CASE REPORT

Uncommon cervical pain due by *Bacillus pumilus* spondylodiscitis in an immunocompetent patient: a case report

Federico Fusini¹, Gabriele Colò², Alessandro Massè³, Massimo Girardo⁴

¹Department of Orthopaedic and Traumatology, Regina Montis Regalis Hospital, ASL CN1, Mondovì (CN), Italy; ²Department of Orthopaedics and Traumatology, Regional Center for Joint Arthroplasty, ASO Alessandria, Alessandria, Italy; ³Department of Orthopaedic and Traumatology, Orthopaedic and Trauma Centre, Città della Salute e della Scienza di Torino, University of Turin, Turin, Italy; ⁴Spine Surgery Unit, Orthopaedic and Trauma Centre, Città della Salute e della Scienza di Torino, University of Turin, Turin, Italy

Abstract. *Case*: A fifty-three year-old military patient, presented with a 3 months neck pain and paraesthesia of upper right extremity after gastroscopy. Radiological examination demonstrated a uncertain degenerative/infective involvement of C5-C6 intervertebral disc. *Conclusion*: The patient underwent anterior debridement with C5-C6 disc excisional biopsy, with microbiological findings of Bacillus pumilus with high bacterial concentration. After biopsy, segment arthrodesis was achieved only with vertebral plate cruentation and 3 months of cervical collar. He completed a six-weeks course of intra-venous antibiotics for the treatment of atypical spondylodiscitis. At one year of follow-up, he had no residual neck pain or neurological signs or symptoms. (www.actabiomedica.it)

Keywords: *Bacillus pumilus*, spondylodiscitis, infection, spine

Introduction

Cervical spondylodiscitis represents a very rare location, and it is often related to pathologies which leads to immunosuppression(1). Clinical presentation could be insidious with a high risk of neurological deterioration, rapid evolution, morbidity and poor response to surgical treatment. A 21% mortality rates has been reported in literature(2).

Bacillus pumilus is a common inhabitant of soil environments, but some strains are known to be plant pathogenic or probiotic for animals(3). Usually, Bacillus spp have little or no pathogenic potential for human subjects, with exception of Bacillus Anthracis and Cereus(4). Only few extremely rare cases of human B. pumilus infections have been reported in literature(5,6).

Detection of positive cultures to *Bacillus sp.* may lead to diagnostic issues since the high prevalence of these bacteria in nature(7). However, they should be considered as a pathogen in case of repeated positive cultures and high concentrations.

Here we describe, to our knowledge, the first report of cervical spondylodiscitis in a healthy male patient due to *Bacillus pumilus*.

The patient was informed about the unusual presentation of the case and he consented for publication.

Case report

A fifty-three male military patient from Lebanon accessed to our Spine Surgery Department for 3 months

persistent cervical pain unresponsive to Paracetamol 1000mg 3 times/daily and Ibuprofen 600mg 2 times/daily for 1 month. Patient reported an insidious onset of cervical pain and occasional upper right extremity paraesthesia, with predominance during night and waking up and no history of previous trauma. Paraesthesia was also present during the day, worsening with neck flection and extension. His medical history was silent, except for gastroesophageal reflux disease (GERD), and a month before cervical pain onset the patient underwent a routine gastroscopy. No other invasive procedures were reported. In addition, no fever, night sweats or weight loss in the last months was reported.

On physical examination, the patient had a normal gait. Cervical range was little decreased in all directions due to discomfort. Romberg and Spurling tests were negative. Motor strength was 5/5 in all muscle groups bilaterally. Touch sensation was intact bilaterally, with hyperreactive tricipital reflex on the right side. Hoffman test was negative on both sides. There was no clonus and Babinsky test was negative.

Conventional and dynamic radiography showed decrease intervertebral disc height with sclerosis and little erosions of inferior vertebral plate of C6 and superior plate of C7 without instability. Computed tomography (CT) scans confirmed sclerosis and highlight erosion of vertebral endplates (Figure. 1) while Magnetic Resonance (MR) with gadolinium showed

signal alteration of C6-C7 intervertebral disc consistent with hypothesis of spondylodiscitis (Figure. 2).

Major findings of laboratory test were reported in table 1.

Due to high suspicious of spondylodiscitis, an anterior open biopsy of intervertebral disc was planned in order to obtain an adequate laboratory material for microbiological examination. A little incision was performed on anterior longitudinal ligament in order to preserve ligament integrity and some residual disc, without any macroscopic features of infection, was left on site.

Intervertebral disc biopsy appeared dampish and degenerated with a grey colour shape; five fragments were collected. Little erosions of vertebral endplates were found and removal of necrotic tissues was performed. After surgical procedure, the patient wore a rigid cervical collar (Aspen Vista® cervical collar) for approximately 3 months.

On five microbiological specimens, four were positive for *Bacillus pumilus* in high concentration (+++, approximately 200-300 CFU), sensible to several antibiotics (Tab I). Because of these results, patient started 6 weeks intravenous antibiotics (Amoxicillin plus Clavulanic Acid 2,2g 3 times/daily plus Ciprofloxacin 400mg 2 times/daily), then switched to oral therapy for another 6 weeks (Amoxicillin 875mg + Clavulanic Acid 125mg 3 times/daily plus Ciprofloxacin 500mg 2 times/daily).

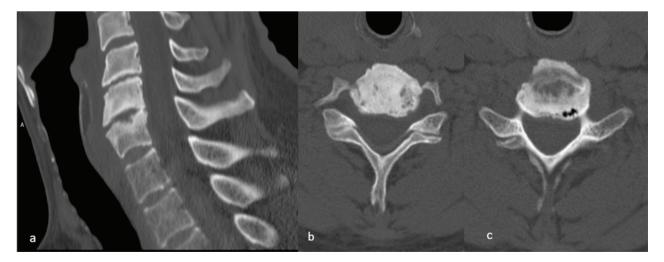


Figure 1. Preoperative CT scan of C6-C7 segment showing reduction of C6-C7 disc height, anterior osteophytotic degeneration of vertebral bodies, initial posterior ankylosis of C6 and C7 vertebral plates (a), sclerosis and little erosion of inferior C6 (b) and superior C7 vertebral plates (c).



Figure 2. Preoperative STIR MR with positive oedema and inflammation of C6 and C7 trabecular bone and C6-C7 intervertebral disc (a), and Gadolinium enhanced MR (b) with medium diffusion into C6-C7 segment, compatible with spondylodiscitis.

Table 1. Major findings of laboratory blood exam, microbiological coltures and antibiotic susceptibility of Bacillus pumilus isolated from disc fragments: we report only interesting values of laboratory blood exams, while normal values were not.

Laboratory test	Value	Reference
WBC	9,90*109/L	4.00 - 10.00 * 10 ⁹ /L
CRP	5,9mg/dL	0-3mg/L
ESR	18mm/h	1-15mm/h
	Microbiological samples	
Samples	Pathogen	Microbial load
Sample 1	Bacillus pumilus	+++
Sample 2	Bacillus pumilus	+++
Sample 3	Bacillus pumilus	+++
Sample 4	negative	negative
Sample 5	Bacillus pumilus	+++
	Antibiogram	
Antibiotic	MIC (μg/ml)	Susceptibility
Ampicillin	0,016	S
Amoxicillin	0,016	S
Ciprofloxacin	0,016	S
Clindamycin	0,032	S
Erythromicin	0,38	S

Antibiogram		
Antibiotic	MIC (μg/ml)	Susceptibility
Imipenem	1	S
Levofloxacin	0,032	S
Teicoplanin	0,032	S
Trimethoprim/sulfamethoxazole	0,016	S
Vancomycin	0,5	S

Laboratory tests normalised after 6 weeks with except for ESR, which returns to normal level after 8 weeks. After 12 weeks, cervical collar was gradually removed, starting with 2 hours/daily to complete wear off in about 2 weeks.

Symptoms progressively decreased and after cervical collar removal, cervical range of motion was comparable with preoperative examination. Radiographic examination at follow up showed resolution of infection and no sign of instability. At one year of follow-up, he had no residual neck pain or neurological signs or symptoms, and radiological examinations were negative for infection.

Discussion

Opportunistic infections are extremely rare in healthy population since immunodepression is one of the most important risk factors. Bacillus pumilus infection are rarely reported in literature and infection of human subject is exceptional.(5) Most of all cases reported in literature are related to pediatric patients or immunodepression-associated pathologies(8,9), while the others showed positive history of major surgery(5). Most of cases reported in literature are related with food poisoning, due to consumption of meat dishes, eggs, baked products or tomato sauce(10) and presenting with fever, bacteremia and gastrointestinal symptoms. In pediatric and newborn patients, sepsis due to Bacillus Pumilus was related to the positioning of intravenous catheter(8), while in oncohematological patients undergoing chemotherapy, this infection causes bacteraemia, with possible exitus due to multiorgans failure(9). In rare cases, localized infection can show up with anthrax-like lesion, with localized skin ulceration associated with necrosis, erythema and edema(11). Also diagnostic/therapeutic invasive procedures could be considered a potential risk factor; in fact, our patient underwent a gastroscopy some times before symptoms onset, while some Authors reported central venous catheter as risk factor(6,7,12).

In case of conventional pathogens, cervical pain and neurological symptoms commonly occurred in association with increase of CRP, ESR and WBC count.(2) In our report we found only a little increase of CRP and ESR, not pathognomonic for infection, with normal WBC count. This difference should be taken in account since *Bacillus pumilus* is only an occasional human pathogen and it is considered a low immunogenic and virulent pathogen. In fact, it is not able to produce the same haemolytic and cytotoxic toxin of other human bacillus pathogen *Bacillus Cereus or Anthracis*, and it is rarely able to synthetize enterotoxins(13). Moreover, as all probiotic *Bacillus spp*, it is able to modulate and inhibit inflammatory response(14).

MR with gadolinium was the most important diagnostic tool in our hands; in fact, only MR showed a compatible picture with spondylodiscitis, while laboratory tests and clinical examination were aspecific. MR has high sensitivity and specificity in diagnosis and differentiation of the type of spondylodiscitis and may reveal signs of spondylodiscitis even in very early stages.(12,15)

Since we only performed an open biopsy and not a complete cervical discectomy, with preservation of the anterior longitudinal ligament and as much disc as possible, no major instability to cervical spine was induced. As showed by dynamic X-rays, the segment was stable during flexion and extension, with good range of motion(16) and without need of hardware placement(17–19). A 3 months of precautionary

cervical collar wear and resolution of infection after antibiotic therapy promoted segment healing.

After surgery, at least 6 weeks of intravenous antibiotics is recommended and they should be prolonged according to type of infection, patient health status and permanence of implanted hardware. In our case, after 6 weeks of intravenous and 6 weeks of oral antibiotics the patient didn't need any further long term suppressive therapy(20).

Diagnosis and correct management of spondy-lodiscitis due to uncommon pathogen are very challenging, requiring high clinical suspect, adequate radiological and laboratory assessment(21). Sometimes, laboratory test could be not clearly predictive of infection, while radiological exams could be compatible. When in doubt, an open biopsy should be taken in to account especially for low pathogenic agents, which could be considered as a contamination, if sample were not directly collected from the surgical field.

Conflicts of interest: Each author declares that he or she has no commercial associations (e.g. consultancies, stock ownership, equity interest, patent/licensing arrangement etc.) that might pose a conflict of interest in connection with the submitted article.

References

- 1. Dunn RN, Castelein S, Held M. Impact of HIV on spontaneous spondylodiscitis. Bone Jt J. 2019;101 B(5):617-620. doi:10.1302/0301-620X.101B5.BJJ-2018-0960.R1
- 2. Urrutia J, Zamora T, Campos M. Cervical pyogenic spinal infections: are they more severe diseases than infections in other vertebral locations? Eur Spine J. 2013;22(12):2815-2820. doi:10.1007/s00586-013-2995-y
- 3. Yuan Y, Gao M. Genomic analysis of a ginger pathogen Bacillus pumilus providing the understanding to the pathogenesis and the novel control strategy. Sci Rep. 2015;5(1):10259. doi:10.1038/srep10259
- Drobniewski FA. Bacillus cereus and related species. Clin Microbiol Rev. 1993;6(4):324-338. doi:10.1128/ CMR.6.4.324
- Borsa BA, Aldağ ME, Tunalı B, Dinç U, Güngördü Dalar Z, Özalp VC. [A sepsis case caused by a rare opportunistic pathogen: Bacillus pumilus]. Mikrobiyol Bul. 2016;50(3):466-470. http://www.ncbi.nlm.nih.gov/pubmed/27525402. Accessed March 4, 2019.
- 6. Bentur H, Dalzell A, Riordan F. Central venous catheter infection with Bacillus pumilus in an immunocompetent

- child: a case report. Ann Clin Microbiol Antimicrob. $2007;6(1):12.\ doi:10.1186/1476-0711-6-12$
- 7. Banerjee C, Bustamante CI, Wharton R, Talley E, Wade JC. Bacillus infections in patients with cancer. Arch Intern Med. 1988;148(8):1769-1774. http://www.ncbi.nlm.nih.gov/pubmed/3401098. Accessed March 4, 2019.
- Kimouli M, Vrioni G, Papadopoulou M, et al. Two cases of severe sepsis caused by Bacillus pumilus in neonatal infants.
 J Med Microbiol. 2012;61(Pt_4):596-599. doi:10.1099/jmm.0.033175-0
- 9. Farhat H, Chachaty E, Antoun S, Nitenberg G, Zahar J-R. Infections à Bacillus et immunodépression, à propos de deux cas. Médecine Mal Infect. 2008;38(11):612-614. doi:10.1016/j.medmal.2008.09.006
- McKillip JL. Prevalence and expression of enterotoxins in Bacillus cereus and other Bacillus spp., a literature review. Antonie van Leeuwenhoek, Int J Gen Mol Microbiol. 2000;77(4):393-399. doi:10.1023/A:1002706906154
- 11. Tena D, Martinez-Torres J a., Perez-Pomata MT, Saez-Nieto JA, Rubio V, Bisquert J. Cutaneous Infection Due to Bacillus pumilus: Report of 3 Cases. Clin Infect Dis. 2007;44(4):e40-e42. doi:10.1086/511077
- 12. Girardo M, Rava A, Aprato A, Massè A, Sabatini L, Fusini F. The treatment of spinal tuberculosis: A single center experience. Minerva Ortop e Traumatol. 2019;70(3):132-136. doi:10.23736/S0394-3410.19.03921-3
- 13. From C, Hormazabal V, Granum PE. Food poisoning associated with pumilacidin-producing Bacillus pumilus in rice. Int J Food Microbiol. 2007;115(3):319–324. doi:10.1016/j. iifoodmicro.2006.11.005
- Duc LH, Hong HA, Barbosa TM, Henriques AO, Cutting SM. Characterization of Bacillus Probiotics Available for Human Use. Appl Environ Microbiol. 2004;70(4):2161-2171. doi:10.1128/AEM.70.4.2161-2171.2004
- 15. Ledermann HP, Schweitzer ME, Morrison WB, Carrino JA. MR Imaging Findings in Spinal Infections: Rules or Myths? Radiology. 2003;228(2):506-514. doi:10.1148/radiol.2282020752
- Rava A, Fusini F, Cinnella P, Massè A, Girardo M. Is cast an option in the treatment of thoracolumbar vertebral fractures? J Craniovertebr Junction Spine. 2019;10(1):51-56. doi:10.4103/jcvjs.JCVJS_8_19
- 17. Trajkovski A, Omerovic S, Hribernik M, Prebil I. Failure Properties and Damage of Cervical Spine Ligaments, Experiments and Modeling. J Biomech Eng. 2014;136(3):031002. doi:10.1115/1.4026424
- 18. Gargiulo G, Girardo M, Rava A, et al. Clinical comparison between simple laminectomy and laminectomy plus posterior instrumentation in surgical treatment of cervical myelopathy. Eur J Orthop Surg Traumatol. 2019;29(5):975-982. doi:10.1007/s00590-019-02395-6
- 19. Girardo M, Rava A, Coniglio A, et al. Importance of polymethylmethacrylate augmentation in the treatment of thoracolumbar osteoporotic vertebral fractures. Minerva Ortop e Traumatol. 2019;70(2):65-69. doi:10.23736/S0394-3410.19.03920-1

- 20. Shad A, Shariff S, Fairbank J, Byren I, Teddy PJMAD, Cadoux-Hudson TAD. Internal fixation for osteomyelitis of cervical spine: the issue of persistence of culture positive infection around the implants. Acta Neurochir (Wien). 2003;145(11):957-960; discussion 960. doi:10.1007/ s00701-003-0129-8
- 21. Colò G, Cavagnaro L, Alessio-Mazzola M, Zanirato A, Felli L, Formica M. Incidence, diagnosis and management of sacroiliitis after spinal surgery: a systematic review of the literature. Musculoskelet Surg. 2019. doi:10.1007/s12306-019-00607-0

Correspondence:

Received: 1 July 2020 Accepted: 20 July 2020 Federico Fusini, M.D. Department of Orthopaedic and Traumatology, Regina Montis Regalis Hospital, ASL CN1, Strada S. Rocchetto 99, 12084, Mondovì (CN), Italy

Phone: 0039 0116933290 E-mail: fusinif@hotmail.com