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Case Report

Pleomorphic adenoma involving soft tissue overlying the anterior border of ramus of the mandible: A rare ectopic presentation

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ABSTRACT

Ectopic pleomorphic adenoma is a rare presentation in sites other than major and minor salivary glands. In literature, cases of pleomorphic adenoma in the soft tissue of the neck, skin of the external auditory canal, subcutaneous layer of nose, and lymph node of mediastinum have been reported. We report a case of ectopic pleomorphic adenoma localized in the soft tissue over the anterior border of the ramus of left side of the mandible.

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1. Introduction

Pleomorphic adenoma is the most common benign tumor of salivary glands. It comprises of about 3–10% of all the tumors of head and neck region and 50% of the major and minor salivary gland neoplasms.¹ Intraorally, hard palate² is the most common presenting site (42.8–68.8%) followed by upper lip³ (15–20%), and cheek⁴ (5.5%). Other sites are throat (2.5%) and retromolar region (0.7%). It may also arise in floor of mouth and alveolar mucosa.³

Other ectopic presentations of pleomorphic adenoma are along the stenson's duct, accessory parotid tissue, and parapharyngeal space.⁵ It may also develop in lymph nodes of the neck due to salivary gland inclusions.¹ In this report, we present a case of rare ectopic presentation of pleomorphic adenoma.

2. Case report

A 37-year-old male patient reported to our unit with the chief complaint of a swelling in the mouth on the left side of lower

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jaw for last four months (Fig. 1). He was apparently well four months back and then he noticed an asymptomatic swelling near the retro-molar region of the left side of the mandible. Swelling progressed gradually to the present size over the period and led to mild discomfort in swallowing and mastication. His systemic history was not contributory.

On clinical examination, extraorally no abnormality was seen. Regional lymph nodes were not enlarged. Intraorally a localized oval-shaped swelling of approximately 3 cm × 3 cm size with well-defined border was present along the junction of lateral border of hard palate and ascending border of the ramus of the mandible on left side. Swelling was firm in consistency and non-tender on palpation. Mucosa over the swelling was normal in color and texture. Mild ulceration of overlying mucosa due to trauma from adjacent tooth was present at the anterior aspect of swelling. Provisional diagnosis of squamous cell carcinoma was made. Lipoma, rhabdomyoma, pleomorphic adenoma, and mucoepidermoid carcinoma were kept in the differential diagnosis.

Routine hematological investigations were within normal limit. Viral markers were negative. Computed tomographic scan revealed an ill-defined, heterogeneously enhancing, soft tissue density, mass lesion of approximate size 22 mm × 20 mm × 18 mm in antero posterior, transverse and cranio-caudal axis respectively in left buccal space involving retro-molar trigone. It was infiltrating left lateral pterygoid muscle medially and deep head of temporalis laterally (Fig. 2). There was no evidence of associated bony erosions. There was evidence of few subcentimetre size lymph nodes in left submandibular area. Incisional biopsy revealed it as pleomorphic adenoma.

We planned for complete excision of the lesion with safe margins. Under local anesthesia, incision was placed over the

Fig. 1 – Preoperative photograph of the lesion involving soft tissue overlying anterior border of ramus of the left side of the mandible.

Fig. 2 – Computed tomographic scan (axial view) revealed an ill-defined, heterogeneously enhancing soft tissue mass lesion of approximately size 22 mm × 20 mm × 18 mm in AP, transverse and cranio-caudal axis respectively in left buccal space involving retromolar trigone.

mucosa, and layerwise dissection was carried out. The tumor was encapsulated between the buccinator and medial pterygoid muscles. After removal of tumor, layerwise closure with 3-0 vicryl suture was done. On histopathological examination, the section revealed proliferation of polygonal and spindle shaped myoepithelial cells in a variable background stroma containing mucoid and myxoid areas at places. Epithelial elements were arranged in duct-like structures, sheets, clumps, and/or interlacing strands and consist of polygonal, spindle, or stellate-shaped cells. The tumor is surrounded by a fibrous pseudocapsule of varying thickness and it extends through normal glandular parenchyma in the form of finger like pseudopodia. This confirmed its diagnosis as pleomorphic adenoma. After one week, wound healing was found satisfactory and sutures were removed. The patient is under regular follow-up with no signs of recurrence.

3. Discussion

Pleomorphic adenomas are the most common benign salivary gland neoplasm. It accounts for 60–70% of all parotid neoplasias, 40–60% of all submandibular neoplasias and 40–70% of minor salivary gland neoplasias.¹ Majority of minor salivary glands are present in hard palate, buccal mucosa, and upper and lower lip. Other sites are floor of the mouth, inferior surface of the tongue, palatoglossal folds, and tonsillar region. Mostly it occurs between 4th and 6th decade of age, with male-female ratio varying from 1:1.4 to 1:1.7.² Clinical presentation includes asymptomatic, slow growing firm mass. Small tumors typically form smooth, mobile, firm lumps but larger tumors tend to become bossellated and may attenuate the overlying skin or mucosa.⁶

Heterotopic salivary gland tissue is an architecturally normal arrangement of salivary tissue found in locations external to major and minor salivary glands.⁷ Pleomorphic adenoma ectopically presents in the soft tissue of the neck, lymph nodes, tongue, mandible, hypophysis, mastoid bone, thyroid, parathyroid, subcutaneous layer of the nose, and skin of the external auditory canal.⁸ Willis proposed three main hypotheses to explain this kind of heterotopias. These were: an abnormal persistence and development of vestigial structures; dislocation of portion of a deficient rudiment during mass movement and development; and abnormal differentiation of the local tissues (heteroplasia).⁹ This may happen due to metaplasia, neoplastic degeneration of ectopic salivary gland tissue or due to the implantation after surgical excision of the salivary gland tumor.¹⁰

Dardick et al.¹¹ stated that neoplastically altered epithelial cells with the multidirectional differentiation property may be responsible for the histogenesis of the pleomorphic adenoma. Pleomorphic adenoma may also develop due to cytogenetic abnormalities, mainly involving the chromosome region 12q 13–15. The putative pleomorphic adenoma gene (PLAG1) has been mapped to chromosome 8q12.⁶

Surgical excision is the choice of treatment.^{1,6} For intraoral lesions, conservative management is preferred by doing extracapsular excision.⁶ The tumor has to be excised in its entirety with an adequate margin to avoid recurrences. It is so because the pleomorphic adenomas are enclosed by a layer of fibrous tissue usually termed as capsule. This capsule might be very thin and the tumor buds may extend from it.^{12,13} The exact cause of recurrence in pleomorphic adenoma is controversial. Intraoperative tumor spillage is believed to be the main cause of local recurrence but several studies have suggested incomplete initial resection to be the principle cause.¹⁴

Pleomorphic adenoma in 5% case can undergo malignant transformation into carcinoma ex pleomorphic adenoma and metastasizing benign mixed tumor. Radiotherapy is not indicated due to the radioresistant behavior of the tumor. Prognosis is excellent (95%) after complete excision.¹⁵

4. Conclusion

Although pleomorphic adenoma in the soft tissue overlying anterior border of the ramus of mandible is a rare presentation, irrespective of site involvement, the management protocol remains same.

Conflicts of interest

The authors have none to declare.

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