CASE REPORT

Vertebral osteomyelitis caused by *Mycobacterium abscessus*

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SUMMARY

Mycobacterium infection caused by non-tuberculous mycobacterial (NTBM) organisms is becoming more common. Although NTBM osteomyelitis is unusual, it can occur in otherwise healthy individuals, but it is also associated with immunocompromised states, such as steroidal therapy and AIDS, and may be observed following trauma. *Mycobacterium avium* is reported to be the most common causative agent, and *Mycobacterium abscessus* has only been described in two cases. We report the case of a 44-year-old man with history of hepatitis C diagnosed with osteomyelitis of the thoracic spine caused by *M abscessus*. We present a literature review of NTBM osteomyelitis and a discussion of its diagnosis and treatment.

BACKGROUND

Mycobacterial infection can present as a localised infection or disseminated disease. The most common form of presentation is pulmonary infection. The increasing prevalence of pulmonary non-tuberculous mycobacterial (NTBM) disease and declining rates of tuberculosis (TB) represent the new epidemiology of mycobacterial disease in the USA. The incidence of TB in the USA declined to 3.8 cases/100 000 population in 2009, resulting in a decrease of 11.4% from 2008, which continued a progressive decline since the 1990s.¹ NTBM seems to have surpassed TB as the leading cause of mycobacterial lung disease in the USA. The NTBM age-adjusted average annual prevalence is 5.4/100 000 for 2004–2006, and the overall prevalence is 4.4/100 000 among men and 6.5/100 000 among women.² Localised infections, such as osteomyelitis, are unusual, and they can be associated with underlying causes. Although *Mycobacterium avium* is the most common, *Mycobacterium abscessus* is unusual. *M abscessus* has clearly been associated with pulmonary infection, especially in cystic fibrosis patients.³ Other common presentations include disseminated infection⁴ and infections of the skin,⁵ ear,⁶ prostate,⁷ breast⁸ and bone.⁹ Osteomyelitis presentations are mainly described for the mastoid,⁹ foot,¹⁰ mandibular bone,¹¹ sternum¹² and vertebrae.¹³

CASE PRESENTATION

A 44-years-old Caucasian man with a medical history of hepatitis C was evaluated for a 1-year history of chronic back pain. The pain started midback and had worsened over time, most recently radiating down to the legs as a shocking sensation and upto the middle of the thigh. Owing

Figure 1 L4–5 spinal compromise.

to worsening of pain and associated symptoms, he decided to seek medical attention. He had never experienced fever, muscle weakness, sensory loss or sphincter incontinence. He did report night sweats. His medical history was notable for a positive purified protein derivative test during childhood, for which he was treated with isoniazid for a long period. His social history was remarkable for intravenous drug abuse. He had been diagnosed with hepatitis C 2 years prior, had never received treatment and was HIV negative.

On initial evaluation, he was afebrile and haemodynamically stable. Physical examination was remarkable only for mild weakness on both lower extremities (4/5), with preserved sensation and tenderness to palpation of the lumbar area. The complete blood count and routine chemistry data were all within normal limits. Radiographs of the thoracic spine illustrated a loss of disc height at L4-S1 and partial collapse at the L5 vertebral body. MRI disclosed discitis at the L4–5 level with paraspinal,

Figure 2 Psoas muscle abscess.

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Table 1 Baseline characteristics of studies

Study	Age	Gender	Pathogen	Underlying condition	Site of involvement	Clinical manifestation	Diagnosis	Antibiotic	Duration	Surgical treatment	Outcome
Aberg <i>et al</i> ¹⁹	49	Male	MAC	AIDS	Thoracic	Back pain, sensory loss	MRI+Bx	ETB, RFP, CLT	Not reported	Yes	Full recover
	49	Male	MAC	AIDS	Thoracic	Numbness and weakness	MRI+Bx	Azitro, cipro	Not reported	Yes	Full recover
Arend <i>et al</i> (2003) ²⁰	41	Female	Atypical	Previous exposure	Not specified	Back pain	X-ray+99Th scan	Cipro, CLT, ETB	18	No	Full recover
	17	Male	MAC	None	Multiple sites	Back pain, fever	99Th scan	CLT, ETB ofloxacin, later just RFB	15	No	Full recover
	29	Male	Atypical	Previous exposure	Lumbar	Back pain, fever	MRI+Bx	CLT, ETB	6	No	Full recover
Bhatia <i>et al</i> (2001) ²¹	70	Male	MAC	None	Lumbar	Back pain, ataxia	MRI+Bx	Not reported	Not reported	Yes	Full recover
Chan <i>et al</i> (2011) ²²	16	Female	<i>M abscessus</i>	Trauma	Thoracic	Back pain	MRI+Bx	CLT	6	No	Full recover
	62	Female	MAC	Trauma	Lumbar	Back pain	MRI+Bx	CLT, ETB, clofazamine	18	Yes	Full recover
Corrales-Medina <i>et al</i> (2001) ²³	35	Male	MAC	AIDS	Thoracic	Numbness	MRI+Bx	INH, RFB, ETB, PZD, CLT	Not reported	No	Full recover
Danesh-Clough <i>et al</i> (2006) ²⁴	75	Female	<i>M xenopi</i>	None	Thoracic	Back pain	MRI+Bx	RFP, ETB	24	Yes	Full recover
Erard <i>et al</i> (2000) ²⁵	36	Female	MAC	AIDS	Lumbar	Back pain	CT+Bx	CLT, RFB, ETB	6	No	Died
Froideveaux <i>et al</i> (1996) ²⁶	28	Male	<i>M xenopi</i>	Previous spine manipulation	Lumbar	Back pain	MRI+Bx	RFB, ETB, INH, PZD, pefloxacin+CLT	18	No	Full recover
Hirakawa <i>et al</i> (2008) ²⁷	18	Female	MAC	Previous exposure	Lumbar	Back pain	MRI+Bx	RFP, ETB, kanamycin, erythromycin, SMZ, TMP	60	Yes	Full recover
Igram <i>et al</i> (1997) ²⁸	39	Male	MAC	None	Thoracic	Back pain	MRI+Bx	Cipro, erythromycin, ETB	Not related	Yes	Full recover
Kim <i>et al</i> (2011) ²⁹	75	Female	<i>M intracellulare</i>	Trauma	Lumbar	Back pain, paraparesis	CT+Bx	ETB, RFP, CLT	3	Yes	Died
Korres <i>et al</i> (2007) ³⁰	55	Male	<i>M chelonae</i>	Immunosuppressed	Multiple site	Back pain	MRI+Bx	Cipro, CLT	7	Yes	Full recover
Kulasegaram <i>et al</i> (2001) ³¹	33	Male	<i>M xenopi</i>	AIDS	Thoracic	Back pain, night sweats	MRI+Bx	RFP, RFB, INH, CLT, ETB, Cipro, amikacin, streptomycin, ETN	6	No	Full recover
Miller <i>et al</i> (1994) ³²	70	Female	<i>M xenopi</i>	None	Thoracic	Back pain, numbness	MRI+Bx	INH, PZD, cipro, ETB	24	Yes	Partial
Nalaboff <i>et al</i> (2000) ³³	50	Male	MAC	AIDS	Thoracic	Back pain	MRI+Bx	Not specified	Not reported	No	Not reported
Otaki <i>et al</i> (2002) ³⁴	51	Female	<i>M intracellulare</i>	Immunosuppressed	Lumbar	Back pain	MRI+Bx	CLT, AZT	Not reported	Yes	Not reported
Petitjean <i>et al</i> (2004) ³⁵	47	Male	<i>M xenopi</i>	None	Lumbar	Back pain	MRI+Bx	CLT	Not reported	Yes	Full recover
		Female	MAC	None	Thoracic	Back pain, sphincter incontinence	MRI+Bx	CLT	Not reported	Yes	Full recover
Pirofsky <i>et al</i> (1993) ³⁶	79	Male	MAC	Immunosuppressed	Thoracic	Back pain, fever	MRI+Bx	INH, ETB, clofazimine, amikacin, cipro	6	No	Died
Prosser <i>et al</i> (1986) ³⁷	27	Male	<i>M fortuitum</i>	None	Thoracic	Chest pain	MRI+Bx	Kanamycin, INH, RFP, ETN	24	Yes	Not reported
	51	Male	<i>M fortuitum</i>	None	Thoracic	Back pain	Not reported	Not specified	Not reported	No	Died
	30	Male	<i>M fortuitum</i>	Trauma	Lumbar	Back pain, fever, paraplegia	Not reported	INH, RFB, ETB, streptomycin, ETN, lincomycin	Not reported	Yes	Full recover

Continued

Table 1 Continued

Study	Age	Gender	Pathogen	Underlying condition	Site of involvement	Clinical manifestation	Diagnosis	Antibiotic	Duration	Surgical treatment	Outcome
	41	Female	<i>M fortuitum</i>	None	Lumbar	Back pain, fever	Not reported	RFP/2; INH+streptomycin, TCL	14	Yes	Partial recover
	18	Male	<i>M kansasii</i>	None	Lumbar	Back pain, fever	Not reported	INH, ETB, streptomycin	Not reported	Yes	Full recover
	55	Female	<i>M xenopi</i>	Immunosuppressed	Lumbar	Back pain	Not reported	INH, RFP, PZD, streptomycin	Not reported	Yes	Full recover
	43	Female	Atypical	Spine manipulation	Lumbar	Back pain	Not reported	Not reported	Not reported	Yes	Full recover
Pruitt <i>et al</i> (1993) ³⁸	17	Female	<i>M chelonae</i>	Immunosuppressed	Lumbar	Back pain	MRI+Bx	CLT	7	Yes	Full recover
Rahman <i>et al</i> (1992) ³⁹	77	Female	<i>M xenopi</i>	None	Lumbar	Back pain	CT+Bx	PZD, RFP, INH, ETB, cipro	12	No	Full recover
Rahman <i>et al</i> (2009) ⁴⁰	51	Male	<i>M chelonae</i>	Drug abuser	Thoracic	Back pain	MRI+Bx	Did not specify	Not reported	Yes	Partial recover
Rose <i>et al</i> (1982) ⁴¹	49	Female	<i>M smitiae</i>	Previous exposure	Lumbar	Back pain	Not reported	INH, RFP, ETB, kanamycin	4	Yes	Full recover
Sarria <i>et al</i> (1998) ¹³	53	Male	<i>M abscessus</i>	Drug abuser	Lumbar	Back pain, night sweats	MRI+Bx	CLT, imipenem, ceftioxin	12	Yes	Partial recover
Shimizu <i>et al</i> (2013) ⁴²	50	Male	MAC	None	Lumbar	Back pain	MRI+Bx	CLT, ETB, RFP	12	No	Full recover
	68	Female	<i>M kansasii</i>	None	Thoracic/lumbar	Back pain	MRI+Bx	INH, RFP, ETB, PZD, AZT	2	No	Full recover
	38	Female	MAC	Immunosuppressed	Thoracic	Back pain	MRI+Bx	CLT, ETB, RFB	4	Yes	Full recover
Suzuki <i>et al</i> (2011) ⁴³	67	Male	MAC	None	Lumbar	Back pain	MRI+Bx	Cycloserine, ETN	24	Yes	Full recover
Telgt <i>et al</i> (1997) ⁴⁴	56	Female	<i>M xenopi</i>	Immunosuppressed	Thoracic	Back pain	MRI+Bx	INH, CLT, cipro	9	No	Full recover
Yano <i>et al</i> (2004) ⁴⁵	33	Male	<i>M kansasii</i>	AIDS	Thoracic	Back pain	MRI+Bx	INH, RFP, PZD, ETB	Not reported	No	Full recover

MAC, *Mycobacterium avium*; Bx, biopsy; ETB, ethambutol; RFB, rifabutin; Cipro, ciprofloxacin; CLT, clarithromycin; AZT, azitromycin; INH, isoniazid; PZD, pyrazinamide; RFP, rifampicin; SMZ, sulfamethoxazole; ETN, ethionamide; TCL, tetracycline.

prevertebral and epidural soft tissue inflammatory changes (figures 1 and 2). The epidural infiltration extended into the vertebral canal posterior to L4 and L5 and compressed the subarachnoid sac. The psoas muscles were compromised bilaterally, with more extensive invasion on the left associated with a 0.5 cm abscess. CT-guided aspiration of the left psoas abscess was performed, and green purulent material was obtained. The material was submitted for culture. Gram staining was negative, and the culture did not grow any agent nor did the fungal culture. An acid-fast bacilli smear initially was negative. *Mycobacterium tuberculosis* PCR was negative, and culture grew *M abscessus*. The patient was started on triple therapy with cefoxitin, amikacin, and clarithromycin. The cultures were susceptible to amikacin, clarithromycin and linezolid but resistant to moxifloxacin, tobramycin and co-trimoxazole. The patient continued on this regimen for 6 months.

OUTCOME AND FOLLOW-UP

At 1-year follow-up, the patient presented with complete resolution of back pain and no neurological symptoms and had resumed his daily activities.

DISCUSSION

Osteomyelitis of the spine is commonly caused by *Staphylococcus aureus* and *M tuberculosis*.¹⁴ NTBM causes of osteomyelitis are very unusual. To better assess the case presentation and its relevance, we studied the main causative agents, epidemiology, predisposing underlying conditions, initial clinical presentation, diagnostic tools, treatment options and clinical outcomes for NTBM vertebral osteomyelitis. We conducted a full systematic search in PubMed and EMBASE from 1966 to July 2012. We included 'vertebral osteomyelitis' as the medical subject for the MEDLINE search, and we searched for case reports as well as case series. We rejected cases with *M tuberculosis* infection, cases that did not present or involve the vertebrae, and papers that did not report the subjects of interest. Our MEDLINE search yielded 489 articles. The EMBASE search did not yield additional studies after the elimination of duplicate results. Through a review of titles and abstracts, 463 studies were rejected for the lack of relevance.

The pooled data provided a total of 28 publications with 41 cases, as demonstrated in table 1. The mean age was 46.2 ±18 years old, and men were prevalent in most of the cases reported (53.6%). The most commonly reported agent was *M avium* (36.5%), followed by *Mycobacterium xenopi* (19.5%). Most patients did not present with a significant underlying condition (31%). Most patients did not present any underlying causative factor (36.5%), and immunocompromised patients with AIDS and steroid users were the most common predisposing underlying conditions (26.8%). The thoracic spine was the most often affected (51%) site of the spine. The diagnosis in all cases involved a biopsy of the infected site, and cultures were essential for identifying the causative agent. Regarding the therapy plan and success of treatment, the four most commonly used drugs were ethambutol (56%), clarithromycin (48.8%), isoniazid (39%) and rifampicin (36.5%). Adjunctive surgical therapy with debridement with either laminar or body resection with spine fusion was reported in the majority of the cases (65%). Clinical improvement or infection cure was reported in 35 cases, where 4 did not present with a 100% successful rate and required additional antibiotic therapy, and another four eventually died due to their underlying disease. Mycobacterial osteomyelitis diagnosis relies on isolation of the bacteria with CT-guided biopsy¹⁵ in culture with a starch-based medium. The American Thoracic Society/Infectious Diseases Society of

America and many other experts^{16 17} have suggested combined therapy with intravenous amikacin with cefoxitin or imipenem and an oral macrolide for 2–4 months for pulmonary presentation of *M abscessus*. After initial therapy, macrolide administration plus at least one other antibiotic agent to which the organism is susceptible should be used for treatment. Currently, there is no standard recommended regimen for treating *M abscessus* osteomyelitis, and the reported regimens in this study were based mainly on the pulmonary indication. Nevertheless, adjunctive surgical therapy can be applied with great success¹⁸ in selected clinical situations.

Learning points

- ▶ Few cases of vertebral osteomyelitis caused by non-tuberculous mycobacterial agents have been described.
- ▶ Vertebral osteomyelitis caused by *Mycobacterium abscessus* is very rare, and only three cases have been described in the literature.
- ▶ The diagnosis of vertebral osteomyelitis involves biopsy of the infected site and cultures.
- ▶ The therapy plan most commonly involves ethambutol, clarithromycin, isoniazid and rifampicin.

Contributors All authors were involved on the writing and review of paper. They were directly involved in patient care as well.

Competing interests None.

Patient consent Obtained.

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