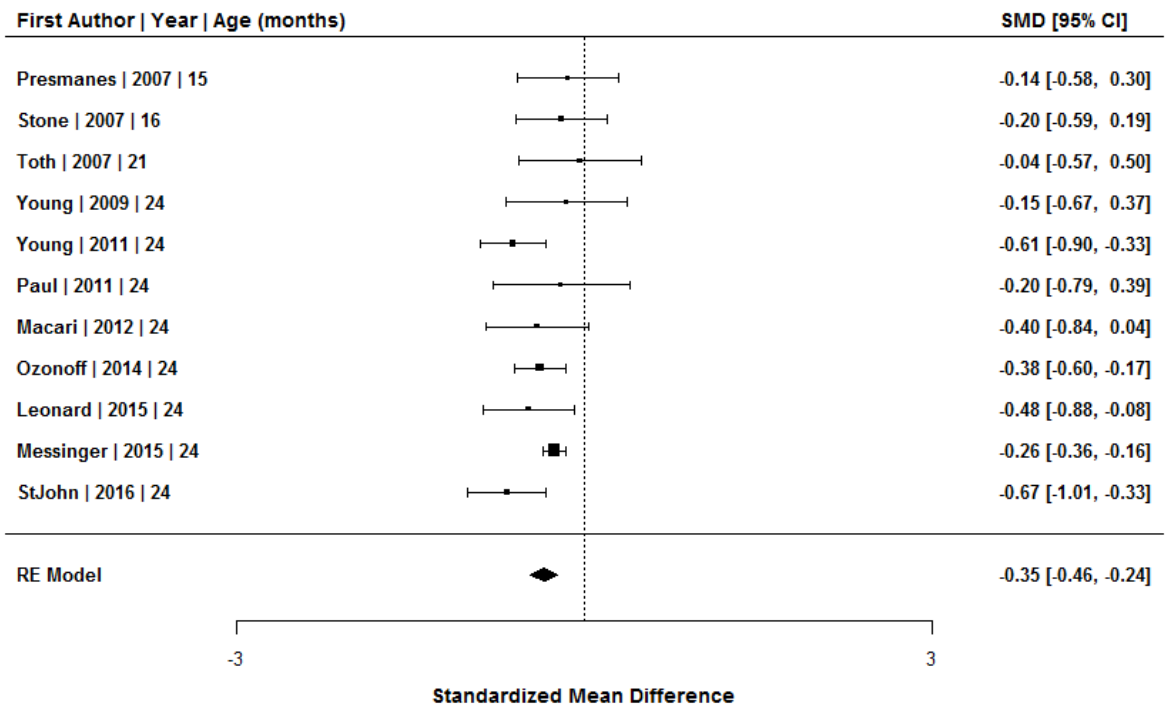


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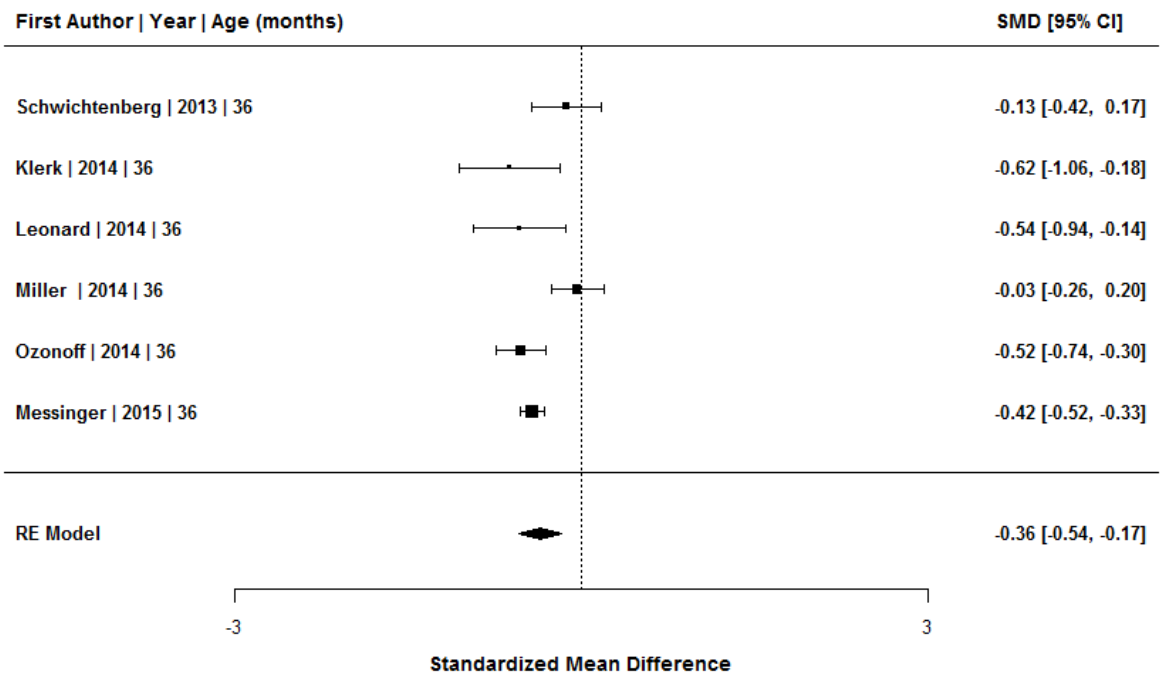
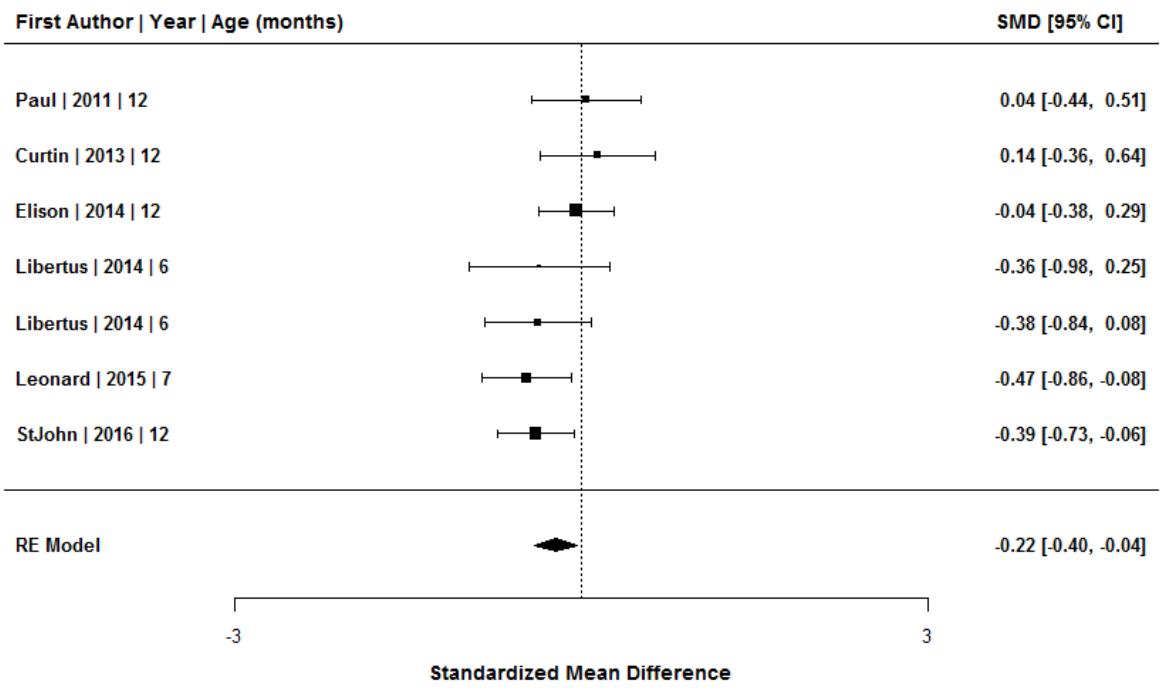
Figure S9.8. Forest plot for fine motor skills at 24 months



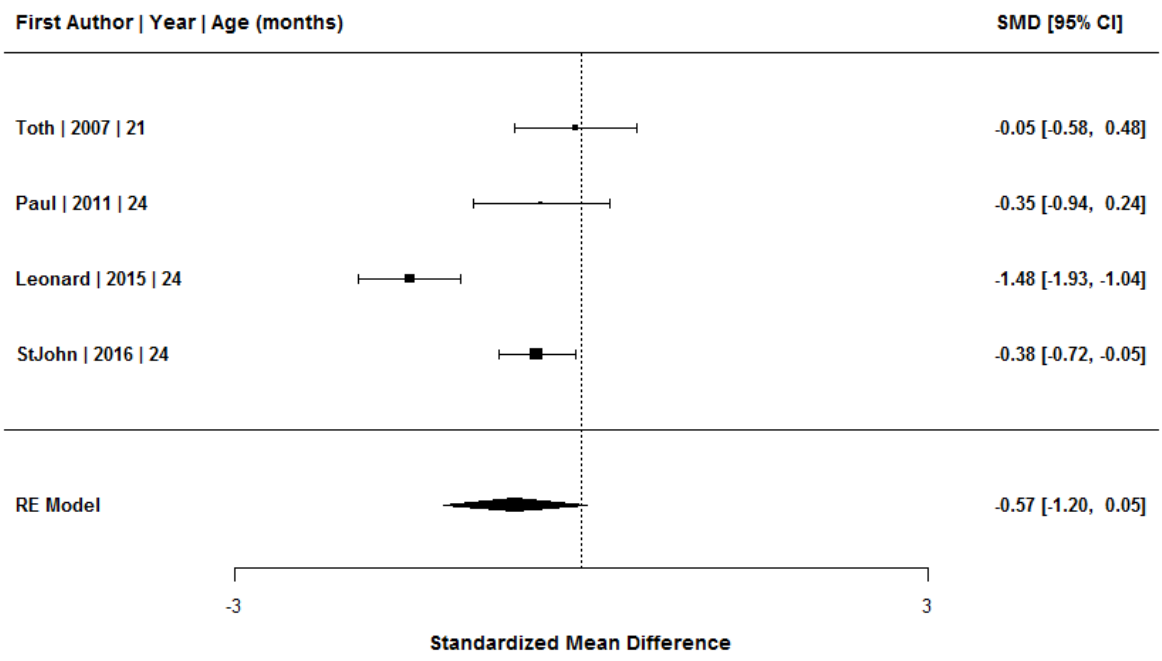
Note. SMD= Standardized mean differences and 95% confidence intervals. A negative value indicates lower scores for the high risk vs. low risk group. The observed effects are drawn proportional to the precision of the estimates.

Figure S9.9. Forest plot for fine motor skills at 36 months



Note. SMD= Standardized mean differences and 95% confidence intervals. A negative value indicates lower scores for the high risk vs. low risk group. The observed effects are drawn proportional to the precision of the estimates.

Figure S9.10. Forest plot for gross motor skills at 12 months



Note. SMD= Standardized mean differences and 95% confidence intervals. A negative value indicates lower scores for the high risk vs. low risk group. The observed effects are drawn proportional to the precision of the estimates.

Figure S9.11. Forest plot for gross motor skills at 24 months

CHAPTER 10

Chapter 10

Language, motor, social support and family quality of life in school–age siblings of children with ASD

The content of this chapter has been published as Garrido, D., Carballo, G., & Garcia-Retamero, R. (under review). Siblings of children with autism spectrum disorders: Social support and quality of life.

Siblings of children with autism spectrum disorders: Social support and quality of life

Autism spectrum disorder (ASD) often has a significant impact on all family members, including parents and siblings of the person who suffers the disorder. The purpose of this study was to explore potential factors that help explain the impact of having an older sibling with ASD on several developmental domains, and to test whether these factors could explain their satisfaction on family quality of life (FQoL). A total of 82 unaffected siblings of children with ASD (Sibs-ASD) and siblings of children with typical development (Sibs-TD) from 4 to 11.9 years old were evaluated in several domains, including intelligence, autistic traits, language, motor skills, social support, and FQoL. Statistical differences were found in motor skills, autistic traits, social support, and satisfaction on FQoL (all with $p<.05$). However, no differences were observed on expressive and receptive language, intelligence, social communication or importance on FQoL. Our results suggest that social support act as a positive factor protecting from the negative effect of having a sibling with ASD on satisfaction of quality of life (r = .23). Social support may be a critical aspect to consider in interventions for improving the satisfaction on FQoL.

I. Introduction

Autism Autism spectrum disorder (ASD) is a neurodevelopmental disorder defined by social-communication challenges and restricted and repetitive behaviors (APA, 2013). Because of these characteristics, ASD might be expected to have an impact on the experiences in families (Gardiner & Iarocci, 2012). For example, parents of children with ASD reported significant levels of stress and depression, lower happiness, and lower family support compared to those parents of children with typical development or other disabilities (Bundy & Kunce, 2009; Higgins, Bailey, & Pearce, 2005; Kogan et al., 2008). These levels of stress and depression could have a negative impact on their family quality of life (FQoL), which is a global construct that reflects family well-being, and has emerged as a good outcome to define the global life situation of families (Mannan, Summers, Turnbull, & Poston, 2006).

To understand the complex experience of families of children with ASD, not only the impact on parents should be considered. Assuming the influence of ASD on the whole family system, parents and siblings (Sibs-ASD) might be affected. Although research reveals an impact on family members, not all the member have a similar experience as a result of having a member with ASD in the family (Hastings et al., 2005). For instance, Lovell and Wetherell (2016) found that Sibs-ASD reported more emotional problems and depressive symptoms than siblings of children with typical development (Sibs-TD). In addition, Meadan, Stoner, and Angell (2010) stated that some characteristics of Sibs-ASD (e.g., social support) might have an influence over FQoL.

Additionally to the experience of having a sibling with ASD, it is important to consider potential factors that could have an impact on Sibs-ASD. In particular, research estimates that 25% of Sibs-ASD show subclinical symptoms of ASD (Georgiades et al., 2013; Messinger et al., 2013). Drumm and Brian (2013) found that Sibs-ASD are at an increased risk for developmental differences, especially in those domains that are compromised in ASD. Receptive and expressive language, and fine and gross motor difficulties are some of these features that have been found among Sibs-ASD from first to third year of life (Paul, Fuerst, Ramsay, Chawarska, & Klin, 2011; see Garrido, Petrova, Watson, Garcia-Retamero, & Carballo, 2017 for a meta-analytic review).

Social communication is another domain that is generally considered as a subclinical feature of ASD in Sibs-ASD (Drumm & Brian, 2013; Miller et al., 2015, Tager-Flusberg et al., 2009). Difficulties in this domain have been described in very young Sibs-ASD (Landa & Garret-Mayer, 2006; Georgiades et al., 2013; Messinger et al., 2013; Miller et al., 2015), but results are mixed. However, there is a dearth of published research on how these deficits in school-age Sibs-ASD are related to other domains of functioning.

Motor, language, and social communication skills in school-age Sibs-ASD are investigated less frequently because most of the research focused on these abilities in Sibs-ASD have been undertaken with young children (Garrido et al., 2017; Gowen & Hamilton, 2013; Mulligan & White, 2012). Therefore, it becomes necessary to evaluate these abilities in older Sibs-ASD, and evaluate if these differences in motor, language, and socio-communication abilities continue or dissipate over time. Some follow-up studies have described different developmental trajectories in Sibs-ASD (Lai, Lombardo, & Baron-Cohen,

2014). For instance, Shepard et al. (2017) did not find differences between Sibs-ASD and Sibs-TD in language at 7 years of life. Similarity, Drumm, Bryson, Zwaigenbaum, and Brian (2015) found that non-ASD Sibs-ASD showed average/above average expressive, receptive and pragmatic language abilities at 8-11 years compared to Sibs-TD. Gamliel, Yirmiya, Jaffe, Manor, and Sigman, (2009) found that forty-one percent of parents of Sibs-ASD reported problems in social communication at age 7 years. However, Shephard et al. (2017) found that Sibs-ASD, in the same age cohort, did not differ significantly from Sibs-TD in social communication abilities.

Despite language, motor development, and social communication, other abilities should be consider in school-age children. As children become older other variables impact over siblings' adjustment such as coping, stress, and social support (Tsai, Cebula, & Fletcher-Watson, 2017). However, less attention has been given to the role of these other domains in Sibs-ASD, such as stress, coping strategies, or social support (Hastings, 2003). Indeed, investigation that evaluates the idea that autistic traits affect sibling adjustment reported mixed results. For instance, although Meyer, Ingersoll, and Hambrick, (2011) found that those Sibs-ASD who exhibit autistic traits might be more likely to report adjustment difficulties, this relationship was completely mediated by maternal depressive symptoms.

At the moment, most studies investigating factors that affect FQoL in families of children with ASD have focused on negative experiences and perceptions (Hastings & Taunt, 2002), although some researchers have begun to examine protective factors (Vasilopoulou & Nisbet, 2016). One such line of research has revealed that social support may improve FQoL in families of children with ASD (Khanna et al., 2011; McStay, Trembath, & Dissanayake, 2014). In that sense, social support has been found to be beneficial to familiar well-being (Ekas, Lickenbrock, & Whitman, 2010; Hastings, 2003; Lin, Orsmond, Coster, & Cohn, 2011). Both informal (e.g., support from friends, extended members and partners) and formal (e.g., professional support) social support have been widely studied in children with ASD and their parents, but not in Sibs-ASD (Tsai et al., 2017).

A formal social support has shown a positive outcome in siblings' adjustment (Tsao, Davenport, & Schmiege, 2012). Hastings (2003) found that higher formal social support was related to fewer adjustment problems in Sibs-ASD. Indeed, a successful adjustment may be moderated by this social support and by the severity of the Sibs-ASD's autism traits. Lowell

and Wetherell (2016) examined the psychophysiological impact of ASD on Sibs-ASD, finding that informal social support in Sibs-ASD (especially from parents and close friends) predicted total depressive symptoms. Moreover, Kaminsky and Dewey (2002) found that higher informal social support was associated with better adjustment at school age. Unfortunately, the relationship between social support in Sibs-ASD and FQoL has not been studied widely in relatives of children with ASD.

In sum, studies of unaffected Sibs-ASD, have found difficulties into mild-childhood (Shephard et al., 2017), and variability in linguistic, and motor areas, social communication, traits of ASD, and social support (see Ben-Yizhak, et al., 2011; Gamliel et al., 2009; Hastings, 2003; Lai et al., 2014; Shephard et al., 2017). Although previous research seems to not provide a strong indication for developmental difficulties in school-age unaffected Sibs-ASD, we are still interested in these domains due to the potential impact of developmental skills on FQoL. Therefore, our main goal was to evaluate several developmental domains in school-age Sibs-ASD, and to test whether either developmental domains or social support in Sibs-ASD could predict family quality of life (FQoL). The current study, which is focused on school-age unaffected Sibs-ASD, has two aims: (a) to evaluate the impact of having a sibling with ASD on social support, considering potential differences in several developmental domains (i.e., expressive language, motor skills, intelligence, and social communication), and traits of ASD, (b) to assess and to determine differences between having or not a sibling with ASD in their FQoL, and (c) to determine whether FQoL might be explained by differences in social support.

II. Method

II.I. Participants

A sample of 82 unaffected siblings of children with ASD (Sibs-ASD, N=43), siblings of children with no family history of ASD (Sibs-TD, N=39) between 4 and 12 years, and their families was recruited from Granada, Spain. All parents signed the informed written consent before participation. The inclusion criterion for families in the Sibs-ASD group was to have another child with ASD according to the DSM-TR-IV (APA, 2000) or DSM-5 (APA, 2013) and ADI-R (Le Couteur, Lord, & Rutter, 2003) or ADOS-G (Lord, Rutter, DiLavore, & Risi, 2002). Moreover,

both group of siblings had to show a typical development. Families in both groups were excluded if siblings received special education, related services (e.g. speech therapy), or had an identified emotional, behavioral o developmental disability (e.g. attention-deficit/ hyperactivity disorder, learning disability, speech delay, Down syndrome, language impairment, or cerebral palsy). An additional criterion for participants in the Sibs-TD group was that participants did not have a previously family history of ASD. The Ethics Committee of the University of Granada approved the methodology of this study.

II.II. Measures

We use different assessment measures: direct measures and indirect measures (parental reports). Parents completed a demographic survey developed for the current study, which included child age, gender, family composition, parental education level, parental age, and gender. Additional specific measures related to receptive and expressive language, motor skills, intelligence, social communication, traits of ASD, social support, and FQoL were also collected, as described below.

Receptive language

The comprehension test of grammatical structures (CEG; Mendoza, Carballo, Muñoz, & Fresneda, 2005) evaluates 20 different grammatical structures with different levels of complexity. This test provides a general score in children from 4 to 12 years.

The Peabody Picture Vocabulary Test (PPVT–III; Dunn, Dunn, & Arribas, 2006) is a test that provides an estimation of the receptive vocabulary ability in children older than two years old.

Expressive language

The Clinical Evaluation of Language Fundamentals –Fourth Edition (CELF-4; Semel, Wiig, & Secord, 2006) is a test for determining if a child (from 5 to 21 years) has a language disorder or delay. For those children under 5, we used The Clinical Evaluation of Language Fundamentals Preschool -Second Edition (CELF-Preeschool-2; Wiig, Secord, & Semel, 2009), which has shown adequate to good levels of reliability with CELF-4 (Semel et al.,

2006). We included the expressive language index (normative mean of 100 and SD of 15), which is an overall measure of expressive language skills.

Motor skills.

The Movement Assessment Battery for Children –Second Edition (MABC-2; Henderson, Sugden, & Barnett, 2007) identifies children (from 3 to 16 years) who have motor function impairment. We evaluated 3 areas: manual dexterity, ball skills, and static and dynamic balance. Moreover, this test provides an overall score of total motor skills. Total scores below the 5th percentile are considered indicative of a conclusive motor problem, and scores between the 5th and 15th percentile range suggest a degree of difficulty (Henderson et al., 2007).

Intelligence.

The Wechsler Intelligence Scale for Children (WISC-IV; Wechsler, 2012) provides a composite score in children from 6 to 16 years. For those children under 6, we used the Wechsler Preschool and Primary Scale of Intelligence -Third Edition (WPPSI-III; Wechsler, 2009). Although we were interested in measuring general intelligence quotient (IQ), we calculate separate correlations for verbal abilities, nonverbal abilities, and full scale IQ are . 610, .953, and .846, respectively, testing that both scales show general IQ along consistent dimensions.

Social Communication.

The Social Communication Questionnaire (SCQ; Rutter, Bailey, & Lord, 2005) is a screening for ASD validated for children older than 4 years. This measure offers a cutoff score (15 points or more) than provides a dimensional measure of ASD symptomatology, and can be used to indicate the likelihood that a child has ASD.

Severity of ASD.

Parents completed the *Gilliam Autism Rating Scale* (GARS; Gilliam, 2004). This scale is a norm referenced screening instrument that helps professionals identify ASD. Moreover, it gathers information about specific characteristics typically noted in ASD (stereotyped behaviors, communication, social interaction, and developmental disturbances, which yield an overall autism quotient). We used the autism quotient as

severity of traits related to ASD (cutoff score of 69 or less indicate a child is “unlikely” to have autism, 70-84 indicate a child “possibly” has autism, or 85 and higher indicate child is “likely” to have autism; Hampton & Strand, 2015).

Social support.

We evaluated social support with the *Structural Social Suppor*t (Berkman, & Syme, 1979). This scale was completed by parents and allowed us to know the social support in terms of interactions face to face: the number of friends, the quality of their relatives’ relationship, and the number of weekly contacts they have with their relatives and friends. The final score is a sum of all items and ranges from 3 to 30, where higher scores indicate more perceived social support. This scale showed a good internal consistency (Cronbach’s = .942).

Family quality of life.

Parents completed the F*amily Quality of Life of People Survey* (FQoLS; Verdugo, Rodriguez, & Sainz, 2009). This instrument evaluates FQoL in two domains (importance and satisfaction), and it includes five factors related to FQoL: emotional well-being, family interaction, financial resources, the role of parents, and physical well-being. This scale showed a good internal consistency in both domains: importance and satisfaction (Cronbach’s = .870, and .738 respectively).

II.III. Data analyses

All statistical analyses were performed using SPSS statistics version 22.0. Descriptive statistics were calculated to characterize the sample, including means, and standard deviations.Independent t-test and chi square analyses were used to compare groups on expressive language, motor skills, intelligence, social communication, severity of traits of ASD, social support, and FQoL. Pearson correlations were conducted to test for associations between the outcome (i.e., satisfaction on FQoL) and the five potential predictor variables (i.e., language, motor skills, social communication, severity of traits of ASD, and social support). To determine the unique influence of each predictor, we conducted multiple regressions. Intelligence and gender were entered as covariates in all analyses.

III. Results

From the whole sample (n = 82), 59.8% (n = 49) were male, with a mean age of 8.58 years (range from 4.00 to 11.92 years). Descriptive statistics for all variables are shown in Table 10.1. No significant differences between groups were found in parental demographic variables (i.e., age, gender, and level of education) or children's demographic variables (i.e., age, and gender). Results showed that there were significant differences between groups in motor skills ($p < .05$) autistic traits ($p < .05$), social support ($p < .001$), and satisfaction on FQoL ($p < .001$). However, there were no differences between groups in vocabulary, receptive language, expressive language, intelligence, social communication, or importance on FQoL (all with $p > .05$). Although there were no differences between groups, we conduced bivariate Pearson's correlations to examine the relation between potential predictors and the satisfaction on FQoL (see Table 10.2).

Table 10.1. Descriptive analysis of Sib-ASD and Sib-TD groups

	Groups		Analysis		
	Sibs-ASD (n = 43)	Sibs-TD (n = 39)	Coeff	*p*	Effect size
Parents' age	35.14 (5.33)	35.51 (6.14)	.087	.769	.001
Parents' gender			.248	.618	.003
Male	5 (12%)	6 (15%)	–	–	–
Female	38 (88%)	33 (85%)	–	–	–
Parents' education			.157	.366	.112
College	30 (70%)	30 (77%)	–	–	–
Some college	2 (5%)	-	–	–	–
High school	11 (25%)	9 (23%)	–	–	–
Children's age	8.22 (2.28)	8.97 (2.02)	2.471	.120	.030
Children's gender			.584	.445	.011
Male	24 (56%)	25 (64%)	–	–	–
Female	19 (44%)	14 (36%)	–	–	–
Receptive vocabulary	75.86 (26.58)	69.59 (29.25)	1.020	.316	.013
Receptive language	44.02 (27.50)	48.10 (28.74)	.426	.516	.005
Expressive language	58.81 (28.47)	67.08 (25.87)	1.861	.176	.023
Motor skills	48.13 (32.61)	60.69 (22.84)	4.005*	.049	.048
Intelligence	107.91 (13.22)	103.13 (9.40)	3.493	.065	.042
Social communication	5.40 (4.86)	3.82 (2.77)	3.165	.079	.038
Severity of ASD	45.51 (26.11)	31.31 (20.86)	7.307*	.008	.084
Social support	6.26 (1.47)	8.67 (1.13)	68.485*	<.001	.461
FQoL importance	22.65 (2.61)	23.01 (1.47)	.590	.445	.007
FQoL satisfaction	18.61 (2.62)	20.47 (2.18)	12.078*	<.001	.131

Effect sizes were computed with eta squared and Cohen's d
* = p<.05

Table 10.2. Correlations between potential predictors and satisfaction on FQoL

	1	2	3	4	5	6	7	8
1. Vocabulary	–	.267*	.416*	.043	-.008	.202	-.042	.077
2. Receptive language	.267*	–	.161	.122	.001	.041	.092	.087
3. Expressive language	.416*	.161	–	.139	.013	-.148	.090	.214
4. Motor skills	.043	.122	.139	–	-.036	-.048	.117	.114
5. Social communication	-.008	.001	.013	-.036	–	.225*	-.127	.030
6. Autistic traits	.202	.041	-.148	-.048	.225*	–	-.158	-.191
7. Social support	-.042	.092	.090	.117	-.127	-.158	–	.417 *
8. Satisfaction on FQoL	.077	.087	.214	.114	.030	-.191	.417*	–

* $p < .05$

We conducted a multiple linear regression analysis with satisfaction on FQoL as outcome variable. Social support, which was highly correlated with satisfaction on FQoL, and the group were included as potential predictors. Vocabulary, receptive language, expressive language, motor skills, social communication, and autistic traits were not included in the analysis because these variables were not related to the outcome (see Table 10.2). The multiple linear regression analysis showed that only social support (β = . 317, $p < .05$) had a significant main effect on satisfaction on FQoL. We tested whether the relationship between social support and FQoL varied as a function of group by adding their interaction. The test of moderation including the interaction between social support and group are presented in Table 10.3, including standardized regression coefficients (*β*s) for each predictor. Results showed that the interaction between group and social support was significant (β = .827, $p < .05$). The completed model accounted for 23% of the total variance [$F(3, 78) = 7.594$, $p < .005$].

Table 10.3. Linear regression analyses to determine the influence of each predictor on satisfaction on FQoL.

				95% CI	
	β	t	*p*	LLCI	ULCI
R^2 = 19, MSE = 2.354					
Group	-.149	-1.088	.280	-2.163	.634
Social Support	.317*	2.314	.023	.065	.868
R^2 = 23, MSE = 5.337					
Group	-7.238*	-2.188	.032	-13.823	-.654
Social Support	-.064	-.194	.846	-.724	.595
Group x Social Support	.827*	2.001	.048	.004	1.649

* = $p < .05$.

Plot of interactions of the effect of social support on FQoL are shown in Figure 10.1. Results indicate that FQoL was particularly low in Sibs-ASD who had relatively low social support. In contrast, FQoL was higher in those Sibs-ASD who have relatively high social support and in Sibs-TD, regardless of their levels of social support. In addition, there was a main effect of group such that social support was lower on average for Sibs-ASD, regardless of their level of social support. Sibs-ASD with lower social support (< 33th percentile) showed a greater reduction in FQoL than those with high social support (> 65th percentile) –a result that is in contrast with that in Sibs-TD, who did not show differences in FQoL as a function of social support (see figure 10.1). To further understand these results, we compared differences between groups in FQoL as a function of level of social support.

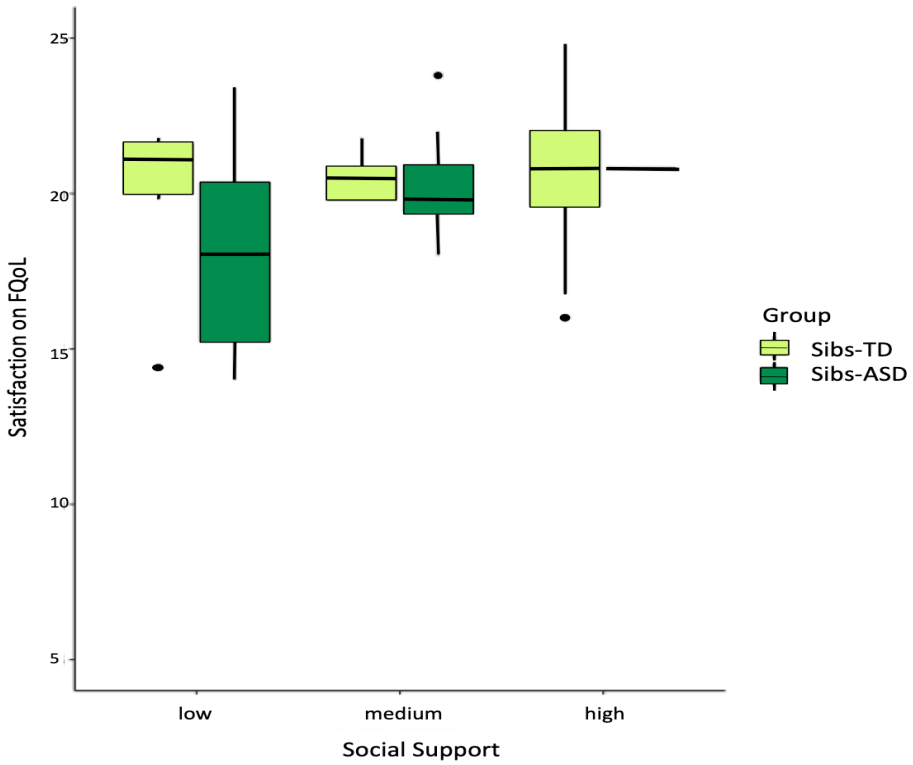


Figure 10.1. A visual representation of the moderation effect of social support (based on terciles) on satisfaction with FQoL by group.

Calculation of simple slopes (see Table 10.4) indicated that the effect of group on FQoL was statistically different at minus one standard deviation from the mean ($p < .05$) on social support (with a medium effect size of d = .50). However, the effects of group on FQoL was not statistically different at mean and plus one standard deviation from the mean (both with $p > .05$).

Table 10.4. Conditional effect of group on FQoL at different values of social support.

					95% CI	
Social Support	Coeff	SE	t	*p*	LLCI	ULCI
5.689	-2.535*	1.122	-2.260	.027	-4.768	-.302
7.439	-1.088	.708	-1.535	.129	-2.498	.323
9.190	.360	.890	.405	.687	-1.411	2.131

*p<.05

IV. Discussion

Research shows that Sibs-ASD are at higher risk for autistic traits and other developmental difficulties than the general population (see Drumm & Brian, 2013). However, research suggests that not all siblings show developmental difficulties (Messinger et al., 2013). Some unaffected Sibs-ASD might show several traits related to ASD or other developmental difficulties, which could be detected by parents. Although these children show a typical development, these features could affect their adjustment and might influenced their family, which would have an impact on their FQoL (Meadan et al., 2010; Szatmari et al., 2016).

We investigated several developmental domains (i.e., language, motor skills, and social communication), traits of ASD, and social support in school-age unaffected Sibs-ASD and Sibs-TD. Moreover, we assessed and described differences in FQoL between groups, and we determined whether FQoL might be explained by differences in social support, developmental domains, or traits of ASD. From all these domains, we have found significant differences between groups in motor skills, severity of autistic traits, satisfaction on FQoL, and social support. However, there were no differences between groups in expressive language skills, intelligence, social communication, or importance on FQoL. Moreover, our results suggest that social support may act as a positive factor protecting the negative effect of having a sibling with ASD on perceptions on satisfaction of quality of life.

These results are in line with previous research showing that during the first years of life siblings of children with ASD may show differences in cognitive, motor, language and/or

social development. However, these results also suggest that these difficulties have not been shown in older Sibs-ASD (Bedford et al., 2012; Ozonoff et al., 2014). Our results indicated that there were no differences between groups in language abilities, as other authors have found (see Ben-Yizhak et al., 2011; Drumm et al., 2015; Hudry et al., 2014; Warren et al., 2012).

In contrast to other studies (e. g., Pilowsky, Yirmiya, Gross-Tsur, & Shalev, 2007; Pisula & Ziegart-Sadowska, 2015), we did not find social communication and language deficits in Sibs-ASD. Our results indicated that the group of Sibs-ASD showed higher levels of traits of ASD but also similar levels of verbal and nonverbal skills than those in the Sibs-TD group. In addition, our data referring to the language in Sibs-ASD group did not agree with those obtained by Gamliel et al. (2009). In particular, they showed that school-age Sibs-ASD showed significantly more cognitive, linguistic, and parent-reported difficulties compadre to Sibs-TD (performance of at least 1.5 SD below average). One explanation of this result is that we measured all developmental skills through directed measures, not parent-reported scales, which could have an impact on the results.

Regarding social communication, several authors have found deficits in Sibs-ASD (see Pisula & Ziegart-Sadowska, 2015 for a review). Other authors have found that young unaffected Sibs-ASD showed lower overall rates in social communication than Sibs-TD (Bontinck, Warreyn, Van der Paelt, Demurie & Roeyers, 2018). However, Pilowsky et al. (2007) found no differences between groups in social communication, as we did. Some studies have not found differences in language between Sibs-ASD and Sibs-TD during the school years. This fact could be explained because only a subset of Sibs-ASD is characterized by lower scores (Miller et al., 2015).

Although social support is important for all members in families of children with ASD, the literature that relates FQoL, social support and psychological and social adjustments in Sibs-ASD is still scarce and contradictory (Vieira & Fernandes, 2013). It is possible, as our results suggest, that a concrete social support plays an important role in the adaptive adjustment of Sibs-ASD, and their FQoL (Tsao et al., 2012; Kaminsky & Dewey, 2002). Our research adds to this literature showing that siblings of children with ASD and social support deficiencies are also vulnerable and experience lower levels of quality of life. For instance, siblings of children with ASD might show positive results when they report higher levels of

social support (e.g. Cebula, 2012; Tomeny, Barry & Fair, 2017). One plausible explanation may be that siblings of children with ASD usually take on additional responsibilities, thus limiting chances for social and peer interaction, which could affect their social support (Moyson & Roeyers, 2012).

Our research contributes to the growing literature on variables of Sibs-ASD that help families with children with ASD improve as much as families with children with TD. Additionally, our results support the wide heterogeneity that it is described throughout research in school-age Sibs-ASD. Although in this range of age there are not so many studies, it is important to recognize some difficulties that have been found in Sibs-ASD, because these difficulties could be magnified by academic requirements and the importance of relationships with other children. To overcome this challenge, Sibs-ASD would need to receive an adequate social support that will have an impact over their FQoL. Nevertheless, this study has several limitations. First, the small sample size does not allow us to generalize results. Second, the majority of participants in this study were mothers of Sibs-ASD. Therefore, the low participation of parents can be considered as another limitation of the work. Third, the Structural Social Support questionnaire may not be a complete measure of social support. Thus the effects of social support found in the present analysis may be considered by limitations of this measure. Finally, having a child with ASD could influence the information reported by parents. Thus, future studies could incorporate opinions from older Sibs-ASD related to FQoL and assess if there are discrepancies between parents and children's opinions.

From our results, several clinical implications could be suggested. First, we support the importance of planning interventions focus on improving social support in Sibs-ASD. Second, perceived social support may serve as potential point of intervention for reducing distress and improving the satisfaction with their FQoL. Finally, differences found between Sibs-ASD and Sibs-TD should be taken into account when making decisions about how to support siblings and what kind of interventions is appropriate for each member of the family. In conclusion, these findings highlight the variability in the developmental abilities of the unaffected school-age children with familiar risk factors and reinforce the need for monitoring development of all Sibs-ASD over multiple time points, as stated Szatmari et al. (2016); not only until the first 3 years of life, but along school years.

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PART V

GENERAL DISCUSSION

CHAPTER 11

Chapter 11
General discussion, conclusions, and clinical implications

I. Discusión general

El principal objetivo de esta tesis ha sido el de contribuir a la Psicología del Desarrollo, la Psicología Cognitivo–Conductual y la Logopedia investigando el impacto que diferentes variables lingüísticas y psicológicas pueden ejercer sobre la detección y el diagnóstico del TEA y la calidad de vida familiar (CdVF) en niños con trastorno del espectro autista (TEA). Para ello, hemos investigado la influencia de diversas variables a lo largo de los nueve capítulos que forman las tres partes de esta tesis, que cubren un amplio periodo desde el nacimiento hasta la adolescencia. Hemos investigado qué señales tempranas y variables lingüísticas (vocalizaciones) están relacionadas con una detección temprana y/o un diagnóstico temprano de TEA (**parte II**). También hemos ampliado la literatura relacionada con las habilidades socio-lingüísticas en el TEA (lenguaje estructural expresivo y comprensivo, comunicación social y comunicación no verbal) y hemos estimado el impacto de la adaptación psicológica, el lenguaje y la comunicación no verbal sobre la CdVF en TEA (**parte III**). Finalmente hemos analizado el impacto que diversas variables procedentes de los padres (participación en una intervención y habilidades numéricas) y de los hermanos (habilidades lingüísticas, motrices y comunicación social) ejercen sobre la CdVF en TEA (**parte IV**). Las principales variables analizadas en estas partes se representan e integran en el modelo que aparece a continuación (véase Figura 11.1)

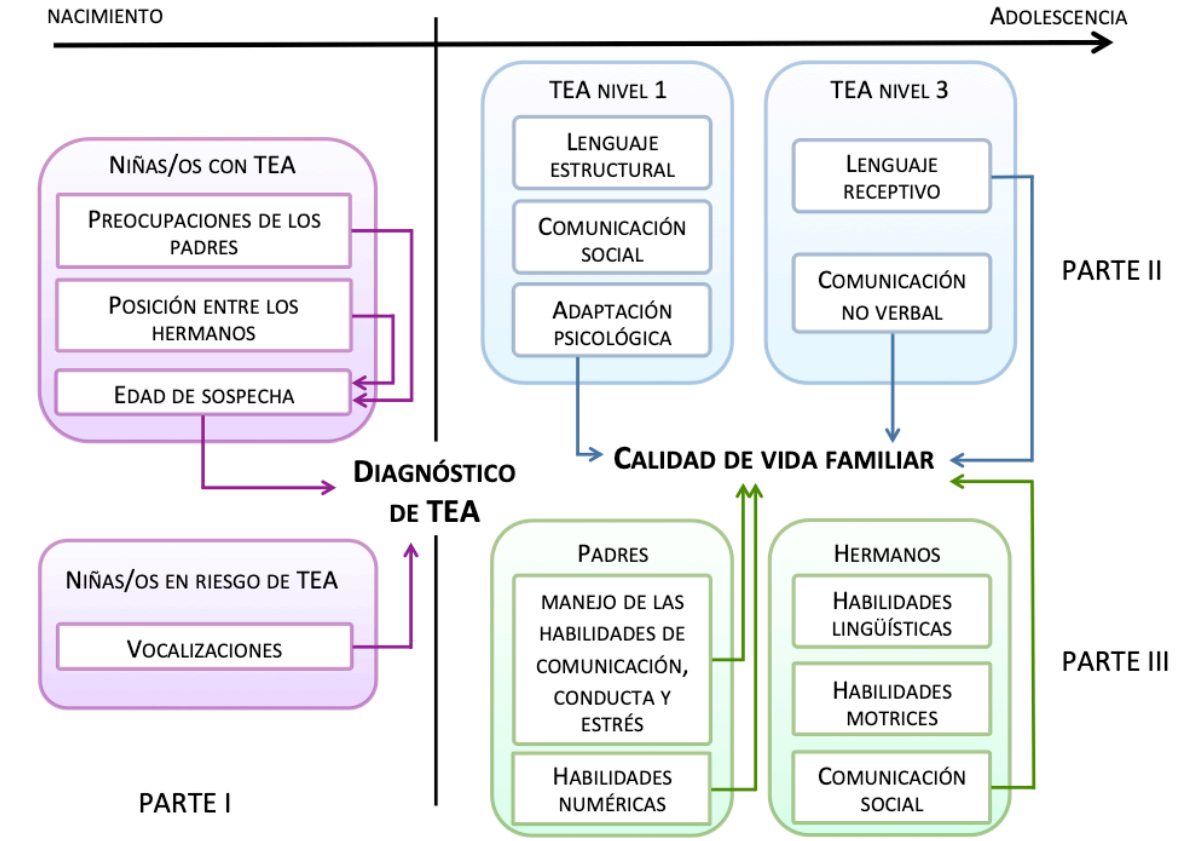

Figura 11.1. Principales variables analizadas en las partes de la tesis

Como mencionamos en la introducción de esta tesis, recibir un diagnóstico lo más temprano posible es esencial para que las familias de niños con TEA puedan acceder a los programas de atención temprana. Desafortunadamente, aunque es el núcleo familiar el que alerta al pediatra en primer lugar de un posible trastorno en el desarrollo, no hay un claro consenso acerca de qué señales son las que tienen un impacto significativo a la hora de reconocer de forma temprana el TEA (Guinchat et al., 2012). Por ello, en esta tesis hemos profundizado en el conocimiento de los factores que están relacionados tanto con la detección como el diagnóstico en TEA y hemos ofrecido algunas de las claves para realizar una detección y un diagnóstico tempranos en TEA (**capítulos 2 y 3**).

Específicamente, hemos identificado diversos factores que pueden acelerar la sospecha del TEA y que pueden interferir, y o alargar en el tiempo la espera entre la sospecha y el diagnóstico formal (**capítulo 2**). Además, en este estudio hemos propuesto un modelo explicativo de la demora del diagnóstico en TEA. En concreto, nuestros resultados informan que la sospecha del TEA por parte de los padres se acelera si los

padres tienen un hijo mayor con desarrollo típico (DT). Sin embargo, esta relación no es lineal, ya que está mediada parcialmente por el tipo de preocupación que expresan los padres. En concreto, las preocupaciones relacionadas con aspectos socio-comunicativos (dificultades o ausencia de sonrisa social, respuesta al nombre, reacciones a la elicitación social, atención conjunta, señalar, contacto ocular e imitación) aceleran la sospecha del TEA, tal y como otros autores señalan (Volkmar, Chawarska, & Klin, 2008; Zwaigenbaum et al., 2005). Sin embargo, nuestros resultados no muestran que otras preocupaciones relacionadas con aspectos de conductas estereotipadas y repetitivas (resistencia al cambio, estereotipias, aleteo de manos], otras preocupaciones del desarrollo no relacionadas con TEA (llanto excesivo, retraso en la marcha, problemas de sueño o alimentación) o las variables socio-demográficas (nivel educativo paterno, edad de los padres y género del niño) afecten a esta relación.

Adicionalmente a las señales que promueven detectar antes el TEA, el tiempo de espera entre la sospecha y el diagnóstico formal es fundamental para acelerar el acceso a los programas de atención temprana. En este sentido, hemos encontrado que la edad de sospecha actúa como mediador entre las preocupaciones de los padres y el tiempo de espera entre la sospecha y el diagnóstico formal. En concreto, nuestros resultados muestran que cuando las consultas acerca del desarrollo se hacen a una edad más temprana, el tiempo que transcurre entre la sospecha y el diagnóstico es mayor. Nuestros datos apoyan los resultados de otros autores que sugieren que cuando los pediatras reciben consultas acerca de niños muy pequeños prefieren adoptar la postura de esperar y ver qué ocurre con el desarrollo del niño (*wait-and-see*). Una posible explicación detrás de esta posición es el conocimiento de que cada niño tiene su propia trayectoria de desarrollo (Klin, Klaiman, & Jones, 2015), lo que podría explicar la demora entre la sospecha y el diagnóstico en TEA (Daniels & Mandell, 2014; Guinchat et al., 2012).

A lo largo de la investigación del TEA, las diferencias en los aspectos socio-comunicativos en el desarrollo no se aprecian antes de los primeros 6 meses de vida (Meek, Robinson, & Jahromi., 2012; Ozonoff et al., 2010; Paul, Fuerst, Ramsay, Chawarska, & Klin, 2011). Sin embargo, aunque la literatura detecta diferencias a una edad temprana, es alrededor del segundo año de vida cuando entre el 80-90% de las señales tempranas son detectadas por los padres (Volkmar et al., 2008; capítulo 2). Por ello, hemos

contribuido a la descripción y evaluación de señales socio-comunicativas y lingüísticas que son fácilmente detectables por los padres antes de los 2 años de vida que potencialmente puedan predecir un diagnóstico de TEA (**capítulo 3**). En este sentido, en este capítulo hemos mostrado que existen diferencias en las vocalizaciones a los 14 meses entre niños que reciben un diagnóstico de TEA, niños que no reciben un diagnóstico de TEA pero se sitúan en el espectro y niños que no tienen un diagnóstico de TEA a los 23 meses.

En concreto, nuestros resultados relacionados con las vocalizaciones analizadas, son consistentes con los informados por Ozonoff et al. (2010) y Winder, Wozniak, Parlade e Iverson, (2013), mostrando que es la intencionalidad comunicativa (es decir, aquellas vocalizaciones que ocurren en un contexto interactivo y que se consideran dirigidas hacia alguien), y no la complejidad de las vocalizaciones, la que marca la diferencia en el desarrollo de los niños con TEA. En suma, los niños con TEA producen menos vocalizaciones con intención comunicativa que los niños que no tienen TEA independientemente de la complejidad de las vocalizaciones (vocalizaciones que incluyen al menos una sílaba completa con la estructura Consonante-Vocal vs. vocalizaciones relacionadas con el habla que no incluyen ninguna sílaba completa con la estructura Consonante-Vocal).

Tras recibir un diagnóstico de TEA, las familias deben hacer frente a multitud de situaciones nuevas y deben adaptarse a las necesidades de sus hijos para potenciar sus capacidades y trabajar las dificultades. En este sentido, la CdVF puede verse afectada y disminuida (Cohen, Holloway, Dominguez-Pareto, & Kuppermann, 2014; Vasilopoulou & Nisbet, 2016). Sin embargo, dada la especial naturaleza del trastorno, en el que fenotípicamente aparece una gran heterogeneidad, se hace indispensable tener en cuenta los diversos niveles de apoyo que acompañan al trastorno (en el ámbito del lenguaje y la comunicación social) para poder ofrecer ayudas específicas en función del nivel de severidad del TEA. Por ello, en la tercera parte hemos ofrecido claves lingüísticas y socio-comunicativas que mejoran la CdVF en TEA teniendo en cuenta el nivel de severidad del TEA.

Específicamente, hemos identificado algunos aspectos diferenciales del lenguaje estructural (expresión y comprensión), comunicación e interacción social (aspectos

pragmáticos), adaptación social (síntomas emocionales, hiperactividad, conducta prosocial y problemas con los compañeros) y su relación con la CdVF en niños de edad escolar con TEA nivel de apoyo 1 (**capítulos 4 y 5**). En concreto, nuestros resultados sugieren que de forma global no existen diferencias significativas en el lenguaje estructural comprensivo (comprensión de estructuras gramaticales) y expresivo (a nivel sintáctico y semántico). Sin embargo, aunque nuestros resultados apoyan la hipótesis de que los niños con TEA nivel de apoyo 1 se consideran gramaticalmente fluidos (Paynter & Peterson, 2010), sí hemos detectado algunas diferencias sutiles que son coherentes con las detectadas por otros autores, como las dificultades en la interpretación de pronombres reflexivos (Perovic, Modyanova, & Wexler, 2013) y que podrían ser útiles a la hora de planificar las intervenciones con esta población (**capítulo 4**).

Por otro lado, los resultados relacionados con la comunicación y la interacción social (como iniciación inadecuada, lenguaje estereotipado, contexto, comunicación no verbal, relaciones sociales e intereses) muestran que sí existen diferencias significativas entre el grupo de niños con TEA nivel de apoyo 1 y el grupo de niños con DT, afirmando los resultados encontrados por Geurts et al. (2004) y Helland (2014). En cuanto a la adaptación social, nuestros resultados sugieren que existen dificultades en el grupo de niños con TEA nivel de apoyo 1 en los aspectos relacionados con síntomas emocionales, hiperactividad, problemas con los compañeros y conducta prosocial, lo que podría explicar en parte la inhibición social típica de estos niños con su entorno (Volden & Phillips, 2010).

Dada la importancia de la adaptación social y las posibles repercusiones en el ámbito social que se plantean a nivel teórico, hemos profundizado en el estudio de la conducta adaptativa en niños con TEA nivel de apoyo 1 y además hemos evaluado su potencial papel en la satisfacción percibida en la CdVF (**capítulo 5**). En este sentido, nuestros resultados arrojan luz sobre la capacidad predictiva y protectora de la conducta adaptativa sobre la CdVF, concordando parcialmente con los resultados obtenidos por otros autores como Eapen y Guan (2016) o McStay, Trembath y Dissanayake (2014). Sin embargo, aunque en nuestro trabajo tener TEA se asocia con un riesgo elevado de presentar problemas de conducta, hiperactividad, dificultades con los compañeros y dificultades emocionales, no todas las variables evaluadas muestran tener dicho valor

predictivo. En concreto, aquellos niños con TEA nivel de apoyo 1 que obtienen una puntuación más alta en conducta prosocial, presentan una mayor satisfacción en CdVF.

Sin embargo, en la población de niños con TEA, no todos tienen lenguaje verbal. Alrededor del 25% muestra limitaciones comunicativas severas y se consideran niños con TEA no verbales (Luyster, Kadlec, Carter, & Tager–Flusberg, 2008). Dada la alta incidencia dentro de esta población de problemas comunicativos severos y la escasa literatura que la estudia, hemos ampliado el conocimiento de las variables lingüísticas que impactan sobre la CdVF en TEA evaluando el lenguaje comprensivo y la comunicación no verbal en niños con TEA nivel de apoyo 3 (**capítulo 6**). Nuestros resultados muestran que la comprensión del lenguaje es especialmente vulnerable en esta población.

De nuestros resultados también se desprende la existencia de dificultades en la comprensión de órdenes verbales (como el seguimiento de órdenes sencillas y complejas), en la comprensión de estructuras gramaticales (especialmente aquellas que no siguen un orden canónico) y la comunicación no verbal, tal y como otros autores afirman (Kjellmer et al., 2012; Rapin & Dunn, 2003; Swensen, Kelley, Fein, & Naigles, 2007). Además, otra aportación de este trabajo es que nuestros resultados ponen de manifiesto que las diferencias en la satisfacción con la CdVF parecen estar ligadas, en parte, con las diferencias en el nivel de comprensión y comunicación no verbal de los niños con TEA nivel de apoyo 3, apoyando los resultados de Park, Yelland, Taffe y Gray (2012).

No obstante, cuando hablamos de CdVF además de los niños con TEA, influyen otros agentes adicionales del entorno familiar más cercano (padres y hermanos). Dado que en la literatura de los últimos años se ha fomentado un acercamiento más comprensivo de la CdVF en el TEA (Vasilopoulou & Nisbet, 2016), en esta tesis hemos evaluado factores protectores que suavizan el impacto del TEA en los familiares más cercanos a los niños con TEA (parte IV; **capítulos 5, 6, 7, 8 y 10**).

Debido a los resultados derivados de esta tesis, en los que hemos constatado que las habilidades lingüísticas y socio-comunicativas están implicadas en la satisfacción de la CdVF (**capítulos 5 y 6**), nos propusimos promover una mejora en estas habilidades en los niños con TEA nivel de apoyo 3 a través de una intervención mediada por padres (**capítulo 7**). Así, tras una intervención breve de 6 semanas, éstos mejoran tanto su satisfacción en la

CdVF como sus habilidades para incrementar la comunicación con sus hijos. En concreto, la mejora en la CdVF se concentra en tres dominios: interacción familiar, papel de padres y bienestar emocional, lo que podría estar relacionado con un incremento en su sentimiento de competencia y su confianza como padres (Ayuda–Pascual, Llorente-Comí, Martos-Pérez, Rodríguez-Basua, & Olmo-Remesa, 2012; Feinberg et al., 2014; Iadarola et al., 2017).

En relación con las habilidades paternas, los resultados muestran que los padres que participan en la intervención mejoran de forma significativa en el manejo de las estrategias lingüístico-comunicativas dirigidas a su hijo (como hablarle en función del nivel de comprensión, valorar cada uno de sus intentos comunicativos, imitar acciones y vocalizaciones de su hijo, responder rápido a estos actos comunicativos y esperar su respuesta sin adelantarse a ella) como otros autores también han demostrado (Matson, Mahan, & Matson, 2009). Sin embargo, aunque los padres muestran mejoras significativas en el manejo de las estrategias relacionadas con el control de la conducta (como responder rápido a las conductas de su hijo, ofrecer refuerzo diferencial y ser contingente tras cada conducta de su hijo), éstas no se mantienen a lo largo del tiempo. Tampoco aparecen diferencias significativas en las estrategias de control del estrés parental (como reconocer las emociones positivas y negativas, evitar las rumiaciones de pensamiento y valorar el esfuerzo personal como padre/madres).

De forma paralela y en línea con el hilo argumental y las aportaciones de esta tesis, hemos evaluado la existencia de otras características personales en los padres que pueden suavizar el impacto del TEA sobre la CdVF (**capítulo 8**). En concreto nuestros resultados muestran por primera vez en una muestra de padres de niños con TEA, que las habilidades numéricas afectan a la CdVF, como se ha constatado en otros trabajos con otras poblaciones (Cokely, Galesic, Schulz, Ghazal, & Garcia-Retamero, 2012). Particularmente, las habilidades numéricas objetivas, pero no las subjetivas, actúan como un factor positivo que suaviza el efecto negativo de tener un hijo con TEA en la satisfacción de la CdVF. Además, este efecto se mantiene aun controlando diversas variables que podrían influir en la CdVF como la severidad del TEA, el nivel educativo de los padres y el apoyo social percibido. Específicamente, aquellos padres de niños con TEA con altas habilidades

numéricas, presentan una satisfacción en la CdVF tres veces mayor que la expresada por familias con bajas habilidades numéricas.

Del mismo modo que algunas características lingüísticas, socio-comunicativas y psicológicas de los propios niños con TEA (**capítulos 5 y 6**) y sus padres (**capítulos 7 y 8**) son específicas y afectan a la CdVF, existen diferencias en el desarrollo temprano de los hermanos de niños con TEA y algunas de estas dificultades ejercen también una influencia sobre la CdVF (**capítulos 9 y 10**). Se estima que alrededor del 25% de los hermanos de niños con TEA presentan algunas características relacionadas con los criterios diagnósticos del TEA que podrían ejercer una influencia en el seno familiar (Georgiades et al., 2013; Messinger et al., 2013). Dado la potencial influencia que también pueden ejercer los hermanos, hemos evaluado aquellas características del desarrollo (lingüísticas, motrices y psicológicas) que pueden ser diferenciales con respecto a hermanos de niños con DT (**capítulos 9 y 10**).

Por este motivo, hemos estimado las diferencias en las habilidades lingüísticas y motrices que aparecían en los hermanos de niños con TEA en los primeros tres años de vida (**capítulo 9**). Así, nuestra revisión meta-analítica muestra que los niños que tienen un hermano mayor con TEA tienen peores habilidades lingüísticas (expresión y comprensión) y peores habilidades motrices (finas). Lamentablemente, no encontramos un número suficiente de trabajos que evaluaran las habilidades motrices gruesas en los hermanos entre 0 y 3 años, para poder establecer conclusiones acerca de esta habilidad en la revisión meta-analítica realizada. Por otro lado, nuestros resultados sugieren que las diferencias analizadas en las habilidades lingüísticas se detectan durante el primer año de vida y son dos veces más grandes que las diferencias detectadas en las habilidades motrices. Además, estas diferencias se mantienen hasta los tres años de edad.

Sin embargo, la literatura existente que evalúa a hermanos de niños con TEA se ha centrado en edades tempranas (Gowen & Hamilton, 2013; Mulligan & White, 2012). Por este motivo, hemos contribuido al conocimiento de las trayectorias en el desarrollo de los hermanos de niños con TEA. En concreto, hemos evaluado qué sucedía con las habilidades lingüísticas, socio-comunicativas y motrices, los rasgos de TEA y el apoyo social en los hermanos de niños con TEA en edad escolar y su potencial relación con la CdVF (**capítulo**

10). Nuestros resultados sugieren que no hay diferencias significativas en las habilidades lingüísticas evaluadas (a nivel expresivo y comprensivo), pero sí las había en las habilidades motrices (finas y gruesas), los rasgos subclínicos de autismo, el apoyo social y la satisfacción en la CdVF entre los hermanos de niños con TEA y los hermanos de niños con DT, tal y como otros autores han sugerido (Drumm, Bryson, Zwaigenbaum, & Brian, 2015; Hudry et al., 2014; Messinger et al., 2013; Provost, Lopez, & Heimerl, 2007).

Por otro lado, nuestros resultados muestran que aquellos hermanos que presentan un mayor apoyo social, presentan un nivel alto de satisfacción en la CdVF. Nuestros datos sugieren que el apoyo social puede servir como un elemento potencial para intervenir en la mejora de la CdVF.

II. Conclusiones

Del trabajo global presentado en esta tesis doctoral se puede extraer las siguientes conclusiones generales:

- Los padres de niños con TEA detectan las señales del trastorno antes si tienen un hijo mayor con DT. Además, las señales relacionadas con los aspectos lingüísticos y socio-comunicativos son las variables más determinantes en la detección. Sin embargo, aquellos padres que detectan antes las señales, esperan más tiempo para recibir un diagnóstico.

- Aunque no existe ningún tipo de vocalización que actúe como un marcador sensible del TEA de forma aislada, examinar la cantidad y la calidad de las vocalizaciones (intencionalidad) junto con otras señales conductuales podrían ser útiles a nivel clínico para predecir un diagnóstico de TEA de forma temprana.

- Los niños con TEA nivel de apoyo 1 presentan un perfil lingüístico con una buena expresividad general tanto semántica como sintáctica. No obstante, también presentan dificultades en comprensión de estructuras gramaticales y en habilidades pragmáticas que podrían estar relacionados con los problemas emocionales y la socialización.

- Los niños con TEA nivel de apoyo 1 muestran dificultades significativas en la conducta adaptativa (síntomas emocionales, problemas de conducta, hiperactividad/falta de atención, problemas de relaciones entre compañeros y conducta prosocial) comparados con niños con DT.

- La conducta prosocial y la comunicación no verbal y la comprensión lingüística predicen en parte la satisfacción en la CdVF en los niños con TEA nivel de apoyo 1 y en los de nivel de apoyo 3.

- Los niños con TEA nivel de apoyo 3 presentan niveles por debajo de los esperado en vocabulario receptivo, comprensión auditiva y comprensión gramatical. Asimismo, la CdVF se ve afectada por los problemas lingüísticos de los niños con TEA de este nivel de apoyo.

- Participar en una intervención breve mediada por padres de niños con TEA, favorece mejoras tanto en el conocimiento sobre el TEA, las estrategias de manejo del lenguaje y la comunicación de sus hijos. También mejoran las estrategias en el manejo conductual y la satisfacción percibida en la CdVF, aunque no se mantuvieron en el tiempo.

- Las habilidades numéricas objetivas (independientemente de otras variables como las habilidades numéricas subjetivas, el apoyo social, el género, el nivel educativo y la severidad del TEA) suavizan el impacto del TEA en la CdVF.

- Los hermanos, de niños con TEA, menores de 3 años presentan peores habilidades lingüísticas y motrices comparados con los hermanos, de niños con DT, menores de 3 años. Estas diferencias aparecen en el primer año de vida y parece que se mantienen hasta el tercer año. Además, las dificultades en habilidades lingüísticas son mayores y se detectan antes que las dificultades en habilidades motrices.

- Los hermanos de niños con TEA en edad escolar no presentan dificultades en habilidades lingüísticas. Sin embargo, sí aparecen diferencias en habilidades motrices, apoyo social, rasgos de autismo y satisfacción en CdVF comparados los hermanos de niños con DT. Además, mostrar niveles altos de apoyo social predice una mejor CdVF.

III. Implicaciones clínicas

De esta tesis doctoral se desprenden una serie de implicaciones que podrían facilitar la práctica clínica de los profesionales que trabajan con niños con TEA y sus familias. En primer lugar, en relación con la detección temprana del TEA y su diagnóstico, los padres que sospechan antes un posible TEA detectan más síntomas relacionados con el lenguaje y la comunicación y tienen hijos mayores. Sin embargo, cuando estos padres comparten sus preocupaciones con los pediatras, a menudo reciben el consejo de esperar y ver qué ocurre con el desarrollo de sus hijos. En este sentido, los pediatras deberían prestar una mayor atención a aquellos síntomas que informan los padres, especialmente cuando se dan bajo ciertas circunstancias (como tener otro hijo mayor y expresar preocupaciones relacionadas con el lenguaje y la comunicación) para poder promover diagnósticos tempranos y el acceso a la atención temprana. Consideramos que informar adecuadamente a los pediatras sobre las señales específicas del TEA y la forma de responder a las preocupaciones de los padres podría mejorar y disminuir el tiempo de demora para recibir un diagnóstico de TEA.

Profundizar en un mayor conocimiento de las señales tempranas que a nivel lingüístico y comunicativo pueden detectar los padres alrededor del primer año de vida, ayudaría a acelerar la detección del TEA. Además, estas claves pueden ser útiles a nivel clínico y guiar las preguntas de los pediatras ante una consulta de un posible trastorno del neurodesarrollo. Específicamente, aunque no detectamos ningún marcador lingüístico exclusivo que identifique un posterior diagnóstico de TEA, parece que es la calidad de las vocalizaciones (es decir, la falta de intencionalidad comunicativa) y no tanto la complejidad de las mismas, las que pueden contribuir a la detección del TEA de forma temprana. Por tanto, nuestros resultados también ofrecen señales fácilmente detectables por personas que no son especialistas en el campo de la psicología o la logopedia.

En segundo lugar, hemos comprobado la importancia de diversas variables lingüísticas y psicológicas en niños con TEA sobre la CdVF. En concreto, los niños con TEA nivel de apoyo 1, muestran dificultades en las habilidades pragmáticas. Por ello se hace indispensable que los profesionales que trabajan con estos niños lo hagan, entre otras, haciendo hincapié en la mejora de estas habilidades. Además, nuestros resultados

subrayan la importancia del desarrollo de la pragmática en diferentes entornos, no solo dentro del núcleo familiar. Por ejemplo, dado que los niños con TEA nivel de apoyo 1 tienden a ser integrados en clases ordinarias, las dificultades del lenguaje detectadas también podrían afectar a la calidad de las relaciones con sus compañeros en el entorno escolar.

Del estudio de la conducta adaptativa se derivan implicaciones para el tratamiento de esta habilidad en niños con TEA nivel de apoyo 1. De manera específica, el TEA de este nivel de apoyo se asocia con un riesgo elevado de dificultades en diversas variables que están directamente relacionadas con la conducta adaptativa (como problemas de conducta, hiperactividad, dificultades con los compañeros y dificultades emocionales). Además, la conducta prosocial predice en parte la satisfacción en la CdVF percibida. Por tanto, prestar especial atención en las intervenciones a aquellas variables relacionadas con las habilidades prosociales, podría tener un efecto directo tanto sobre el funcionamiento adaptativo en los niños con TEA como sobre la CdVF.

Otras implicaciones clínicas en relación a los niños con TEA, nivel de apoyo 3, afectan a la comprensión del lenguaje (a nivel gramatical, lenguaje no verbal y comunicación social). Aunque el deterioro de la comprensión no es un criterio diagnóstico, sí debe tenerse en cuenta en la intervención. Además, la mejora en estas habilidades también implicaría una mejora en la satisfacción de la CdVF, dado que las diferencias en el nivel de lenguaje comprensivo y de comunicación de los niños con TEA nivel de apoyo 3 y DT explican las diferencias en el nivel de CdVF en estas familias.

En tercer lugar, en relación a las variables de los familiares de primer grado de niños con TEA que explican la CdVF también se infieren importantes implicaciones para la práctica clínica. En concreto, tras la participación en una intervención mediada por padres, se describieron mejorías en la satisfacción percibida en la CdVF (interacción familiar, papel de padres y bienestar emocional) y en el manejo de la comunicación e interacción padres-hijos y el manejo conductual. Los padres de niños con TEA son capaces de adquirir, en un breve periodo de tiempo, algunas estrategias para mejorar las habilidades de sus hijos. Además, esta intervención también puede tener un efecto positivo transversal en la motivación de los padres para involucrarse en las intervenciones con sus hijos. De forma

global, la principal implicación clínica de este estudio radica en ofrecer algunas claves para identificar intervenciones breves que parecen ser especialmente prometedoras para mejorar el bienestar de las familias de niños con TEA.

Además de los beneficios comentados tras la participación en una intervención mediada por padres, se han mostrado variables individuales que pueden afectar a la CdVF. En concreto, un adecuado nivel de habilidades objetivas numéricas puede ayudar a las familias de niños con TEA a percibir una mayor satisfacción en su CdVf. Una de las implicaciones de este trabajo radica en un posible efecto de las habilidades numéricas objetivas en la reducción de la carga de información que los padres tienen que integrar a la hora de tomar decisiones y ayudar en el manejo de la condición de su hijo. Por lo tanto, nuestros resultados contribuyen al crecimiento en el conocimiento de las características que hacen a los padres de niño con TEA más resilientes.

La revisión meta-analítica de los hermanos de niños con TEA menores de 3 años muestra que aunque no existe un patrón homogéneo y estable de dificultades lingüísticas y motrices, sí existen algunos aspectos atípicos en el desarrollo del lenguaje y la motricidad. En consecuencia, vigilar el desarrollo de los hermanos de niños con TEA es fundamental para asegurar el progreso de las habilidades lingüísticas y motrices y trabajar con las dificultades que aparecen en esta población.

En los hermanos de niños con TEA en edad escolar, hemos subrayado la importancia de planificar intervenciones centradas en la mejora del apoyo social. Aunque aproximadamente solo el 20% de los casos los hermanos de niños con TEA presentan el diagnóstico, el entorno familiar donde se desarrollan puede tener consecuencias en sus habilidades y la CdVF. Adicionalmente, las diferencias individuales que se detectaron entre los hermanos de niños con TEA (como los rasgos de autismo) también se deberían tener en cuenta cuando se toman decisiones sobre la mejora en el apoyo social.

En suma, las implicaciones planteadas pueden potenciar, en los diferentes profesionales que trabajan con niños con TEA y sus familias, el diseño de los objetivos de la detección temprana del TEA y las intervenciones para mejorar la CdVF, especialmente aquellas destinadas a la intervención con niños con TEA, sus padres y sus hermanos.

En investigaciones futuras sobre población con TEA se debería hacer hincapié en la evaluación de otros factores lingüísticos y comunicativos adicionales a la hora de detectar el TEA. También se debería profundizar en las dificultades gramaticales a nivel escrito y en las habilidades de lenguaje oral (como las habilidades pragmáticas), ya que podrían estar relacionadas con las dificultades descritas en esta tesis doctoral en comprensión gramatical oral y lenguaje expresivo. Además, futuros estudios deberían estudiar otras variables individuales de los familiares de primer grado que puedan suavizar el impacto del TEA en la familia y los mecanismo que pueden implicar tener una mayor satisfacción en su CdVF.

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INTERNATIONAL DOCTORATE

The main goal of this thesis was to contribute to Developmental Psychology, Cognitive Behavioral Psychology, and Speech Science investigating the impact of several linguistic and psychological factors on detection and diagnosis of autism spectrum disorder (ASD), and family quality of life (FQoL). For this purpose, we have investigated the influence of several variables throughout nine chapters, which integrate the three parts of this thesis. We have covered an extended period of time from childbirth to adolescence. Specifically, we have investigated to what extent early signs and linguistic variables (i.e., vocalizations) were related to an early detection and/or an early diagnosis of ASD (**Part II**). Moreover, we have expanded the literature related to socio-linguistic skills in ASD (expressive and comprehensive language, social communication, and nonverbal communication), and we have estimated the impact of psychological adaptation, language, and nonverbal communication on FQoL in ASD (**Part III**). Finally, we have analyzed the impact of different variables from parents (i.e., participating in an intervention, and numeracy skills) and siblings (i.e., linguistic skills, motor skills, and social communication) on FQoL in ASD (**Part IV**).

I. Conclusions

We can draw the following conclusions from the results from research included in this thesis:

- Parents detect early signs related to ASD if they have an older child with typical development (TD). Moreover, those symptoms related to linguistic and socio-communicative aspects are crucial in early detection. However, parents who report symptoms earlier have to cope with a longer time lag until they receive a formal diagnosis.

- Research suggests that there is no vocalization type that acts as a sensitive marker of ASD on its own. However, the quantity and quality of early vocalizations (i.e., intention) combined with other early behavioral features may be useful for clinicians in making an early diagnosis of ASD.

- Children with ASD level of support 1 demonstrate a linguistic profile with good general expressive structure both at semantic and syntax levels. Nevertheless, these children show difficulties related to comprehension of grammar structures, and social pragmatics which could be related to emotional and social problems.

- Children with ASD level of support 1 show more difficulties in adaptive behavior (i.e., emotional symptoms, behavioral problems, hyperactivity/lack of attention, problems with peers, and prosocial behavior) compared to children with TD.
- Prosocial behavior, nonverbal communication, and linguistic comprehension predict satisfaction on FQoL in children with ASD level of support 1 and level of support 3.
- Children with ASD level of support 3 show lower levels of receptive vocabulary, auditory comprehension, and grammar comprehension than children with TD. Furthermore, FQoL is influenced by language problems of children with this level of support.
- A brief parent mediated intervention can lead to improvement in knowledge related to ASD, parental management on strategies related to language and communication skills, as well as improvements in their satisfaction on FQoL.
- Objective numeracy skills buffer the impact of ASD on FQoL regardless of other variables such as subjective numeracy skills, social support, gender, education, and ASD severity.
- Under 3-year-old siblings of children with ASD (HR) have less adequate linguistic and motor skills compared to under 3-year-old siblings of children with TD (LR). These differences are detectable during the first year, and tend to remain until they are three years old. Moreover, linguistic difficulties are larger and could be detected earlier than motor difficulties.
- School-age siblings of children with ASD (Sibs-ASD) show no differences on general expressive language skills. However, they show differences on motor skills, social support, traits of autism, and satisfaction on FQoL compared to school-age siblings of children with TD (Sibs-TD). Additionally, those children who show higher levels of social support show better FQoL.

II. Clinical implications

The research included in this doctoral thesis can have a number of implications which could facilitate the clinical practice of those professionals who work with children affected by ASD as well as their relatives. First, this research shows that parents who detect signs earlier also report more concerns about language and communication problems and often have an older son with TD. However, when they share their concerns with health professionals, they often receive the advice of "wait and see". This result suggests that health professionals should give more attention to these parents' concerns, especially under certain circumstances to promote an early diagnosis and access to early intervention. This result also suggests that informing pediatricians about ASD signals and how to respond to parents' questions or concerns properly might enhance their response to parents' concerns and, consequently, decrease the time until a formal diagnosis of ASD.

Furthermore, parents' knowledge about early signs of ASD at a linguistic and communicative level can help them improve early detection of ASD. These signals might be useful at a clinical level and guide what questions should be asked by health care professionals to address parents' concerns related to a neurodevelopmental disorder. Although none of the vocalization variables we examine are likely to be a very sensitive marker on its own, examining the quality of vocalizations (i.e., intentional communication) but not the complexity of them, may be clinically useful in predicting later ASD diagnosis. Thus, the research reported in this thesis offers early signals easily detectable by people who are not specialists in the fields of psychology or speech.

Secondly, we test the significance of several linguistic and psychological variables in children with ASD that influence FQoL. In practice, children with ASD level of support 1 show pragmatic difficulties. It is essential that professionals who work with these children focus on and help families improve these abilities. Our results highlight the importance of the development of pragmatics in different settings, not only within the family. For example, given that children with ASD level of support 1 tend to be integrated in ordinary schools, detecting language difficulties could have an impact on the quality of the relationships with their peers.

Some implications for the treatment derive from the study of adaptive behavior in children with ASD level of support 1. Particularly at this level, ASD is associated with a high risk of difficulties in several variables that are directly associated with the adaptive behavior (such as behavioral problems, hyperactivity, problems with their peers, and emotional problems). Moreover, prosocial behaviors predict the perceived satisfaction on FQoL. Thus, pay particular attention to those variables related to prosocial skills may have a direct effect on both children's adaptive function and FQoL.

Other clinical implications, related to children with ASD level of support 3, affect language comprehension (at a grammatical, nonverbal language, and social communication level). Although comprehension impairments are not diagnostic criteria, they should be taken into account in interventions. Moreover, improving these skills would also imply a higher level of satisfaction in FQoL, since differences in comprehensive language level and communication from children with ASD level of support 3 and TD, could explain differences in satisfaction level of FQoL in these families.

Third, the research reported in this thesis can have important implications about the variables related to first degree relatives from children with ASD that could explain FQoL. After completion of a brief parent mediated intervention, we found improvements in the satisfaction perceived in FQoL (i.e., family interaction, parental role, and emotional well-being) and in the management of communication and the interaction between parents and children. Parents of children with ASD are capable of acquiring, in a brief period of time, some strategies to enhance their children's abilities. In addition, this intervention may have a positive transactional effect on the motivation of the parents in order to get involved in their children's interventions. Overall, the main clinical implication of this study consists in providing some keys to identify brief interventions that may be especially promising for the research on quality of life for these families.

Besides these benefits after completion of a brief mediated intervention, we show additional individual variables that could have an impact on FQoL. Specifically, the research reported in this thesis shows that adequate levels of objective numeracy can help families of children with ASD improve their FQoL. A possible implication from this study is that objective numeracy skills can reduce the burden of overloaded information and decision vulnerability,

helping parents manage their children's condition. Thus, these results contribute to the growing literature on parental characteristics that make parents resilient and help families with children with ASD thrive as much as families with children with TD do.

Although the results of our meta–analysis reflects that the patterns of altered linguistic and motor skills in HR are not stable, there are certainly atypical aspects of linguistic and motor skills which yield differences in group comparisons between them and LR. Accordingly, monitoring the development of HR is crucial to ensure the progress of linguistic and motor skills, and work with those difficulties that are likely to appear among this population.

We highlight the importance of planning interventions focused on improving Sibs–ASD social support. Although nearly 20% of Sibs-ASD has a diagnosis, the family environment where they develop might have an impact on their abilities and their FQoL. Moreover, individual differences detected among Sibs-ASD (such as autism traits) should be taken into account when making decisions about how to improve the social support.

In conclusion, these implications could enhance, among those professionals who work with children with ASD and their families, the design of goals on early detection of ASD and interventions to further improve their FQoL; specially, those intended to work with children with ASD, their parents, and their siblings.

CURRICULUM VITAE

Nací en Granada, España. En 2012 me licencié en Psicología por la Universidad de Granada. Realicé el máster de investigación de Psicología de la Salud de la Universidad de Granada. Desde 2012 a 2015 trabajé como Psicóloga a cargo del Departamento de Psicología Infantil del centro de Psicología Terapia y Salud en las Gabias (Granada). Estoy homologada como Psicóloga General Sanitaria por la Junta de Andalucía. En 2015 comencé los estudios de doctorado con una beca predoctoral (programa nacional FPU) del Ministerio Español deEducación, Cultura y Deporte en la Universidad de Granada. Para esta tesis, he pasado seis meses en la Universidad de Carolina del Norte en Chapel Hill (UNC) en EE.UU. y tres meses en la Universidad de Göttingen (UMG) en Alemania. Durante los estudios de doctorado, además de los artículos de la tesis, he sido autora de otros tres artículos publicados en revistas revisadas por pares y soy revisora de cuatro revistas científicas de impacto. He participado en más de 30 comunicaciones en congresos científicos nacionales e internacionales y he colaborado en la organización de diversas jornadas. He impartido docencia tanto en el grado de logopedia como en el de psicología y he sido invitada como docente para impartir diversos cursos y jornadas organizados tanto por organismos públicos como asociaciones de autismo. También soy miembro del Colegio Oficial de la Psicología (AO-07513), de la Asociación Española de Profesionales del Autismo (AETAPI), de la International Society for Autism Research (INSAR) y del grupo de investigación de Aprendizaje, Emoción, y Decisiones de la Universidad de Granada.

Dunia Garrido was born in Granada, Spain. In 2012 she graduated from the BA program in Psychology at University of Granada. She then completed a research master in Health Psychology at the University of Granada. From 2012 to 2015 Dunia work as a Psychologist, in charge of the Child Psychology Department in Terapia y Salud in Las Gabias (Granada). In 2015 Dunia started her predoctoral fellowship (FPU program) to complete doctoral studies from the Spanish Ministry of Education, Culture and Sport at the University of Granada. For her thesis Dunia spent six months at University of North Carolina at Chapel Hill (UNC) in the USA and three months at University of Göttingen (UMG) in Germany. During her doctorate studies, beyond the articles of her thesis, she was author of another three articles published in peer–reviewed journal, and she is reviewer of four scientific journals. Dunia has participate in more than 30 communications in national and international scientific meetings, and she has collaborate in the organization of several scientific conferences. Dunia has taught at the university of Granada both Psychology and Speech therapy, and she has been invited as speaker in several workshops and conferences organized by public agencies and associations of autism. Dunia is also a member of the Official Association of Psychologists of Spain (AO-07513), of the Spanish Association of Professionals related to Autism (AETAPI), the International Society for Autism Research (INSAR) and of the Learning, Emotion, and Decisions Research Groups at the University of Granada.

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