CASE REPORT

Delayed diagnosis of chronic postoperative sternal infection: a rare case of sternal tuberculosis

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SUMMARY

Sternal osteomyelitis secondary to mycobacterium tuberculosis (TB) is rare, with <1% of musculoskeletal TB cases reported. The recurrent scenario is unresolving infection and delayed diagnosis. A 75-year-old woman presented with a persistently discharging sternal wound 10 months after coronary artery bypass grafting. Multiple antibiotics, wound debridement and removal of sternal wires was attempted; however, progression to local osteomyelitis and sternoclavicular joint destruction occurred. Tissue biopsies were finally sent for mycobacterial culture testing positive for *Mycobacterium tuberculosis*. High index of suspicion is necessary for diagnosis of sternal tuberculosis, confirmed through timely microbiological investigations. MRI may identify soft-tissue and bone oedema characteristic of TB osteomyelitis. This patient had no TB risk factors. The source of infection is unclear and warrants further investigation. Sternal TB osteomyelitis is uncommon and largely reported through case reports, thus management and indications for surgery remain undefined. If sensitive, standard TB four-drug regimen may be trialled.

BACKGROUND

This patient was unfortunately reviewed and treated with inadequate antibiotics for 6 years by both the cardiothoracic surgery team and infectious disease team before diagnosis of the causative organism and appropriate treatment was commenced. The aim of this case report is to create awareness and highlight the importance of timely mycobacterium cultures in cases of non-resolving infections of the sternum.

CASE PRESENTATION

A 75-year-old woman underwent uncomplicated coronary artery bypass graft (CABG) surgery that involved harvesting and grafting of the left internal mammary artery. Her postoperative stay in hospital was uneventful. She was discharged from care following a satisfactory 6-week clinic review.

She was referred back for surgical review 10 months later when her sternal wound showed signs of infection and broke down. There was dehiscence of the wound in the superior aspect with discharge of frank pus. Clinical examination revealed a deep sinus 2×2cm in size with underlying necrotic material. Wound swabs taken for routine microscopy and cultures revealed no growth. A CT scan confirmed a sinus tract extending to the first sternotomy wire and dehiscence of the underlying bone leaving a 2cm gap between the hemisternal edges.

Although no infectious growth was identified in the wound swabs, an empirical trial of antibiotics was attempted.

As the infection persisted into the 18th month following the initial CABG surgery, surgical management was carried out by way of removal of the first sternal wire and debridement of the wound. Laboratory samples from the wound and sternal wire cultured positive for *Staphylococcus aureus* and *Acinetobacter*. Treatment with intravenous ceftriaxone and oral ciprofloxacin to cover for *Acinetobacter* was administered, under the guidance of the infectious disease unit. A long course of doxycycline and intravenous tigecycline was also administered.

Persistent inflammation led to an ultrasound (US)-guided biopsy 4 years following the initial CABG surgery, which identified amorphous granular debris but no malignant cells. Repeat CT scan at this stage revealed local osteomyelitis and disruption of the right sternoclavicular joint. At this point, the surgical team removed the remainder of the sternal wires and conducted a further wound debridement including excision of an abscess cavity. Histopathology identified the presence of granulomata on a background of chronic inflammation on two occasions; however, Ziehl-Neelsen and periodic acid-Schiff-diastase (PASD) stains for acid-fast bacilli were negative and therefore the samples were not sent for mycobacterial cultures.

Further antibiotic therapy was administered with little effect. There were nine separate admissions for wound debridement and intravenous antibiotic therapy with little success. Re-sternotomy and second-look operations were avoided in view of risk of damage to grafts performed in the initial CABG. The wound was not large enough for negative pressure therapy.

Six years postoperatively, there were still signs of chronic inflammation and persistent ooze from the wound. Tissue biopsies from the sternal wound were sent for mycobacterial cultures, prompted by reports of infection with *Mycobacterium chimaera* transmitted from heater-cooler units incorporated into the cardiopulmonary bypass machine.¹ This tissue biopsy was positive for mycobacterium tuberculosis (TB).

There were no obvious TB risk factors or family history of TB. Although the patient was brought up on a farm, she did not recall drinking unpasteurised milk and had no travel history. She had received BCG vaccine as a young girl as per protocol at the time. Of note, she was not immunosuppressed at any

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Figure 1 Sinugram showing extension of contrast into sinus of about 2 cm deep.

point during treatment for this infection. The patient remained systemically well and on diagnosis was started on quadruple antibiotic therapy (rifampicin, isoniazid, pyrazinamide, ethambutol) for tuberculosis treatment. As per trust guidelines, isolation was not required for non-respiratory tract TB.

INVESTIGATIONS

Microbiology

The key diagnostic investigation in diagnosis of tuberculosis is a positive culture. From the outset, a number of swabs were taken from the wound site. As explained, these were not sent for mycobacterial culture and were positive for *S. aureus* and *Acinetobacter*. The antimicrobial treatment of these organisms was guided by the infectious disease unit.

Elevation in inflammatory markers such as C reactive protein coincided with operative procedures with the highest recorded level just over 110 mg/L.

Histopathology/microbiology

An US-guided biopsy was also carried out at this stage and samples sent for microbiology and histopathology to exclude malignancy. No malignant cells were detected; however, the presence of granulomata instigated Ziehl-Neelsen and PASD stains, which were found to be negative.

Ziehl-Neelsen staining is severely limited in the diagnosis of TB having low sensitivity and is unsuccessful if mycobacteria are less than $10^4/\text{mL}$.²

PCR and Mantoux testing was not carried out as initially there was little suspicion of TB.

Radiological imaging

A sinugram was carried out in the initial stages to identify the extent of the sinus. This is depicted in [figure 1](#) with extension of contrast along 2 cm down to the sternum.

Repeated CT scans were carried out to assess progress of the sternal infection, with evidence of destruction of the right sternoclavicular joint at 4 years as depicted in [figure 2](#).

Figure 2 Sections from CT scan showing osteomyelitis and disruption of the right sternoclavicular joint marked by a red arrow.

The sinugram supported the decision for removal of the first sternal wire and debridement, while persistent deterioration as evidenced on CT scan prompted removal of the rest of the wires and further wound debridement. As discussed below, MRI would have been more adequate to identify bone oedema, which is a typical characteristic of TB osteomyelitis.

TREATMENT

The mycobacterium was not resistant to standard quadruple antibiotic therapy (rifampicin, isoniazid, pyrazinamide, ethambutol) for tuberculosis and the combined tablet was started with good effect. This was administered for 2 months, after which combined rifampicin and isoniazid was given for 4 months. During the duration of treatment, rivaroxaban (for chronic

atrial fibrillation) was switched to daily enoxaparin. The patient expressed a personal preference for enoxaparin over warfarin.

OUTCOME AND FOLLOW-UP

The patient was regularly followed up in infectious disease outpatient clinic and cardiothoracic surgery outpatients with excellent results and resolution of infection. The function of the sternoclavicular joint was preserved and the patient was able to return to work.

DISCUSSION

Sternal osteomyelitis secondary to mycobacterium tuberculosis is rare and makes up less than 1% of musculoskeletal TB cases reported.^{3,4} The spine and hip are the more common skeletal sites for TB infection.⁵ The recurrent scenario in the scant number of case reports is non-resolving infection and delayed diagnosis.

Studies from the Asian and African subcontinents report the largest number of patients with TB on a global scale³; however, even in such endemic regions, TB infection of the sternum is rare.⁶ Interestingly, this patient has no history of possible exposure to TB and therefore the source of the infection is unclear. In light of recent reports of mycobacterium chimaera infections secondary to heater-cooler units of the cardiopulmonary bypass machine,¹ the source may warrant further investigation.

Diagnosis of sternal TB infections are mainly through timely microbiological investigations, with cultures being more reliable than medical imaging. From the latter, CT or MRI techniques are more indicative than chest X-rays in forming a possible diagnosis. MRIs are particularly able to identify bone and soft-tissue oedema.⁶ Increased intensity of normal bone marrow on T2-weighted imaging is a typical characteristic of TB osteomyelitis.⁵

The sternal body and manubrium are the sections of sternum most commonly affected by TB infection.⁴ This is followed by sternotomy sites and the inferior aspect of the sternal body.³ Infection at the sternoclavicular joint, as evidenced by this patient, is less frequent.

On overall review, apart from her age, there were no other factors associated with increased risk of sternal infection such as insulin-dependent diabetes, bilateral internal mammary grafting or re-exploration for bleeding among others.⁷

This case report describes a mycobacterium sensitive to the recognised TB drugs and therefore the patient was started on a standard four-drug regimen. This has shown to be effective in most patients with over 25% requiring concomitant surgical intervention.^{3,4} Indications for surgery are at present unclear due to lack of long-term follow-up and low case numbers.⁶

On diagnosis, the local trust policy for the management of tuberculosis was used in planning management. This is based on the National Institute for Health and Care Excellence clinical guideline 117 Tuberculosis: clinical diagnosis and management of tuberculosis, and measures for its prevention and control. Isolation and identification of patient contacts was not necessary for a non-respiratory tuberculosis infection.⁸

In conclusion, a high index of suspicion is necessary for diagnosis of sternal tuberculosis. Although rare, early cultures for TB

can avoid a delay in diagnosis and commencement of targeted therapy, which occurred in the present case. Furthermore, while surgical intervention for debridement and/or excision of sinuses seems to be a common theme in cases published, good results are reported with the four-drug antituberculous regimen in cases where the mycobacterium is not already resistant to these antibiotics. Investigation of the possible source for mycobacterium tuberculosis infection of the sternum may be warranted in patients with no risk factors.

Learning points

- ▶ A high index of suspicion is key for diagnosis of mycobacterium tuberculosis (TB) infection in a patient with no risk factors. Particularly in similar cases of non-resolving infection, patients should be investigated for possible mycobacteria and other uncommon infection sources.
- ▶ MRI is a useful diagnostic tool in identifying intensity in the bone marrow typical of osteomyelitis secondary to TB. This was not used in this case scenario.
- ▶ Timely microbiological investigations are crucial for diagnosis.
- ▶ Management involves the use of a multidisciplinary team with a role for infectious disease physicians as well as cardiothoracic surgeons. Case reports have indicated that wound debridement and targeted antibiotic therapy in tuberculosis infection of the sternum have both shown good effect, singularly and in combination.

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