Cerebrospinal fluid shunt-associated meningitis caused by *Gordonia sputi*: case report and review of the literature

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SUMMARY

We report the first known case of cerebrospinal fluid (CSF) shunt-associated meningitis caused by *Gordonia sputi* and review published cases of *Gordonia* CNS infections.

Keywords: *Gordonia sputi*, cerebrospinal fluid, shunt, meningitis, central nervous system infection.

INTRODUCTION

Gordonia spp are uncommon but emerging pathogens, mainly in the setting of medical device-related infections. Central nervous system (CNS) infections by *Gordonia* spp have only been reported anecdotally.

CASE REPORT

An 82-year-old man was admitted to hospital due to a 2-week course of limb weakness, urinary incontinence and malaise. The patient's medical history included diabetes mellitus with retinopathy and nephropathy. Four years prior to presentation, he had been diagnosed normal pressure hydrocephalus and a CSF ventriculo-peritoneal shunt was placed.

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At the time of admission, the patient was misdiagnosed with urinary tract infection and empirical treatment with ceftriaxone was initiated. On the first day in hospital, he developed vespertine fever, and neurological and cognitive impairment. Urine culture was negative. According to these findings, CSF-shunt dysfunction caused by an infection was suspected. Consequently, a magnetic resonance image study was performed which confirmed a worsened CSF dynamic, with a duplicated aqueductal stroke volume compared to previous studies. In addition to that, a CSF valve sample was analyzed, showing 12 leukocytes per mm^3 with 90% granulocytes, normal glucose and protein values. A Gram staining showed Gram-positive rods. Therefore, CSF-shunt associated meningitis diagnosis was established. In this clinical context, age, poor specific clinical presentation, CSF findings (both biochemical and microbiological), and no extra-meningeal foci of infection raised the suspicion of *Listeria monocytogenes* etiology [1]. As a result, antibiotic therapy was changed to ampicillin, associated with vancomycin and ceftriaxone until definitive culture results.

CSF valve culture yielded few rough, brownish colonies at the third day of incubation in chocolate agar plates (Figure 1). On the sixth day in hospital, fever persisted and empirical therapy was changed to linezolid and meropenem to cover the possibility of nosocomial secondary infection. Surgical intervention for shunt withdrawal was scheduled on the eleventh day. A ventricular CSF specimen obtained in surgery contained 262 white cells per mm^3 with 60% granulocytes, high protein (130.5 mg/dL) and low glucose levels (73 mg/dL - blood glucose level 224 mg/dL). Both ventricular CSF and peritoneal shunt tip cultures were positive for the same Gram-positive rods. The patient persisted febrile two days after surgery, but isolations were not detected on blood cultures. An abdominal TC and a transthoracic echocardiogram allowed to discard an abdominal abscess and infective endocarditis, respectively. A week after CSF shunt removal and after 18 days of systemic antibiotic therapy, the patient started to improve, subsiding fever, and slowly recovering neurological status.

The Gram-positive rod isolated in all cultures was identified using matrix-assisted laser desorption ionization-time of flight mass spectrometry (MALDI-TOF system) as *Gordonia sputi* with a low score (1.65 and 1.57, respectively). The strain was sent to the Reference Laboratory for Taxonomy (National Centre for Microbiology, National Institute of Health Carlos III) which confirmed the previous identification by 16S rRNA gene sequencing. Antimicrobial drug susceptibility was performed using E-test method in chocolate agar and susceptibility data were interpreted according to the Clinical and Laboratory Standards Institutes [2]. *Gordonia sputi* was susceptible to all antibiotics tested (Table 1).

Two weeks after CSF shunt withdrawal, a negative CSF culture confirmed the bacterial eradication. The patient completed 4 weeks of intravenous antibiotic and was discharged continuing oral linezolid. On the follow up, the patient completed 3 months of linezolid. A new assessment confirmed negative CSF culture after 5 months of antibiotic withdrawal. After 18 months the patient maintains good neurological status and has not been scheduled for another CSF shunt placement.

Table 1 - *Gordonia sputi* susceptibility test (minimum inhibitory concentrations, MICs).

Antibiotics	*MIC (μg/mL)*
Amoxicillin	≤0.125
Ceftriaxone	≤0.5
Imipenem	≤0.042
Gentamicin	≤0.38
Vancomycin	≤1.5
Ciprofloxacin	≤0.032
Linezolid	≤0.5

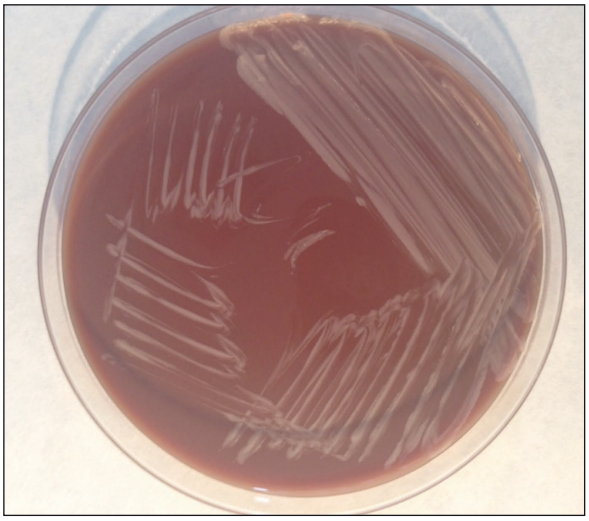

Figure 1 - Agar chocolate plaque with gram-positive rods colonies. Eventually identified as *Gordonia sputi*.

DISCUSSION

Few case reports of *Gordonia* genus infections have been reported in medical literature, but it seems they are increasing in recent years. *Gordonia* spp are coryneform bacteria, slow growing Gram-positive, slightly acid-fast actinomycetes. These rods show intermediate characteristics between *Nocardia*, *Rhodococcus* and even *Mycobacterium*. Previously classified as *Rhodococcus* species, comparative analysis of ribosomal 16S RNA by Tsukamura first distinguished the genus *Gordonia* [3]. *G. sputi*, *G. bronchialis* and *G. terrae* have been reported as the major pathogenic strains within this genus, which includes more than 30 species [4-6].

Microbiologists can find laborious the identification of *Gordonia* species, since their phenotypical characteristics are shared with similar microorganisms. Currently, the principal technique to identify *Gordonia* is 16S rRNA gene sequencing, which is only done at referral centers. However, mass spectrometry systems as MALDI-TOF MS, represent a promising method for identification [7].

Most case reports of *Gordonia* infections have involved hosts immunodeficient or wearing medical devices, mainly indwelling catheters. The first pathogenic role of *Gordonia* species was described in sternal wound infections and mediastinitis after coronary artery by-bass [8]. Most frequently reported cases of *Gordonia* infection involve patients with catheter-related bacteremia, occasionally complicated by infective endocarditis [9, 10]. The newest reports are case series of *Gordonia* peritoneal-dialysis related peritonitis [7, 11]. Immunosuppressed patients are generally at higher risk for developing *Gordonia* infections, mainly patients that carry medical devices. Nevertheless, *Gordonia* human infections have also been described in immunocompetent hosts [12].

There is no standardized treatment for *Gordonia* infections. In contrast to some other actinomycetes, *Gordonia* are generally susceptible to many antimicrobial drugs. Some authors suggest initial therapy with carbapenems or fluoroquinolones in combination with an aminoglycoside [5]. Most isolates are susceptible to vancomycin. However, trimethoprim-sulfamethoxazole, which is often used to treat *Nocardia* infection, has poor activity against *Gordonia* species. *Gordonia* isolates are also susceptible to linezolid [4]. Antimicrobial regimen should be guided by *in vitro* susceptibility test results.

The duration of therapy is unclear and should be based on the host's underlying immune function and clinical response. When dealing with medical devices *Gordonia* infection, persistence or recurrence of positive culture is common if the device is not removed. Therefore, withdrawal of the implanted device is recommended. Central nervous system infections caused by *Gordonia* species are rare. To our knowledge, there are only 4 medical reports in the literature (Table 2) [5, 13-15]. There are several characteristics that highlight the singularity of our case. To our knowledge, this is the first reported case of central nervous system infection caused by *Gordonia sputi*. Therefore, the clinical management was challenging.

CSF-shunt associated meningitis was suspected on the basis of the clinical presentation and CSF valve sample findings, and confirmed with ventricular CSF sample and several positive culture results. Differences of biochemical findings and rate of positive culture depending on the place from where CSF is obtained (shunt, ventricular, lumbar) are described, but the preferable one is not well established [16].

We chose to initiate the diagnostic process with a CSF shunt sample which seemed to be the easiest and less aggressive procedure given the clinical status and comorbidities of our patient. In addi-

Table 2 - Reported central nervous system infections with *Gordonia spp.*

Year of publication [reference]	*No. of cases*	*Gordonia species*	*Source*	*Microbiological definitive identification*	*Presentation (Comorbidity)*
1985 [12]	1	*G. aurantiaca*	CSF*	Biochemical test	Meningitis (Hairy cell leukemia)
1994 [13]	1	*G. terrae*		16S rRNA gene sequencing	Brain abscess (Surgical drain, malignant tumor)
1997 [14]	1	*G. terrae*	CSF, brain biopsy (Gram)	16S rRNA gene sequencing	Meningitis and brain abscesses (Immunocompetent)
2007 [4]	1	*G. bronchialis*	CSF	16S rRNA gene sequencing	Meningitis (Intracranial shunt, premature neonate)
Own case	1	*G. sputi*	CSF, shunt tip	16S rRNA gene sequencing	Meningitis (CSF Shunt and diabetes)

*CSF: cerebrospinal fluid

tion, its rate of positive culture is the greatest described [16].
Following other medical devices infections by *Gordonia* and the current recommendations for managing CSF shunt-related meningitis, removal of shunt catheter was imperative [16]. Our patient maintained fever even after surgery, and systemic broad spectrum antibiotics were needed to finally control the infection.
Linezolid was chosen due to the possibility of oral administration, its diffusion to central nervous system and bacterial susceptibility. The choice of a long suppressive antibiotic regimen was based on *Gordonia* genus similarity to *Nocardia* and *Actinomyces* infection. Another remarkable aspect of our case was the timing of the infection, since the shunt had been placed 4 years before the infection appeared, while shunt meningitis is typically contracted intra-operatively and usually develops a month after the surgery [16].
Finally our patient was affected by diabetes mellitus as the only underlying condition, suggesting that, for the physiopathology of the infection, the presence of a medical device is a risk factor more important than the immune status. This case prompted us to look for more cases of *Gordonia* infection at our institution. We found other two cases of *Gordonia* infection (*G. sputi* and *G. rubropertintca*) in the last 20 years, both catheter-related bloodstream infection in patients involved in hemodialysis [17].
In both patients catheter withdrawal was warranted to control the infection.

CONCLUSIONS

The present report describes the first case of *Gordonia sputi* CNS infection and the first ventriculo-peritoneal shunt infection caused by this microorganism. *Gordonia* spp is an emerging pathogen, mainly related to medical devices, which should be added to the list of pathogens that may cause CSF shunt infection. Systemic broad spectrum antibiotics and removal of the device usually warrant control of the infection, but the appropriate length of antibiotic course is unknown.

Conflict of interest. On behalf of all authors, the corresponding author states that there is no conflict of interest.

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