Unusual presentation of more common disease/injury

Shoulder pain and dysphagia with an unexpected cause

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Summary

A 31-year-old female pharmacist of Nigerian origin, now resident in London, described 4 months of worsening left-sided subscapular pain despite the use of increasingly potent analgesia. She also described progressive dysphagia, first to solids and later to liquid foods. She remained systemically well, with no associated symptoms and normal physiological observations. In light of raised plasma inflammatory markers and chest radiography demonstrating a widened paraspinal stripe, the patient underwent CT and subsequent MRI of the thorax and upper spine. This revealed bony destruction of multiple higher thoracic vertebrae, with an associated epidural abscess mediating spinal cord impingement at T5. A large prevertebral collection spanning C7–T9 directly compressing and displacing the oesophagus was demonstrated. These findings collectively suggested spinal tuberculosis (Pott's disease); PCR confirmed the presence of *Mycobacterium tuberculosis*. The patient was successfully treated with oral anti-tuberculous chemotherapy and physiotherapy.

BACKGROUND

Globally, more than two billion people are thought to be infected by tuberculosis, with prevalence highest in sub-Saharan Africa and lowest in the developed world.¹ While pulmonary disease is the principal manifestation, spinal tuberculosis – as first described in 1782 by

Sir Percival Pott – accounts for 1% of all cases.² We here describe an unusual case of Pott's presenting as chronic shoulder pain and dysphagia in a patient who remained systemically well throughout. The case is particularly of note for certain features found rarely in Pott's, specifically the involvement of higher thoracic vertebrae and

Figure 1 Chest radiograph demonstrating widening of the left superior mediastinum with displacement of the left paraspinal stripe (arrow).

extrinsic oesophageal compression from a tuberculous abscess.

CASE PRESENTATION

A 31-year-old Nigerian national, working in London as a pharmacist since arriving in the UK 3 years ago, presented with a 4-month history of well-localised left-sided subscapular pain, which began shortly after a brief trip home. No history of trauma or other obvious precipitant was elicited. She noted that the pain was making it increasingly difficult to reach for medications and other equipment from shelves above shoulder height at the pharmacy in which she worked. She otherwise felt well, denying any sensorimotor disturbance, weight loss, fever, cough or sputum production. Her medical history consisted solely of sickle cell trait. Examination revealed mild tenderness over the infero-medial aspect of the left scapula, along with pain and some limitation of movement beyond 90 degrees of flexion or abduction of the left shoulder. There was no evidence of neurological deficit, active synovitis, deforming arthropathy or lymphadenopathy. Simple analgesia to treat a presumed muscular injury initially proved successful.

The patient represented 4 weeks later complaining principally of dysphagia. This had initially been for solids only, but now was starting to affect her intake of liquid foods too. She denied any difficulty in the actual action of swallowing, but described the sensation of food 'getting stuck' whenever she tried to do so. In addition, her shoulder pain

had returned and was now severe enough to keep her awake at night. Initial investigations included blood tests (revealing an erythrocyte sedimentation rate of 45 mm/h and C reactive protein of 46 mg/l) and chest radiography showing clear lung fields but widening of the left superior paraspinal stripe (figure 1), highly suggestive of posterior mediastinal pathology. Given this suggestion in the context of chronic bone pain, the patient was referred for an urgent orthopaedic opinion.

Review of her social history by the orthopaedic team revealed that she lived alone, had no recent sexual partners and did not smoke or use illicit drugs. She denied any unwell contacts. Thorough systems enquiry revealed nothing new of note. Examination was unchanged to a month earlier, and physiological observations remained normal.

INVESTIGATIONS

Further blood tests revealed vitamin D deficiency (25-hydroxycholecalciferol <10 ng/ml). Full blood count, urea and electrolytes, liver function tests, serum electrophoresis and plasma-corrected calcium were all within acceptable limits. Extractable nuclear antigens were not detected, and HIV serology was negative.

CT (and subsequent MRI) of the thorax and spine revealed almost complete anterior vertebral body destruction in the region of T2–T5 with marked loss of the intervening disc spaces. Also noted was a large prevertebral collection spanning C7–T9 with associated epidural abscess

Figure 2 Transverse CT demonstrating fluid density paraspinal collection (arrow 1), bony destruction of the T2 vertebral body (arrow 2) and oesophageal compression (arrow 3).

at T3–T6 and clear evidence of spinal cord impingement at T5 (figure 2). Abscess-mediated oesophageal compression and displacement was noted at multiple levels (figures 2 and 3). Clear lung fields and normal abdominal viscera confirmed a complete lack of extra-spinal disease, with no mediastinal lymphadenopathy noted aside from a single necrotic node at the subcarinal level. Collectively, these appearances were felt to be most consistent with spinal tuberculosis.

Spinal aspiration via a posterior approach was performed using a 14G catheter directed under CT-guided fluoroscopy. This yielded 30 ml of a green viscous solution rich in leucocytes. Gram staining and staining for acid-fast bacilli along with standard bacterial culture were all negative; however, mycobacterial culture and PCR confirmed the presence of *Mycobacterium tuberculosis*.

DIFFERENTIAL DIAGNOSIS

Prior to aspiration, abscess formation secondary to pyogenic bacteria was considered high on the list of differential diagnoses. However, such patients often reveal

an obvious source of possible bacterial inoculation (eg, recent surgery) during review of the history and would be expected to have much more significant constitutional upset in the presence of such a burden of disease as demonstrated here. Brucellosis is a zoonotic infection capable of a very wide variety of manifestations, including osteomyelitis, but again often first presents as 'pyrexia of uncertain origin' rather than with localising features. Given the history of sickle cell trait, a salmonella osteomyelitis was also considered, but again felt less likely in the absence of systemic upset and the extraordinary size of the paraspinal collection.

The presence of raised inflammatory markers and posterior mediastinal pathology might also be explained by malignancy. Extrinsic compression of the oesophagus through the mass effects either of solid organ tumours or enlarged lymph nodes (reflecting either tumour metastasis or primary lymphoproliferative disease) is well characterised. Similarly, pathological fracture of the vertebrae and spinal cord syndromes are common complications of bony metastasis.

Figure 3 Sagittal MRI T2-weighted image with gadolinium enhancement. An enhancing paraspinal collection is seen extending into the spinal canal, causing spinal cord impingement (hollow arrows) and oesophageal displacement (filled arrows).

TREATMENT

The patient was started immediately on combination oral anti-tuberculous chemotherapy (isoniazid, rifampicin, pyrazinamide and ethambutol); at 2 months, she was able to reduce this regimen to combined isoniazid and rifampicin only. Later microbiology results confirmed the organism to be fully sensitive to all standard anti-tuberculous medications. In keeping with guidelines published in the UK, she was treated with 6 months of chemotherapy in total.³ There was neither clinical nor radiological suspicion of meningeal tuberculous disease, and as such no indication felt for either a more prolonged course of treatment or the addition of corticosteroid therapy.

At the time of diagnosis, the patient was transferred to a tertiary orthopaedic centre. It was felt that surgical intervention to stabilise the concerned vertebrae was not warranted in the absence of clinical features of true spinal cord compression. Instead, she was started on a course of intensive physiotherapy with regular orthopaedic follow-up.

OUTCOME AND FOLLOW-UP

At the time of writing, the patient has recently finished her course of chemotherapy. Her dysphagia has long since resolved and her shoulder pain continues to gradually improve. She is still requiring physiotherapy for the latter problem, but has been able to return to full-time work. A recent repeat MRI demonstrates no evidence of a residual collection; some vertebral body inflammation is still present, but the oesophagus and spinal cord appear to be fully intact.

DISCUSSION

Pott's disease represents a diagnostic challenge to even the most experienced clinicians, with up to 40% of cases being initially misdiagnosed due to the non-specific nature of its presentation.⁴ Our patient's presenting symptom of atraumatic chronic focal pain out of context to clinical findings is often typical of Pott's,⁵ with other common presentations including leg weakness, gibbus and the presence of a palpable spinal mass.⁶ Surprisingly, pulmonary involvement and systemic upset are often absent in Pott's, with both occurring in less than half of cases,^{4 7} making the diagnosis even more elusive. The fact that the first clue to the diagnosis here was a radiological subtlety rather than clinical suspicion is somewhat typical of cases in the developed world, and one with profound prognostic implications. Specifically, it is a reflection of this trend towards late diagnosis of Pott's in areas of low incidence that means up to 70% of all new cases already demonstrate evidence of spinal cord compression at diagnosis.⁴

Review of the epidemiology of Pott's disease suggests that our patient fits the typical profile of cases diagnosed in the developed world. A retrospective review from France of all musculoskeletal tuberculosis cases diagnosed between 1980 and 1994 found that 68% of patients were foreign-born,⁸ the vast majority of whom were born in Africa. Interestingly, none in this series were HIV positive, suggesting that in contrast to pulmonary tuberculosis, the incidence of spinal tuberculosis has been unaffected

by the HIV pandemic. Assuming that this patient had undergone primary infection when still in Nigeria and taking into consideration that she too was HIV negative, it is interesting to hypothesise what risk factors made her prone to reactivation of tuberculosis. Given that vitamin D deficiency has been shown in vitro to be associated with reduced phagocyte activity,⁹ one explanation implicates the patient's almost undetectable plasma 25-hydroxycholecalciferol concentrations. A series of 210 patients with active tuberculosis in London found that 76% were deficient in vitamin D, with the majority of these patients being of non-South East Asian or non-European descent.¹⁰ Furthermore, Africans immigrating to western countries tend to become deficient in vitamin D,¹¹ which may impair the host defence to latent tuberculosis.

Certain elements of the case are particularly unique. Pott's disease most commonly involves lower thoracic, thoracolumbar and lumbosacral vertebrae^{12 13} but rarely higher levels as encountered here; anterior vertebral body destruction with subsequent disc narrowing and prevertebral collections is, however, typical.¹³ Tuberculosis presenting as dysphagia is also unusual – in the largest series of such patients in the literature (a total of 14 patients), radiological findings suggested that the underlying mechanism of dysphagia was extrinsic oesophageal compression by subcarinal lymph nodes in seven patients and tracheo-oesophageal fistula in four.¹⁴ The only previously reported cases of dysphagia secondary to direct compression by a Pott's cold abscess are seen in a series of seven patients with the exceptionally rare diagnosis of cranio-cervical Pott's,¹⁵ with dysphagia tending to be a late feature in such cases. To our knowledge, ours is the first reported case in the literature of a thoracic Pott's spinal collection causing dysphasia by direct oesophageal compression, and as such demonstrates the unparalleled size of the collection in this patient. With such a burden of tuberculous disease, it makes it all the more surprising that the patient had such vague symptoms, minimal systemic inflammatory response and no true spinal cord compression at presentation.

Optimal management of the patient with Pott's involves input from physicians, orthopaedic surgeons and neurosurgeons, but the exact role that surgery has to play in the disease remains controversial. Where neurological signs are present some would advocate immediate surgery in combination with standard anti-tuberculous chemotherapy in order to achieve immediate skeletal stability⁶; however, the present mainstay of treatment is for adoption of a more conservative approach wherever possible. In a retrospective review of 70 patients with thoracic tuberculosis, 44 of whom had abscess formation and 7 of whom had signs of cord compression, a conservative approach was successful in all but one patient.¹⁶ The exclusion criteria used in this study – namely, advanced neurological deficit (grade 3/5 power or less), symptoms refractory to anti-tuberculous chemotherapy and significant kyphosis – are now widely regarded as the major absolute indications for surgery.¹⁶ Although long courses of anti-tuberculous chemotherapy were previously the norm, it is now broadly accepted that this is no more efficacious than a 6-month course.¹⁷

Learning points

- ▶ The diagnosis of Pott's disease should be considered in all patients from high-risk populations presenting with an unusual constellation of musculoskeletal symptoms and those from other systems.
- ▶ As patients with Pott's often lack the classic constitutional upset that accompanies pulmonary disease, diagnosis is often delayed until after the onset of cord compression with potentially devastating consequences.
- ▶ Dysphagia is a rare but important possible manifestation of tuberculosis, and a symptom that tends to resolve well with appropriate treatment.
- ▶ Vertebral deposits and bony destruction in the context of pain, weight loss and neurological symptoms is a common presentation of metastatic malignancy. With the ever-increasing incidence of tuberculosis, Pott's must be on the differential in those fitting the epidemiological profile.
- ▶ Anti-tuberculous chemotherapy remains the mainstay of treatment in Pott's, even where abscess formation and impingement onto spinal structures is seen.

Competing interests None.

Patient consent Obtained.

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