CASE REPORT

Actinomycotic osteomyelitis of the cranial vault presenting with headache: an unusual presentation

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SUMMARY

A case of left parietal calvarial actinomycotic osteomyelitis in a young woman is described. She had no predisposing illnesses. She had delivered a live child at term and presented in the puerperal period. No extracranial focus of infection was identified. She responded well to a combination of surgery and medical therapy and had an excellent outcome. The authors emphasise the importance of establishing a histopathological diagnosis since radiological signs are non-specific and unreliable.

BACKGROUND

Actinomycosis is a chronic suppurative bacterial infection caused by the Gram-positive bacillus *Actinomyces israelii*.^{1–3} The most common sites of infection are the cervicofacial area (63%), abdominal wall (22%) and the thorax (15%).^{4–6} Calvarial and intracranial involvement are exceedingly rare (2% of all cases).⁴ An atypical clinical presentation of actinomycosis with osteomyelitis of the cranial vault in a young immune-competent woman is described and the relevant literature is reviewed.

CASE PRESENTATION

A 32-year-old woman presented to the hospital 2 weeks after she delivered a healthy child at term. She developed mild (visual analogue scale (VAS) score=2), persistent, aching left hemicranial headache soon after delivery. She was initially thought to have migraine and was treated elsewhere with a combination of analgesics and sumatriptan. However, her headache grew progressively worse over a week (VAS=7). She developed nausea and had occasional episodes of vomiting in relation to the headache. She had no other symptoms and gave no history of trauma to the head. She gave no history of fever, seizures, photophobia or phonophobia. The headache was not associated with physical activity; there was no diurnal variation in pain. Her medical history was unremarkable; she gave no prior history of migraine or headache. Ocular fundi were normal. Systemic examination was normal and no neurological deficits were detected. An initial clinical diagnosis of cortical venous thrombosis was considered at this time.

INVESTIGATIONS

Blood investigations revealed haemoglobin to be 9.4 g% and the total leucocyte count to be marginally elevated at 12 600/mm³. Serological tests for HIV 1 and 2 were negative. A cranial CT revealed

thickening of the bone of the left parietal skull vault with obliteration of the diploic space (figure 1A, B). There was gross irregularity of the contour of the inner table of the skull, with hair-like bone projections into the epidural space being seen. There was an underlying epidural fluid collection that did not enhance on contrast administration (figure 1C).

DIFFERENTIAL DIAGNOSIS

The possibility of a haematological malignancy was considered, in view of the anaemia and radiological evidence of expansion of the diploic space. A possible diagnosis of chronic osteomyelitis was also considered due to the presence of an enhancing epidural collection. However, in the absence of preceding trauma, this was difficult to explain.

TREATMENT

Drainage of the epidural fluid collection and biopsy of the involved bone were performed via a left parietal burr hole. The epidural fluid was clear, serosanguinous and odourless. Histopathological examination and Gram staining of the curetted bone material revealed Gram-positive, paraminosalicylic acid positive bacteria, diagnostic of *A. israelii* (figure 1E).

OUTCOME AND FOLLOW-UP

The patient was administered intravenous cloxacillin for 1 week (500 mg intravenously every 6 h). Subsequently, she was treated with oral phenoxymethyl penicillin for 6 months. Her headache gradually subsided and a repeat cranial CT after 6 months revealed near complete resolution of the radiological abnormalities, including the irregularity in the inner table of the skull (figure 1D).

DISCUSSION

The *Actinomyces* are Gram-positive, anaerobic, filamentous bacteria that normally colonise the vagina, colon and mouth. They cause chronic skin and soft tissue infections when inoculated into sites other than their natural habitats.^{1–4} The most frequent site of infection is the craniofacial area; intracranial involvement or spread of infection is very rare.^{4–6} Intracranial involvement, when encountered, probably occurs either via haematogenous dissemination or due to contiguous spread of infection from the paranasal sinuses, middle ear or teeth.⁷

Among the intracranial lesions caused by *Actinomyces*, brain abscess is the most common, (67%) followed by disseminated meningoencephalitis

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Figure 1 (A) Preoperative CT axial section with bone windows showing the irregular calvarial bone. The overlying soft tissue swelling of the scalp can also be appreciated. (B) Coronal reconstruction of the involved areas, showing the extent of bone involvement. (C) The thin epidural collection is evident. (D) CT after 6 months of treatment demonstrating a normal bone contour. (E) H&E-stained section ($\times 100$) showing the basophilic bacterial cell surrounded by an inflammatory infiltrate.

(13%) and subdural empyema (7%).⁷ Skull base and temporal bone osteomyelitis also have been reported, secondary to spread of infection from the oral cavity.^{7 8} However, epidural infection and calvarial osteomyelitis are by far the rarest manifestations. Only a few reports of isolated calvarial osteomyelitis exist in the literature.^{1 4 9} Calvarial lesions are generally indolent and may present with intractable epilepsy.⁴ Radiological findings are usually atypical and non-specific and the only method of establishing a diagnosis is histopathological examination of involved bone.^{10 11}

Our patient did not have any obvious risk factors or an extra-calvarial primary source of infection. It is probable that the state of relative immunosuppression in the puerperal period (due to hormonal changes) might have precipitated the haematogenous spread of the organism from its ambient existence in the genital tract.¹² There is no other ready explanation for such an infection. This makes the present case unique, since nearly all reported cases of calvarial or cerebral actinomycosis had a primary extracranial focus of infection.

The mainstay of management for actinomycosis is medical therapy.¹³ Surgical drainage of a pus collection is warranted only if the collection is large or to establish a diagnosis.^{13–16} The antibacterial agent of choice is penicillin.¹³ The exact duration of treatment required is not well established. However, these infections generally require prolonged therapy with oral

penicillin to effect a cure.^{4 13} The duration of therapy may be as long as 6 months and may extend even longer depending on the radiological resolution.

Competing interests None.

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Learning points

- Calvarial osteomyelitis due to actinomycotic infection is a rare entity but should be considered in the differential diagnosis of atypical osteomyelitis.
- Patients may present with chronic headache and no other symptoms.
- Early diagnosis, combined with surgical and aggressive medical therapy can result in a cure.
- Since there are no radiological signs specific to this entity, histopathological examination is the mainstay of establishing the diagnosis.

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