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Case Report

Cervicofacial actinomycosis with chronic parotid sialadenitis-mimicking malignant neoplasm of parotid gland: a rare case

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ABSTRACT

Cervicofacial actinomycosis is an uncommon disease caused by *Actinomycosis israelii*. Chronic 'sialadenitis' of parotid gland is an insidious inflammatory disorder and may form a fibrous mass. We report a case of 55 years old female presenting with mass in right parotid region-on imaging-malignant neoplasm of parotid gland. But on histopathology-actinomycosis and chronic parotid sialadenitis.

Keywords: Actinomycosis, Chronic parotid sialadenitis, Superficial parotidectomy

INTRODUCTION

Actinomycosis israeili is a gram-positive bacterium. They are harmless but become virulent when there is breach in mucosa and presence of co pathogens. Cervicofacial actinomycosis still occurs occasionally and the lesion can mimic many other diseases, including neoplasms of the head and neck causing difficulty in diagnosis.[1,2]

Classically actinomycosis manifests as a slow growing, painless mass with multiple draining sinus tract on the skin or oral mucosa and occasionally presence of thick, yellow exudate with characteristic sulphur granules.

Chronic sialadenitis of the parotid gland is an uncommon clinical condition characterized by intermittent, often painful parotid swelling and/or discharge of pus and inflammatory changes in the affected gland.[3,4] Chronic sialadenitis of the parotid gland is an insidious inflammatory disorder which tends to progress and maybe lead to the formation of a fibrous mass, chronic sialadenitis is a major disorder that can cause the salivary hypofunction.

CASE REPORT

A 55-year-old female was admitted in the general surgery department with complains of a progressive painful swelling in the right parotid region with trismus and restricted mouth opening of 4 weeks duration. Unable to eat and loss of weight. The patient did not give any history of trauma or tooth extraction. No history of fever. She had consulted dental surgeon and received medicine with no relief. She belongs to poor socioeconomic class. History of chronic tobacco chewer.

On local examination: Swelling in right parotid region of face 4×4 cm, diffuse, firm to hard in consistency with irregular margins, fixity to overlying skin, no discoloration (Figure 1 and 2) local temperature normal, softness on top of swelling, mobility restricted. Mouth opening restricted allowing only 1 finger. On intraoral examination few teeth missing. No caries. Skin excoriation after FNAC. No sinuses in the wound and no granules.

Routine hematological examination was normal.

Figure 1: Clinical image on presentation.

Figure 2: Clinical image lateral view.

Ultrasonography revealed hypoechoic lesion in right parotid with cystic area and solid component within. Enlarged lymph nodes in right submandibular region neoplastic origin.

CECT face and neck-a large lobulated heterogeneously enhancing mass lesion of size 5.4×3.3 cm in right parotid space involving the right parotid gland superficial and deep lobe, masseter muscle, buccinator muscle and portion of temporalis, multiple discrete cervical lymph nodes level I, II, III. These features are likely suggestive of neoplastic pathology likely malignancy of right parotid gland with metastatic type of lymphadenopathy.

FNAC from swelling suggestive of organizing suppurative lesion.

Smear for staining and culture sensitivity (from discharge). Gram stain-plenty of pus cells seen. No organism seen. ZN stain-no acid-fast bacilli seen. Culture sensitivity report sterile. X-ray chest was normal.

The patient undergone surgery-superficial parotidectomy with mass excision (Figure 3-5). Ulcerated skin over the mass excised.

The mass was adherent to the parotid gland. Excised in toto. The parotid gland was bulky and not looking like malignant so superficial lobe of the parotid gland removed; muscles not excised. 2-3 Lymph nodes removed which was enlarged. Post operative period uneventful. Discharged on tenth day. Post operative patient has facial paralysis due to neuropraxia. Slowly recovered.

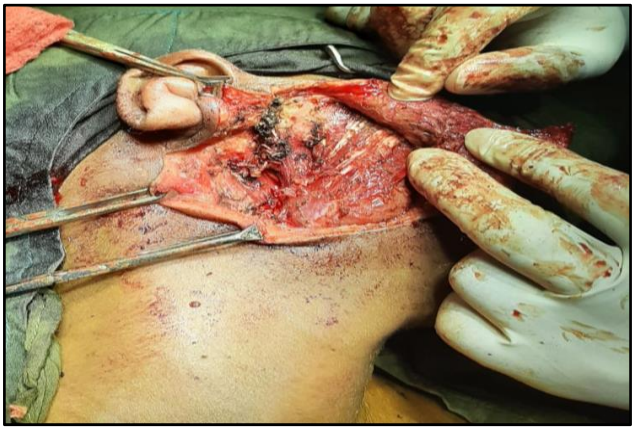

Figure 3: Post operative clearance of disease.

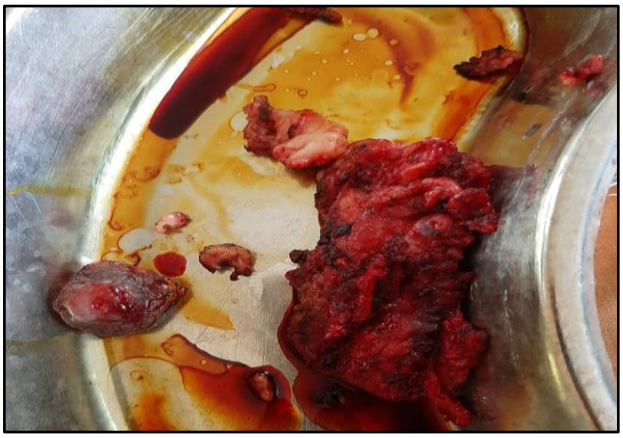

Figure 4: Excised specimen.

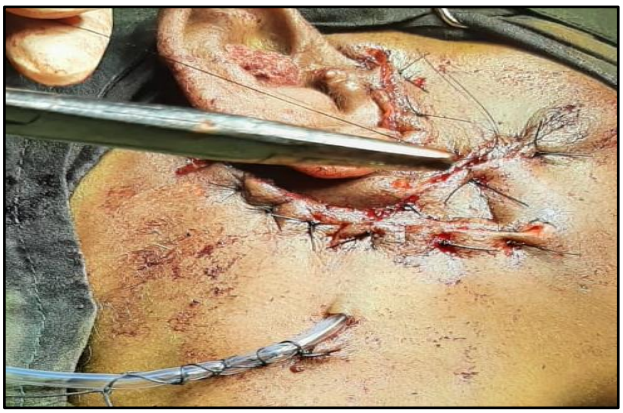

Figure 5: Post op image.

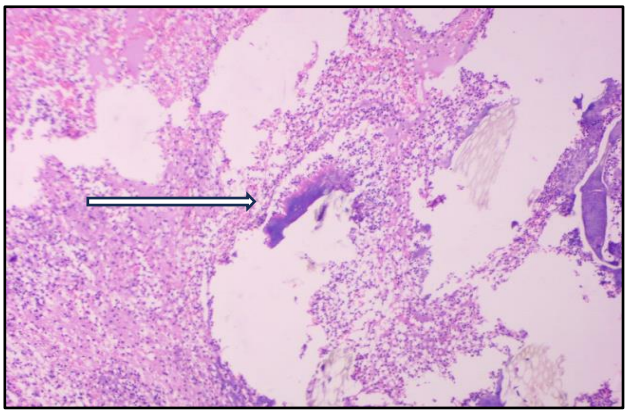

Figure 6: Actinomycosis under 10×HE.

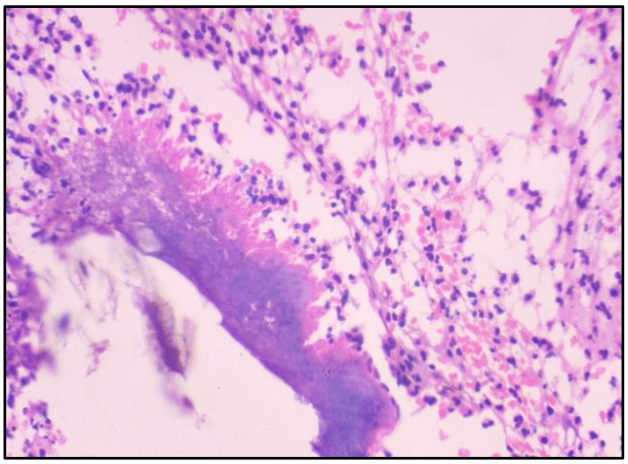

Figure 7: Actinomycosis under 40×HE.

Histopathology

Parotid: ducts and acini with moderate to severe infiltration of lymphocytes. Areas of fibrosis, hyperplastic lymph node.

Skin covered mass pseudo cystic structure, deep to this fragment of vegetative foreign body surrounded by acute inflammatory reaction, areas of fibrosis. Dark basophilic structures having eosinophilic sun ray appearance surrounded by strong neutrophilic aggregates-actinomycosis and parotid chronic sialadenitis.

DISCUSSION

Actinomycosis is a chronic, suppurative, granulomatous, bacterial disease primarily caused by *Actinomycosis israelii*, a gram positive, non-acid fast, anaerobic or microaerophilic filamentous branched bacterium, difficult to culture.[5-7] The bacteria are normal inhabitant of oral cavity and found in caries teeth, tonsillar crepts, gingival crevices.[6] They are harmless but become virulent when there is breach in the mucosa and presence of co pathogens. Cervicofacial actinomycosis still occurs occasionally and the lesion can mimic many other diseases, including neoplasms of the head and neck causing difficulty in diagnosis.[7]

Actinomycotic infection is usually caused by tooth extraction, odontogenic infection or trauma.[8] The etiology was unclear in our patient because she had no identifiable portal of entry. However, actinomycosis in parotid region involving masticator muscles, buccal and temporal and vegetative foreign body in the mass with chronic parotid sialadenitis has not been reported previously.

Chronic sialadenitis of the parotid gland usually diagnosed as recurrent parotitis, non-obstructive parotitis, obstructive parotitis, or benign lymphosialadenopathy.[9]

In actinomycosis, imaging techniques like USG and CECT show inconsistent findings and are non-contributing to positive diagnosis. Histopathology is the gold standard for diagnosis.[10] The disease is difficult to diagnose as it commonly mimics like tuberculosis, chronic granulomatous lesion and malignancy.[4] Hence it is considered as the most misdiagnosed disease, and is listed as a rare disease by the office of rare disease (ORD) of the national institute of health (NIH).

Management of cervicofacial actinomycosis includes antibiotics and surgery.

In chronic sialadenitis of the parotid gland initial management consists of antibiotics, analgesics and major surgical procedures (superficial parotidectomy) in the reported case.[11,12]

In our case, clinically and on imaging it was not possible to differentiate two pathologies separately. On imaging-USG and CECT diagnosed as malignant neoplasm of parotid with secondaries in lymph nodes.

On histopathological examination we found, the mass covered with skin showed-actinomycosis and fragments of vegetative foreign body surrounded by acute inflammatory reaction. Parotid gland shows infiltration of lymphocytes. Follicular hyperplastic lymph node identified, which were inconsistent with the imaging interpretation of neoplasm. Thus, a very rare case of actinomycosis and vegetative foreign body in the parotid region along with chronic sialadenitis of parotid gland.

CONCLUSION

Mass in parotid region can't be diagnosed clinically and on imaging. Imaging may confuse as in this case. Histopathology is the gold standard process after resection. We think, the choice of treatment in such cases is radical resection of the lesion and superficial parotidectomy, that to be decided on operation table.

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