A Non-Sinus Forming Mandibular Actinomycotic Osteomyelitis with a Submandibular Gland Swelling - A Case Mimicking a Periapical Infection

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INTRODUCTION

Involvement of the submandibular gland in actinomycotic osteomyelitis in the absence of sinus is rare. Cervicofacial form is characterized by contiguous spread, suppurative osteomyelitic & granulomatous inflammation of the mandible and formation of multiple abscesses and hallmark discharging sinuses draining sero-sanguinous fluid containing sulphur granules.

As the imaging finding of this entity is rarely described, in this case report, a rare case of mandibular actinomycotic osteomyelitis, with a submandibular swelling, but without draining sinus is reported. The imaging findings of cone beam computed tomography (CBCT) & ultrasonography (US) of the case are discussed with a review.

In subacute stage, mandibular actinomycosis may show no hallmark sinuses. Sclerotic margins around the lesion may be found on radiograph. Imaging is useful in ruling out clinical diagnostic challenge when it includes involvement of mandible & submandibular region.

Cervicofacial actinomycosis commonly occurs as perimandibular infection, rarely as osteomyelitis of the mandible.[1] It may present as two distinct morphological patterns; first, "lumpy jaw," and second, simulating an acute pyogenic infection affecting the submandibular area, discharging sinus being a hallmark finding.[1] Other variant reported include chronic osteitis, osteolytic lesion, hard nodule on the tongue, lockjaw, periapical, or paradental abscess. Diagnosis of cases presenting in multiple areas in the absence of multiple discharging sinuses is a challenge.[2] Usefulness the imaging findings are rarely enumerated.

This report is of a twin presentation of a non - sinus forming actinomycotic mandibular osteomyelitis and a submandibular swelling, along with a review of various diagnostic imaging features.

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PRESENTATION OF CASE

In the third decade, a male patient presented with six - month - old painful swelling below the angle of the lower jaw. History revealed, fracture of the right mandible in an accident a year back, treated with inter-maxillary fixation. A few months later, the patient returned with a painful right mandibular tooth mimicking a periapical infection. The attempted endodontic treatment failed to relieve the symptoms. The tooth was extracted due to a persistent infection, in spite of endodontic treatment. Three months later, a soft and painless swelling appeared in the right submandibular region, which gradually enlarged to become firm and painful. There was no history of draining sinus, fever, cough, weight loss. Medical, family, and psychosocial history was unremarkable. Despite the repeated medication of antibiotics and analgesics, partial remissions and exacerbations of the swelling continued.

Extraoral examination revealed a diffuse mild swelling over the right angle of the mandible, and an oval, 2.5 cm x 3.5 cm, swelling in the ipsilateral submandibular region 1 cm anterior to the sternocleidomastoid muscle, 1 cm below the lower border of the mandible, with well - defined borders, smooth surface, & bluish - purple coloured overlying skin. (Figure1). Swelling in the submandibular gland region was firm, tender, non – fluctuant. The skin over the swelling was indurated, but no sinus/pus discharge was present. Right submandibular tender lymphadenopathy was present.

On intraoral examination, there was no source of dental sepsis in the molar region. On palpation, the vestibule showed mild obliteration of the vestibule and tenderness. The panoramic image revealed ill-defined radiolucency, 3.05 x 1.68 cm in size, in the mandible extending from the second molar till the anterior ramus, resorbing the external oblique ridge. The lesion showed a moth-eaten appearance, with irregular sclerotic borders. (Fig. 2). CBCT showed an osteolytic lesion, mild expansion of the buccolingual cortical plate predominantly in tooth no. 48 region, effacement, and disruption of the lingual cortex, with irregular areas of dense trabecular pattern (Fig. 3). Radiographic impression was chronic osteomyelitis of the mandible.

Figure1A & B. 1A. Patient Photograph Showing Diffuse Mild Swelling Over the Right Angle of the Mandible & Submandibular Swelling; 1B. Resolution of the Lesion After 6 Months

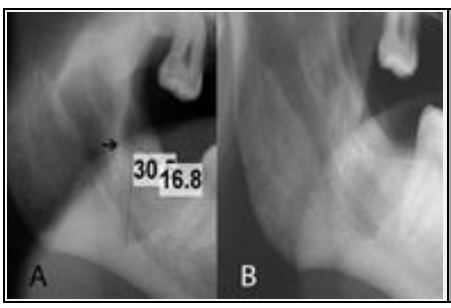

Figure2A & B. 2A. Panoramic Radiograph Showing Motheaten Type of Radiolucency & a Surrounding Sclerotic Border.2B. Post Treatment Image Showing Resolution.

An ultrasound examination suggested an inflammatory lesion in the submandibular gland. It showed a roughly circular hypoechogenic area in the right submandibular region measuring 2.1 x 1.4 cm, with multiple echogenic foci, the largest measuring 2.5 mm, oedematous changes in the overlying skin, subcutaneous tissue. (Fig. 3) Fine needle aspiration cytology (FNAC) from the submandibular swelling confirmed the final diagnosis as actinomycosis. (Fig. 3)

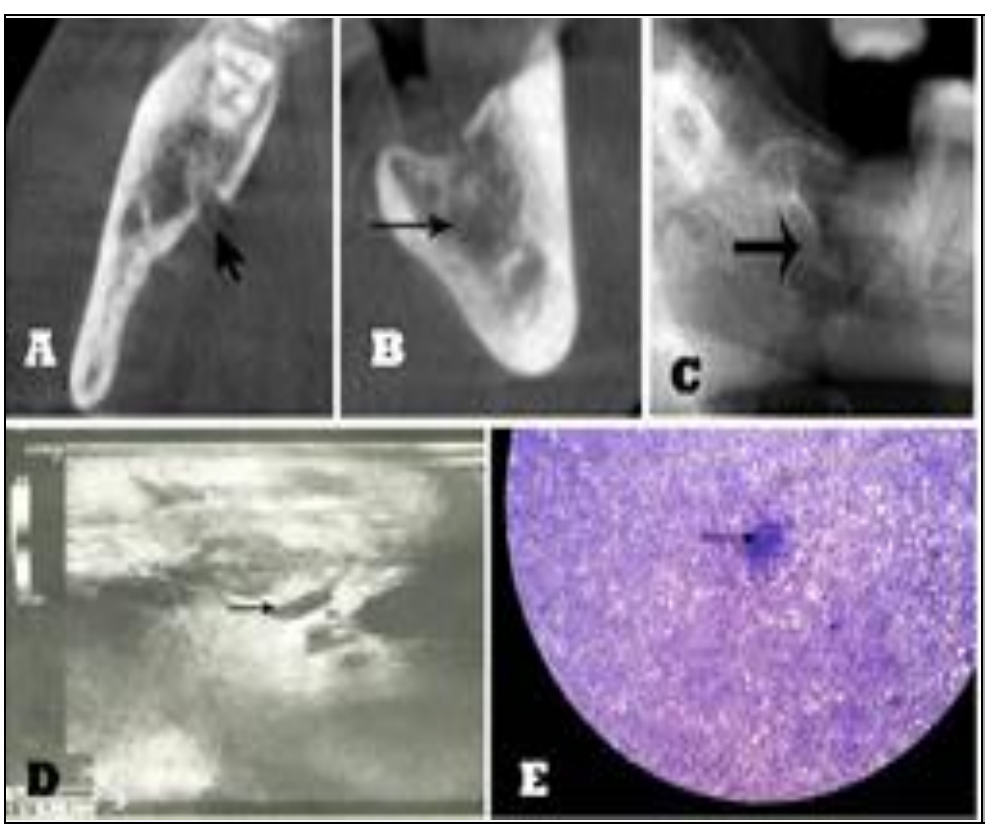

Figure3A, 3B, 3C, 3D & 3E. 3A. Axial Section CBCT Showing Mixed Density Lesion and Lingual Cortical Breach (Arrow). 3B. Coronal Section CBCT Showing Mixed Density Lesion (Arrow) with Defined Sclerosing Periphery. 3C. 2D Tomographic View Showing Disruption of the Cortex. 3D. US Image Showing Circular Hypoechogenic Area in Right Submandibular Gland with Multiple Echogenic Foci (Arrow) & Oedematous Changes Skin and Subcutaneous Tissue. 3E. FNAC Demonstrated Few Colonies of Actinomycosis (Arrow) against the Background of Haemorrhage and Neutrophils.

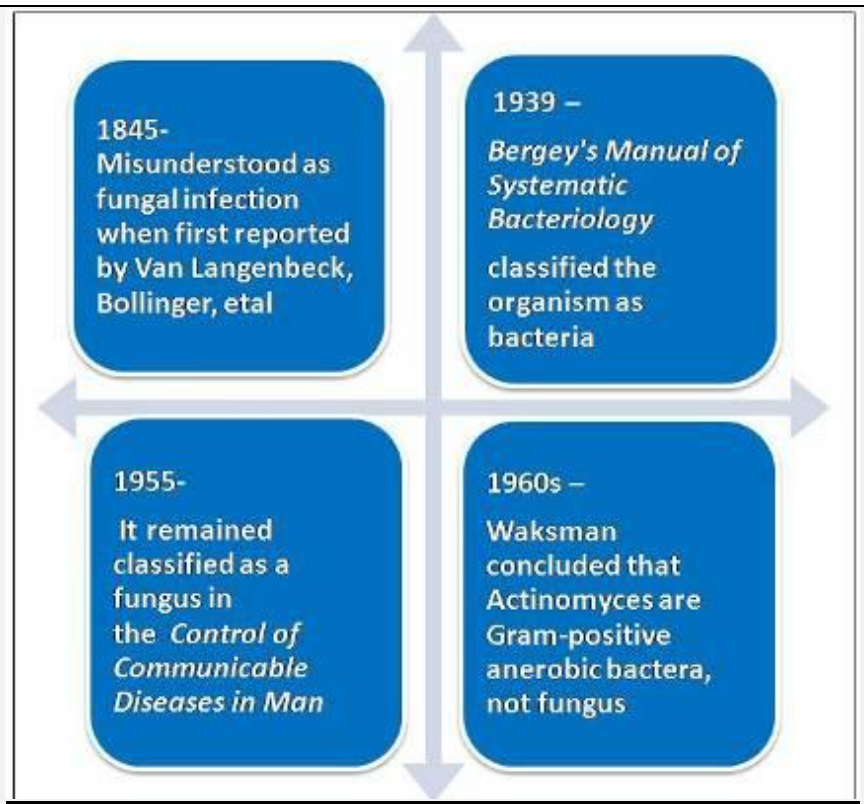

Figure 4. Classification of Actinomyces, Controversies / Timelines

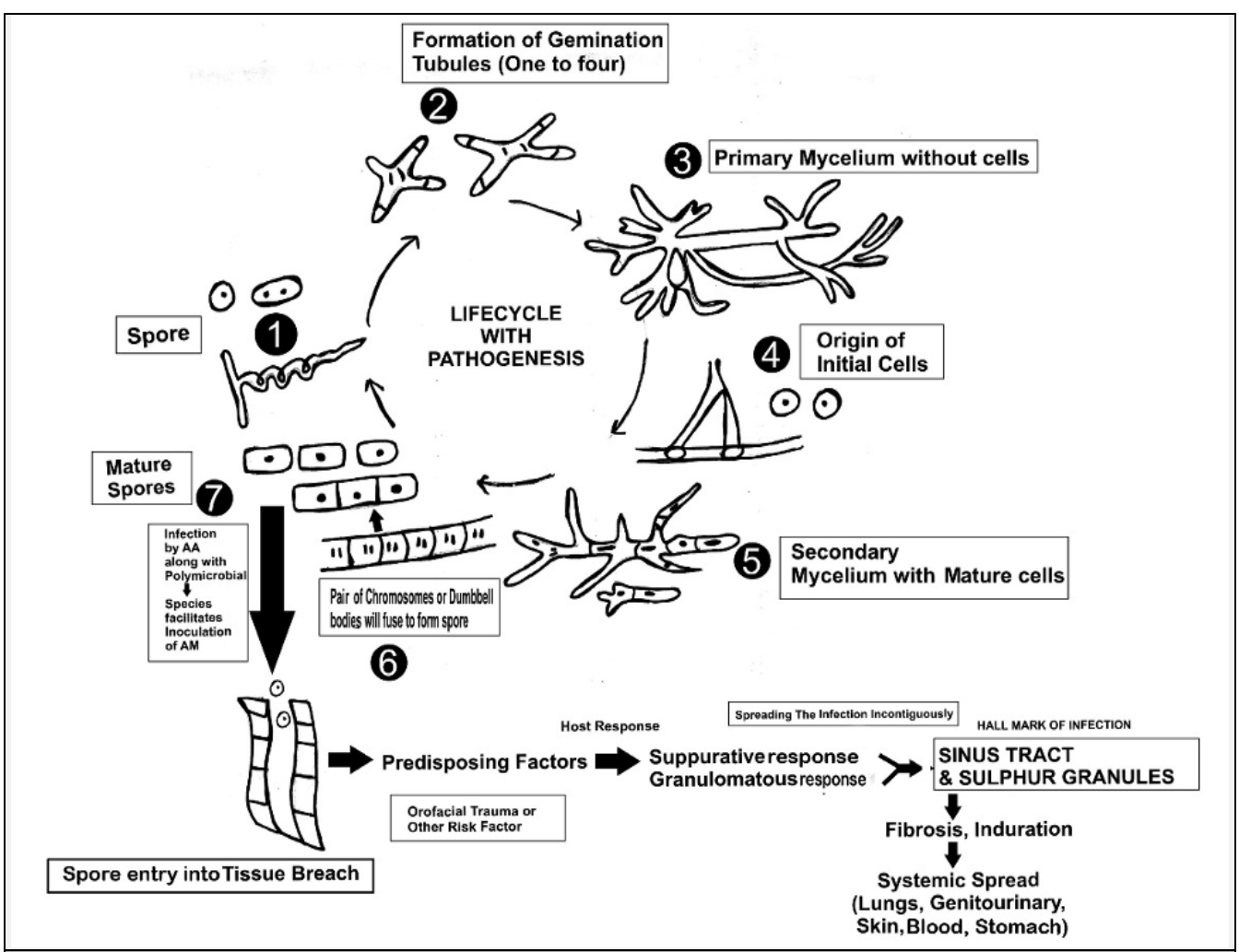

Figure5. Actinomycosis - Lifecycle and Pathogenesis

CLINICAL DIAGNOSIS

Inflammatory swelling of the right submandibular space, secondary to a residual infectious source in the tooth 48 region.

DIFFERENTIAL DIAGNOSIS

Residual abscess, chronic suppurative osteomyelitis, antibioma, and TB lymphadenitis

PATHOLOGICAL DISCUSSION

Actinomyces, earlier classified as a fungal lesion, are now classified as bacteria.[3] (Fig. 4). The spores follow a cycle of haploid & diploid generations by the process of reduction division and conjugation. Spores gain access into the body in the presence of characteristic risk factors.[4] (Fig. 5). Cervical actinomycotic infection without the hallmark draining sinus is rare. Characteristic series of diagnostic manifestations are chronicity, extensive propagation across tissue planes, and firm to hard mass like features often mimicking malignancy, multiple draining sinuses, and frequent remissions & relapses.[5] The most commonly involved site is the angle of the jaw or the submandibular region, followed by cheek, submental space, masticator space, and temporomandibular joint.[6] As in the present case, dual presentation of mandibular osteomyelitis and a submandibular swelling, in the absence of discharging sinus, may be mistaken as dental abscess involving the submandibular space. However, 40 % of cases report a discharging sinus. It is not clear if this finding is absent in patients with good host resistance, low virulence of the organism, or subclinical disease.

Cervicofacial actinomycosis infection of the mandible is often a peri mandibular, granulomatous infection. However, the mandibular osteomyelitis with a submandibular gland swelling is rare, as reported here. Bone infection (periostitis and osteomyelitis) may develop in 11.7 % of cases.[7] Reports show sclerosis within the lesion, but not a sclerotic periphery, around a minimal expansile, moth-eaten type of radiolucency as seen in the present case. It is not clear if this characteristic is pathognomonic of the disease or is a feature in subclinical forms.

Not many studies reported CBCT findings of cervicofacial actinomycosis. In the present case, CBCT was useful in showing the buccolingual expansion, irregular areas of dense trabecular pattern, disruption of the lingual cortex, & status of the mandibular canal. Various imaging findings of actinomycotic mandibular osteomyelitis, described in the literature reported between 1970 to 2017, include lytic and sclerotic lesion, minimal expansion, destruction of cortices on computerised tomography (CT), heterogeneous lesion on magnetic resonance imaging (MRI), homogenous radiotracer uptake on bone scan. The lesion may extend into the adjacent skin, fat tissues, muscle, masticator spaces, and the salivary gland.[1,2,5-16] (Table 1).

Feature	Description
Site	Mandible angle & ramus (CT),[6,8,9,10] (PAN),[1,9,10] (CBCT).[11] ramus (plain film),[12] (CT),[13] body (PAN),[5,14] (CT),[14] (MRI).[14] incisor area (PAN,CT, MRI).[2]
Size	None described.
Shape	None described
Periphery	Irregular (CT).[2], Ill - defined (PAN)[5,11,9] (CBCT)[11], (CT)[14]
Internal structure	Cortical thickening of vertical ramus (plain film),[12] thickening with lucencies (PAN),[15] osteolytic areas with sequestra, osteolytic & osteosclerotic areas (PAN),[5] osteolytic lesion crossing midline with floating teeth (PAN),[14] destruction of labial bone (CT),[2] mixed density lesion (CT),[9] focal sclerotic change within the lesion (CT),[6] osteolytic lesion (CT),[16] sequestrum within a radiolucent capsule(CT),[10] radiolucency (CBCT),[11] heterogeneous high signal intensity in mandibular bone lesion (MRI),[16] heterogeneous low signal intensity lesion (MRI),[14] homogeneous intense uptake pattern on gallium scintigraphy (Scintigraphy).[16]
Effect on surrounding structure	Expansion of vestibular & lingual cortex (CT),[9] extensive demineralization of buccal & lingual cortical plate (CT),[14] perforation of buccal and lingual cortex, widening of PDL space, indistinct mandibular canal (CBCT),[11] cortical bone destruction (US).[8]
Extension into surrounding structures	Right sub-masseteric and pterygomandibular space, chest (CT),[2] submandibular space, adjacent fat plane and fascial thickening (CT),[6] in front of the right sternocleidomastoid muscle (CT),[13] masticator space, the skin, masseter muscle, and parotid gland, (CT),[16] muscles of mastication (MRI),[14] buccal space.[17]
Submandibular gland involvement	Peripheral ring - enhancing lesion with central suppurative necrosis in the right submandibular space, relatively homogeneous enhancement that has small necrotic areas or cystic portions (CT).[6] Elliptical mass with a focal skin defect / sinus, strongly enhanced mass, with several small non-enhanced areas, soft - tissue infiltration to the adjacent fat plane and fascial thickening (CT).[6] Expansive large mass (approximately 4.5cm in size), located in front of the right sternocleidomastoid muscle (CT).[13]

Table 1. Features of Mandibular Actinomycotic Infection Described on Different Types Imaging

Ultrasound examination of cervical swelling is useful to rule out other causes like submandibular gland involvement, tubercular lymphadenitis, and other tumours. Reports on US findings are rare. In the present case, the US showed hypoechoic areas with multiple echogenic areas. On US examination, an actinomycotic lesion showed a mass with relatively homogeneous enhancement that has small necrotic areas or cystic portions. The abundant granulation tissue, fibrosis & necrosis may result in such enhancement on ultrasound imaging.[6] In the subacute stage, mandibular actinomycosis may present as a slowly progressing osteomyelitis with no draining sinuses that may easily mimic a periapical infection. Sclerotic margins around the lesion may be found on the radiograph. Involvement of mandible & submandibular region challenges clinical diagnostic skills, & CBCT, US are useful in such cases. Prolonged treatment is required to control the disease, as in the present case.

DISCUSSION OF MANAGEMENT

Pharmacotherapy included-Amoxycillin and Clavulanic acid 625mg TID and Diclofenac Sodium BID & Serratiopeptidase. six months of follow up gave complete remission of clinical signs and symptoms. (Fig. 1B), The post-therapy panoramic radiograph showed partial resolution of the lesion. (Fig. 2B.) (Time line of the case is given in the table). The patient reported no adverse event. He reported improved quality of life in the physical, psychological, and social domains.

Time - line			
SYMPTOM - Swelling on right submandibular area and pain in 48 region. History of trauma 1 year back, intermaxillary fixation, 1 month later pain in 48, root canal treatment of 48, followed by extraction of 48. Three months later submandibular swelling appeared. Failed repeated attempts to treat by local doctors.			
Time	Clinical Exam	Radiography / Cytology	Therapy
1st visit	Provisional Diagnosis: Residual dental infection involving submandibular space aspiration from the lesion	Panoramic - Rarefying lesion near the extraction socket of tooth 48. Requested for ultrasound & CBCT Sample sent for culture & sensitivity Request for cytopathology	Antibiotics Anti - inflammatory Serratiopeptidase
72 hrs		Culture report - ve	
1week	Pain reduced, swelling persistent	Cytology diagnosis: Actinomycosis Ultrasonography: Inflammatory swelling of submandibular gland	No adverse effects reported. Continued the medication.
2 weeks	Significant reduction in size of the swelling.		Patient followed up telephonically. Every fortnight. No recurrence
6 months	Complete remission of the swellings	Follow-up panoramic radiograph showed partial resolution of the lesion.	Patient reported a generalized physical and psychological well-being.

Table 2. Timeline of Clinical Workup

FINAL DIAGNOSIS

A non-sinus forming mandibular subacute actinomycotic osteomyelitis with submandibular gland involvement.

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