Case Report

Submandibular gland actinomycosis: A rare disease – Worth to mention

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Abstract

Cervicofacial actinomycosis (AM) is a well-documented entity; however, primary AM of the submandibular gland is infrequent. The diagnosis is difficult due to its nonspecific clinical presentation and it usually mimics chronic granulomatous infection or malignant lesion. We report the case of a young female with AM of submandibular gland, presented as recurrent infection of submandibular gland, underwent its excision and confirmed on microscopy as *Actinomyces*.

Keywords: *Actinomyces*, actinomycosis, granulomatous, submandibular gland

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INTRODUCTION

Actinomyces is a Gram-positive anaerobic bacterium that inhabits the mouth, digestive and urogenital tracts. Cervicofacial actinomycosis (AM) is the most frequently encountered infection and commonly involves the parotid and the mandible.[1] They are often misdiagnosed due to nonspecific clinical presentation and similarity with infectious lesion or infrequently malignancy.[2] Hereby, we report a rare case of AM of submandibular gland that was clinically diagnosed as suppurative infection.

CASE REPORT

A 19-year-old girl presented with complaints of a swelling in the right submandibular region for the past 5 years. Swelling was gradually progressive in size and without any accompanying pain. She had neither pain in swelling nor change in size of swelling following food intake. There was no history of fever, cough, nasal or throat complaints. She had no history of similar swelling on the contralateral side.

Her general physical examination was unremarkable. Her neck examination showed a ~3 cm × 2 cm sized firm, nontender swelling with well-defined margin and lobulated surface in the right submandibular region. It was freely mobile with normal skin over it. Her left submandibular region was normal with no palpable lymphadenopathy. Her ear examination showed intact bilateral tympanic membrane.

Her laboratory investigations were within normal limits and as follows: hemoglobin – 12.5 g/dl; leukocyte count – 6620/cumm; RBS – 98 mg/dl; viral markers were nonreactive. Chest radiograph was obtained and showed no features of tuberculosis. Ultrasound of the neck reported bulky submandibular gland on the right side with heterogeneous echotexture likely submandibular gland sialadenitis. Further to

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characterize the lesion, fine-needle aspiration cytology (FNAC) of the right submandibular swelling was done. It was reported as acute suppurative lesion featuring polymorphs, plasma cells, macrophages and lymphocytes in a background of necrosis. Occasional clusters of ductal epithelial cells were noted, and Ziehl–Neelsen staining for acid-fast bacilli was negative. Amoxicillin–clavulanic acid combination was started and continued for 3 weeks, but not responded.

In view of long-standing symptoms and suppurative lesion on FNAC, the right submandibular gland was excised. Intraoperatively, importantly, facial vein was divided between ligatures and hypoglossal nerve preserved. The entire right submandibular gland was removed and sent for histopathology. In her postoperative course, she had right marginal mandibular paresis, which was managed with injection dexamethasone and facial exercises. Her neck drain was removed on the 2nd postoperative period. Her paresis gradually improved and she was discharged on the 4th POD on oral antibiotics for 1 week.

Gross examination of the specimen showed submandibular gland measuring ~3.5 cm × 3.5 cm × 1.8 cm and weighing 10.4 g with unremarkable cut surface [Figure 1a]. Her histopathology showed seromucinous glands with ducts and lymphoplasmacytic infiltrates. Dilated duct was seen with vegetative material and actinomycotic colonies (Gram-positive) [Figure 1b-d]. Six reactive lymph nodes were also identified. No granuloma or atypia or malignancy was eminent in the section examined.

At 6-month follow-up, her paresis was completely resolved with no further sign of actinomycotic infection.

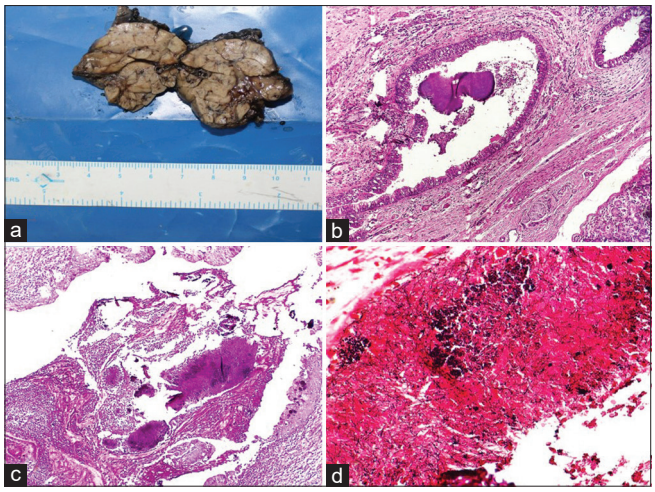

Figure 1: (a) Cut section of the submandibular gland appears normal grossly; (b and c) microphotograph showing *Actinomyces* colonies and surrounding inflammation within submandibular duct (H & E, ×100); (d) microphotograph showing Gram-positive bacilli (Gram stain, ×400)

DISCUSSION

AM is a commensal of oral cavity, but may present as chronic, suppurative infection in cervicofacial region. AM may involve the mandible, cheeks, chin, submaxillary ramus and parotid gland, but submandibular gland has been involved in very few instances.[3,4] Our case seems to be third in the reviewed available literature on submandibular gland actinomycosis (SGA).

AM is more prevalent in young male (3:1),[3,4] but we encountered SGA in a young girl, which is rare to find. The predisposing factors of AM are poor oral hygiene, odontogenic infection, dental extractions and surgical intervention. Poor oral hygiene and dehydrated oral cavity may lead to concentrated saliva and also mucosal breakdown, which may progress to AM infection.[1,5] We did not find any such predisposing factor in her and considered it spontaneous.

The clinical diagnosis is often a challenge due to its nonspecific symptoms and signs. The disease is often characterized by an abscess formation surrounded by a granulomatous inflammatory reaction. It may imitate infectious as well as noninfectious diseases such as granulomatous disease or malignancy.[5,6] In our case, it was primarily diagnosed as long-standing nonpainful swelling and likely suppurative infection on needle cytology.

Hematological and biochemical parameters are usually noncontributory, similar to our case. The patient may have low hemoglobin, mild leukocytosis, elevated C-reactive protein and erythrocyte sedimentation rate. Cultures from sinuses or aspiration cytology may help, but one needs to be vigilant about its possibility. Gram staining may show beaded, branched, Gram-positive filamentous rods and cultures usually require 2–3 weeks to grow *Actinomyces*.[6] In the current era, polymerase chain reaction and nucleic acid probes are quick methods to diagnose, but not readily available. We got only findings of acute bacterial infection on FNAC, but no definitive evidence of SGA.

Radiological investigations such as computed tomography and magnetic resonance imaging also play a supportive role in showing osteomyelitis with surrounding inflammatory changes, but still have no definite signs to diagnose AM.[1,5] We got only US done which also reported sialadenitis.

Medical management usually fails for SGA. Most *Actinomyces* species are vulnerable to beta-lactams, so long course of oral amoxicillin is usually preferred. Acceptable alternatives comprise macrolides, tetracycline and cephalosporins,

which has a superior bone permeation, but still needs long course.[6,7] The patient in the present instance was put on amoxicillin–clavulanic acid combination, but not responded.

The definitive way to diagnose SGA is histological examination, but it is suspected clinically whenever there is a presence of painless progressive indurated mass in retromandibular area, persistent abscesses with multiple draining sinus, or recurrent infection of submandibular gland. Hence, surgical management is the optimal choice for SGA, which ranges from drainage of abscess, marsupialization of sinus tracts, debridement of necrotic bone tissue to complete excision of disease gland.[3,6] In this case, in view of long-standing swelling, nonresponder to medical management and suppurative infection on FNAC leads us to perform complete submandibular gland excision.

The prevention of cervicofacial AM should be the goal. Maintenance of proper oral hygiene with regular removal of plaques limits the *Actinomyces* to form dense colonization and subsequent infection.[2]

Hence, recurrent infection of submandibular gland, not responding to antibiotics, SGA should be kept as differential diagnosis and might need surgical treatment for symptomatic relief and definitive diagnosis.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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