Case Report

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Mandibular *Actinomyces* osteomyelitis mimicking osteosarcoma

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Introduction: *Actinomyces* is a relatively uncommon cause of infection of the head and neck. However, its presentation is usually atypical and can be difficult to recognize. In this case report, we present the diagnostic and therapeutic challenges that can arise from cervicofacial actinomycosis.

Case presentation: A 26-year-old woman from Northern Manitoba in her second trimester of pregnancy presented to the emergency department with a 2-month history of progressive pain and swelling of her right mandible. Imaging revealed lytic lesions involving the angle and ramus of the right mandible with possibility of osteosarcoma, and she was taken for hemimandibulectomy. Intraoperative specimens revealed *Actinomyces odontolyticus*. She was subsequently started on piperacillin–tazobactam that was later stepped down to amoxicillin–clavulanate with radiographic and clinical improvement of her osteomyelitis.

Conclusion: Cervicofacial actinomycosis may mimic several neoplastic, granulomatous and infectious conditions, and often require tissue culture for diagnosis. Therapy often necessitates a combination of surgical resection and prolonged medical therapy, which is tailored to the burden of disease in the individual patient.

Keywords: cervicofacial actinomycosis; osteomyelitis; penicillin.

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Introduction

Actinomyces are filamentous Gram-positive microaerophilic bacilli that are commensal organisms of the oropharynx, gastrointestinal tract and urogenital tract. These organisms are well known to cause odontogenic and cervicofacial infections, including gingivitis, periapical abscesses, periodontitis and surrounding soft tissue infection, but occasionally they are also the causative organism in pneumonia, empyema, abscesses, osteomyelitis and pericarditis.

Osteomyelitis of the mandible is a rare complication of odontogenic infections that frequently develops from contiguous spread of a chronic dental infection with *Actinomyces*. It presents as an indolent course with swelling of the surrounding soft tissue that may or may not be painful. Diagnosis is particularly challenging, as *Actinomyces* osteomyelitis closely resembles other infectious and noninfectious aetiologies, both clinically and radiographically. Diagnosis is confirmed by demonstration of actinomycotic

granules in necrotic bone tissue and growth on anaerobic culture obtained from surgical biopsy.

We describe a case of mandibular *Actinomyces* osteomyelitis that posed a diagnostic challenge.

Case report

A 26-year-old woman from Northern Manitoba in her second trimester of pregnancy presented to the emergency department with a 2-month history of progressive pain and swelling of her right mandible. About 1 month prior, she had presented to her dentist with similar symptoms. She was prescribed a short course of amoxicillin without demonstrable improvement in her symptoms. On initial examination, she had extensive dental caries with several missing teeth and significant swelling over the angle of the mandible extending to the temporomandibular joint. Sub-mandibular lymph nodes were noted without tenderness on palpation. There were no fistulizing tracts observed.

Fig. 1. Panoramic radiograph showing right mandibular osseous lesion and cortical erosion, and poor dentition.

Investigations

A panoramic radiograph of her mandible showed osseous and cortical erosion on the right ramus (Fig. 1). A computed tomography scan of her facial bones showed a lytic lesion involving the angle and ramus of the mandible extending to the right mandibular condyle with extension of the lesion into the adjacent masseter and pterygoid muscles (Fig. 2). The deep cervical and submandibular lymph nodes were not enlarged on imaging.

Maxillofacial surgery was primarily considered because of suspected osteosarcoma of the mandible, and the patient was taken into the operating room for an extensive surgical procedure. During this procedure, significant granulation tissue was seen on the anterior border of the ramus with a lytic loculated lesion seen underneath the periosteum from which frank purulent discharge was expressed. Several areas of significantly decorticated and rough osteomyelitic bone were debrided.

Diagnosis

Intraoperative culture of the mandible revealed *Actinomyces odontolyticus*, which was sensitive to penicillin. Intraoperative pathology specimens of the affected bone revealed intense acute and chronic inflammation. Special stains (including Gomori's methenamine silver stain) showed no micro-organisms.

Treatment

Three weeks after the mandibular debridement, a hemimandibulectomy with mandibular and temporomandibular joint reconstruction was performed. The patient was treated with piperacillin–tazobactam for 6 weeks and then stepped down to amoxicillin–clavulanate for a further 6 months.

Outcome and follow-up

Subsequent computed tomography imaging confirmed interval regression of the extent of osteomyelitis. She

subsequently underwent routine total replacement of the temporomandibular joint and has continued to remain well on serial follow-up visits.

Discussion

Although cervicofacial *Actinomyces* osteomyelitis is uncommon in an immunocompetent host, it should be in the differential diagnosis when there is a suspicion of contiguous odontogenic soft tissue infection. The most common inciting events are dental caries, dental manipulation and oromaxillofacial trauma facilitating mucosal breakdown and spread of infection (Volante *et al.*, 2005). Other risk factors for acquiring *Actinomyces* infections include poor oral hygiene, diabetes mellitus, steroid and bisphosphonate therapy, leukaemia with chemotherapy, human immunodeficiency virus co-infection, prior transplant with immunosuppression and heavy alcohol intake (Wong *et al.*, 2011).

Several features in the presentation of this case were suggestive of *Actinomyces* infection. The chronicity of presentation was characteristic: a painless, fluctuant mass progressively enlarging over the course of months (Wong *et al.*, 2011). The incomplete response to a short course of empiric antibiotics may have represented temporary improvement with inevitable relapse (Russo, 2015). Actinomycosis is known to spread contiguously, crossing tissue planes, progressing from periodontitis to myofasciitis and osteomyelitis as observed here. It sometimes also involves the pharynx, larynx, tonsils and paranasal sinuses. Although not seen in this patient, sinus tracts within the mass may develop and then heal over time (Russo, 2015). Unlike in most cervicofacial

Fig. 2. Computed tomography showing lytic osseous lesions of the ramus of the right mandible, in transverse view.

infections, regional lymphadenopathy is uncommon (Ganepalli *et al.*, 2015).

Submandibular involvement is the most common presentation of cervicofacial actinomycosis; less frequently, the temporomandibular joint maybe involved among other locations. The diagnosis is particularly challenging as *Actinomyces* osteomyelitis mimics several infectious and noninfectious pathologies. The differential diagnosis includes osteosarcoma, metastases, lymphoma, and diseases involving granuloma formation such as Langerhans cell histiocytosis, sarcoidosis and tuberculosis. Computed tomography and magnetic resonance imaging are useful in determining the extent of infection and for preoperative planning. The challenge is to suspect and treat *Actinomyces* infection in order to avoid unnecessary surgery (Russo, 2015; Bartkowski *et al.*, 1998).

In light of this, direct specimen culture is the cornerstone for diagnosis of *Actinomyces*. The ideal specimen contains pus or tissue and is obtained prior to the initiation of antibiotics. Swab specimens should be avoided. Specimens are cultured on selective anaerobic culture medium and may take up to 3 weeks to grow (Russo, 2015; Wong *et al.*, 2011). Histopathological findings suggestive of *Actinomyces* infection are Gram-positive filamentous rods and sulfur granules, which are collections of inflammatory cells surrounded by hyphae, degenerated bacteria and proliferating fibroblasts. It is important to note that sulfur granules are commonly missed, which may have occurred in this case (Russo, 2015; Kubo & Osada, 1980). Gram, Gomori methenamine silver and Giemsa stains are helpful for visualization of these features (Brook, 2008).

The management of cervicofacial actinomycosis includes antibiotics with or without surgical debridement as even in extensive infection antibiotics alone may be sufficient (Russo, 2015). A recent retrospective review of resistance patterns in *Actinomyces* species shows near complete susceptibility to β -lactam antibiotics with and without a β -lactamase inhibitor, carbapenems, tetracyclines and vancomycin. In contrast, isolates were almost completely resistant to metronidazole (Steininger & Willinger, 2015). As odontogenic infections are often polymicrobial, the initial antibiotic choice should be appropriately broad with 4–6 weeks of intravenous antibiotics and followed by step down to broad oral antibiotics. A first line regimen may consist of piperacillin–tazobactam to cover both aerobes and oral anaerobes and β -lactamase producers with step down to an oral antibiotic with similar coverage such as amoxicillin–clavulanate.

Historically, surgical debridement has played a role where there are areas of necrotic tissue, fistulizing sinus tracts and cases in whom malignancy is difficult to exclude such as in this case (Wong *et al.*, 2011). Evidence from several case reports of actinomycosis suggests that medical therapy alone may be sufficient for cure of even severe infections (Fu & Tsai, 2010; Colmegna *et al.*, 2003; Hawnaur *et al.*, 1999). While the traditional approach is to treat with antibiotics for up to 12 months, this prolonged duration may not be necessary. Treatment regimens should be individualized based on individual clinical presentation. Factors that affect duration of treatment include initial burden of disease, the site of infection, extent of surgical debridement and the clinical and radiological response to therapy. Close follow-up is essential in determining the trajectory of the treatment course and the need for surgical revision, especially, in patients where compromised immunity or socio-economic factors have contributed to the burden of disease.

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