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CASE REPORT

Emergent genital infection by *Leptotrichia trevisanii*

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Abstract
We report the first case of an association between *Leptotrichia trevisanii* and an episode of pelvic inflammatory disease (PID) and the second case of the isolation of this infection in the cervical canal. A 45-yr-old woman was admitted to our emergency department with clinical and radiological signs and symptoms compatible with an episode of PID. She was hospitalized for intravenous antibiotic control and treatment and the subsequent surgical drainage of abscesses. Cultures were taken throughout the process, but only cultures from cervical canal exudate were positive, with the growth of *L. trevisanii* species. It appears important to carry out a complete microbiological screening, not limited to conventional agents, on adequate clinical samples to detect possible infectious agents that may be missed in these cases.

Keywords Genital infection · *Leptotrichia trevisani* · Emerging infection

Introduction

Bacteria of the genus *Leptotrichia* are anaerobic Gram-negative but also aerotolerant and adapt to growth under CO_2-supplemented aerobiotic conditions. The genus includes seven species: *L. buccalis, L. goodfellowii, L. hofstadii, L. hongkongensis, L. shahii, L. trevisanii*, and *L. wadei*. They colonize mucosae and can be found in the oral cavity, gastrointestinal tract, urogenital system, and female genital tract. Their presence has been associated with periodontal disease, oral cavity abscesses, infectious endocarditis, acute appendicitis, bacterial vaginosis, salpingitis, and bacteremia in neutropenic patients with oral mucosal damage [1–7]. Severe systemic infections, most frequently with *L. buccalis*, have been reported in immunosuppressed patients [2, 8]. However, *L. trevisanii* species has only been detected in blood, dental plaque, stool, and amniotic fluid cultures (Table 1). We present the first report of an association between this species and an episode of pelvic inflammatory disease (PID) and the second report of its isolation in the lower genital tract, specifically in the cervical canal. We analyze the clinical, diagnostic, and therapeutic data obtained for this patient and other factors that may elucidate the pathogenesis.

Case report

We report the case of a 45-year-old woman with no medical or surgical history of interest who had carried a copper intrauterine device (IUD) for 5 years. The patient had no toxic habits and denied any high-risk sexual relationships. She visited the Obstetrics and Gynecology Emergency Department with a 1-month history of intermittent pain in left iliac fossa that had intensified during the previous week and become localized in the mesogastrium and epigastrium. She described general malaise and intermittent episodes of diarrhea but no other symptoms or signs of interest. Her vital constants were normal, with no fever. Diffuse pain was produced by abdominal palpation. Vaginal examination showed non-specific leucorrhea, and she reported pain with mobilization of the cervix during vaginal palpation. Analytical

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Table 1 Main clinical manifestations in patients with infection by *Leptotrichia trevisanii*. Modified from Eribe et al. [18]

Case	Microorganism isolated	Sex, age	Clinical symptoms	Sample	References
1	*Leptotrichia trevisanii*	F, 74	Odynophagia, oral lesion, fever, and pneumonia	Blood	[4]
2	*Leptotrichia trevisanii*	M, 53	Multiple myeloma, chemotherapy, mucositis	Blood	[9]
3	*Leptotrichia trevisanii*	M, 56	Multiple myeloma, chemotherapy, mucositis	Blood	[9]
4	*Leptotrichia trevisanii*	F, 63	Acute myeloid leukemia, febrile neutropenia, chemotherapy, mucositis	Blood	[9]
5	*Leptotrichia trevisanii*	F, 12	Acute myeloid leukemia, febrile neutropenia, stomatitis, chemotherapy	Blood	[10]
6	*Leptotrichia trevisanii*	M, 66	Esophageal carcinoma, chemotherapy, febrile neutropenia, dysphagia, esophageal lesion	Blood	[10]
7	*Leptotrichia trevisanii*	M, 69	Diffuse large B-cell lymphoma, mucositis, fever, diarrhea, hematopoietic cell transplant	Blood, Stools	[11]
8	*Leptotrichia trevisanii*	M, 62	Multiple myeloma, odynophagia, mucositis, febrile neutropenia, persistent catarrhal symptoms	Sangre	[11]
9	*Leptotrichia trevisanii*	M, 7	Burkitt's lymphoma and fever	Blood	[12]
10	*Leptotrichia trevisanii*	M, 37	Diffuse large B-cell lymphoma and fever	Blood	[13]
11	*Leptotrichia trevisanii*	M, 65	Plasmablastic lymphoma and fever	Blood	[13]
12	*Leptotrichia trevisanii*	F, 34	Stomach and colon cancer and diarrhea	Blood	[13]
13	*Leptotrichia trevisanii*	M, 19	Ewing's sarcoma and fever	Blood	[13]
14	*Leptotrichia trevisanii*	F, 36	Chorioamnionitis at 19 weeks of gestation, fever, amniotic sac prolapse	Amniotic fluid	[14]

results revealed leukocytosis with left shift and elevated CRP count. Transvaginal ultrasound revealed a loculated image occupying the pouch of Douglas that contained a hyper-refractive 33-mm image. Findings of abdominal and pelvic CT with contrast suggested PID, with left oophoritis, bilateral salpingitis, and liquid in pelvis, apparently loculated. Cultures were taken of the cervical canal exudate [15], and the patient was hospitalized with intravenous analgesia and antibiotherapy of 600 mg clindamycin/6 h and 240 mg gentamicin/24 h. Analytical results improved over the next 3 days but the clinical symptoms did not; therefore, laparoscopic surgery was performed to drain the abscesses and pelvic cavity and remove the IUD, taking cultures of purulent liquid from abscesses and cavity. Analytical results continued to normalize and clinical symptoms also improved, and the antibiotherapy was changed to 600 mg oral clindamycin/6 h and 1 g intramuscular ceftriaxone/12 h. The patient was discharged at 5 days post-surgery with a prescription for oral 500 mg ciprofloxacin/12 h for 7 days to complete the antibiotherapy. PCR study (BD Max, Becton–Dickinson Diagnostics, Sparks, MD) of cervical canal exudate was negative for *Chlamydia trachomatis, Neisseria gonorrhoeae, Mycoplasma genitalium, Mycoplasma hominis*, and *Ureaplasma urealyticum*. After 48-h incubation in CO_2 in blood agar medium (Becton Dickinson, Spain) and chocolate agar medium (Becton Dickinson), colonies identified as *L. trevisanii* by the Maldi-Tof technique (Bruker Biotyper, Billerica, MA, USA) were isolated in the cervical canal culture (Fig. 1) (score 2.103), which was confirmed by 16S rRNA gene sequencing (performed in Centro Nacional de Microbiología, Majadahonda, Madrid, Spain). *E* test was performed according to 2017 CLSI criteria for anaerobic

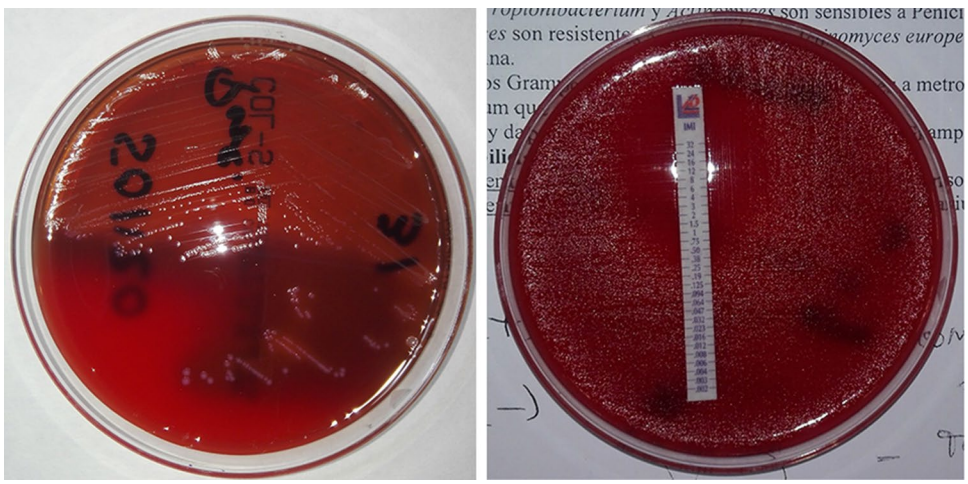

Fig. 1 Image of colonies of *Leptotrichia trevisanii* in blood agar culture medium after 48-h incubation under anaerobic conditions (left) and in the presence of imipenem antibiotic by the *E* test (right)

Table 2 Antibiotic susceptibility of *Leptotrichia trevisanii* according to the *E* test

Antibiotic	MIC (in mg/L)	Clinical category
Clindamycin	0.032	S
Amoxicillin/clavulanic acid	0.25	S
Penicillin	0.25	S
Imipenem	0.25	S
Metronidazole	0.38	S
Moxifloxacin	12.0	R

MIC minimum inhibitory concentration

Gram-negative bacilli (CMI value in mg/L) on Brucella agar supplemented with hemin (5 µg/mL), vitamin K1 (1 µg/mL), and laked sheep blood (5% v/v) (Becton Dickinson, BD, Franklin Lakes, NJ, USA), 36 °C ± 1 °C, anaerobically, 48 h. The results are shown in Table 2. Cultures of intraoperative samples were all negative.

Discussion

Pelvic inflammatory disease is an acute or subacute infection of the female upper genital tract that typically ascends from the cervical canal and involves the uterus, fallopian tubes, ovaries, and even adjacent organs. It can cause endometritis, salpingitis, oophoritis, peritonitis, perihepatitis, or tubo-ovarian abscesses. PID risk factors include age < 25 years, multiple sexual partners, non-use of barrier methods, previous PID, the co-presence of other sexually transmitted diseases or bacterial vaginosis, IUD placement (for 3 months post-insertion) or any other diagnostic or therapeutic intervention in the uterus [16].

PID develops in two stages, the first with inflammation of pelvic soft tissues and the second with the formation of intra-abdominal abscesses. Up to 85% of PID cases are caused by sexually transmitted pathogens, mainly *Neisseria gonorrhoeae* and *Chlamydia trachomatis*, while a small proportion is caused by bacterial vaginosis agents (e.g., *Peptostreptococcus* spp., *Leptotrichia* spp., *Gardnerella, Bacteroides* spp., or *Atopobium* spp.) or by enteric (e.g., *Escherichia coli, Bacteroides fragilis*, group B streptococci, and *Campylobacter* spp.) or respiratory (e.g., *Haemophilus* spp., *Streptococcus pneumoniae*, group A streptococci, and *Staphylococcus aureus*) microorganisms that colonize the lower genital tract [18]. Cytomegalovirus, *M. hominis, U.urealyticum*, and *M. genitalium* can also be associated with some cases of PID. Regardless of the etiology, the therapeutic approach should be based on PID as a mixed (polymicrobial) infection by facultative and strict anaerobic bacteria.

Microbiological diagnosis is conducted by culture and by molecular study (e.g., PCR) of samples of cervical canal exudate and any infection focus detected (e.g., pyometra, abscesses, etc.). *L. trevisanii*, associated with PID in the present case, has only previously been detected in blood, dental plaque, stool, and amniotic fluid cultures [4, 9–14, 17], with one possible case of its identification in the genital tract of a pregnant woman with chorioamnionitis [14]. Hence, inadequate data are available to elucidate the relationship between other species of the genus *Leptotrichia* and bacterial vaginosis or to determine the association of this infection with PID development.

Leptotrichia travesanii has most frequently been detected in blood, which may have been the vehicle for its spread to the patient's lower genital tract. This microorganism has also been reported in dental plaque and has been associated with the development of periodontal abscesses; therefore, the oral cavity is another plausible source of this infection (e.g., during oral sex). *L. travesanii* has also been detected in stools, so that the gut flora may also be the origin of this type of infection [11], with infection of the genital tract caused by transfer from the anus during post-defecation wiping or certain sexual practices or by the presence of recto-vaginal fistulas.

The lack of previous reports of this infection in the female lower genital tract may be attributable to the usually incomplete nature of microbiological screening, frequently limited to the search by PCR of the most prevalent agents in these cases, such as *N. gonorrhoeae* and *C. trachomatis*.

Activity against *L. travesanii* has been reported for penicillin, ampicillin, oxacillin, ampicillin/sulbactam, amoxicillin/clavulanic, clindamycin, metronidazole, rifampicin, tetracycline, imipenem, and chloramphenicol [2, 11, 18]; however, inadequate data are available to indicate the antibiotherapy of choice.

In the present patient, intravenous clindamycin and gentamicin improved analytical but not clinical parameters. After the surgery, the patient was changed to intravenous clindamycin and ceftriaxone with a favorable clinical outcome. The patient was prescribed with oral ciprofloxacin at hospital discharge, but this antibiotic is not active against anaerobic bacteria and the antibiogram showed moxifloxacin resistance. It can, therefore, be assumed that the infection had been successfully remitted by the previous treatments.

In conclusion, this is the first report of genital infection by *L. trevisanii* in the cervical canal. This species can be identified in a simple manner by MALDI-TOF, and its detection at this localization may become more frequent if the appropriate diagnostic methods are systematically implemented.

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Compliance with ethical standards

Ethical statement The study protocol was carried out in accordance with the Declaration of Helsinki. This was a non-interventional study with no additional investigation to routine procedures. Biological material was only used for infection standard diagnostics following physicians' prescriptions. No additional sampling or modification of the routine sampling protocol was performed. Data analyses were carried out using an anonymous database. For these reasons, ethics committee approval was considered unnecessary according to national guidelines. The Infectious Diseases and Clinical Microbiology Clinical Management Unit of the University Hospital Virgen de las Nieves of Granada (Spain) granted permission to access and use the data.

Conflict of interest None.

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