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Case Report

Ear, Nose and Throat Section

Pleomorphic Adenoma of Submandibular Gland: A Rare Case Report

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ABSTRACT

A benign mixed tumour, or Pleomorphic Adenoma (PA), is the most prevalent form of salivary gland neoplasm. Submandibular gland involvement is rather rare, accounting for 8-10% of all salivary gland tumours, but the parotid gland is the site of most occurrences. The present case report describes a rare case of PA that originated from the right submandibular gland. Here, the authors present a case of a 38-year-old female who presented with the main complaint of right side cervical swelling (located inferior to angle of mandible) since two years, insidious in onset and gradually progressive in nature. On examination the swelling was firm, well defined, mobile, measuring approximately 2×2 cm. Rest of the Ear, Nose and Throat (ENT) examination was normal. Ultrasonography (USG) of local region was done which demonstrated a well-defined hypoechoic lesion in the right submandibular gland measuring 18×12 mm showing posterior acoustic enhancement and mild internal vascularity on colour Doppler. PA needs consideration. Fine Needle Aspiration Cytology (FNAC) and Computed Tomography (CT) scan were done further to confirm the diagnosis. Histopathological examination of the excised specimen confirmed the lesion as a PA. The postoperative period was uneventful, and no signs of recurrence were noted during a six-month follow-up. The current case underscores the importance of considering PA in the differential diagnosis of persistent neck swellings and illustrates the diagnostic complexity posed by submandibular gland tumours. It highlights the critical role of histopathological confirmation in establishing a definitive diagnosis and emphasises the need for early surgical intervention to ensure optimal outcomes and minimise the risk of recurrence.

Keywords: Cervical swelling, Contrast-enhanced computed tomography, Fine needle aspiration cytology, Histopathological investigation, Salivary glands

CASE REPORT

A 38-year-old female presented to the Outpatient Department (OPD) with a complaint of right-sided cervical swelling located inferior to the angle of the mandible, persisting for the past two years [Table/Fig-1]. The swelling was insidious in onset and gradually progressive. Associated symptoms included fever without chills (patient had low grade fever 100°F during the time of presentation and hospital stay) and rigours, dryness of the mouth, and intermittent right ear pain for the past 15 days. These are the additional symptoms with which patient presented to our OPD. Here, dryness of mouth (which was not so significant, absence of systemic features) along with other symptoms helped us to rule out Sjögren's syndrome.

[Table/Fig-1]: Patient with right-sided swelling.

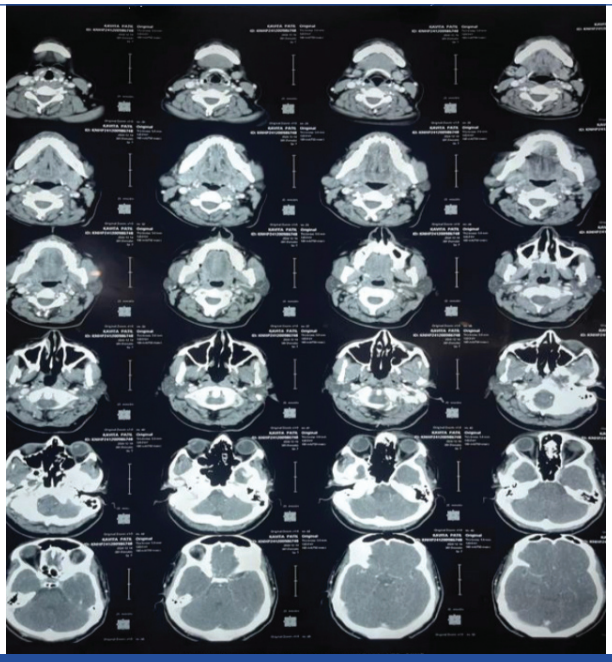
The patient is a known case of hypothyroidism and has been receiving tablet Levothyroxine sodium 100 µg once daily for the past one year. There was no history of recent dental procedures. No additional otorhinolaryngological complaints were reported.

On clinical examination, a well-defined swelling, mobile mass measuring approximately 2×2 cm was found on the right side of the neck. The anatomical extent of the swelling was noted as; superiorly, 0.5 cm below the angle of the mandible; inferiorly, up to 5 cm from the midpoint of the clavicle; anteriorly, 4 cm from the mentum, and posteriorly, 3 cm from the right mastoid tip. The overlying skin appeared normal with no signs of erythema or ulceration. On palpation, the findings were consistent with inspection. The swelling was firm, mobile, solitary, and non tender. Differential diagnosis was lymphadenopathy, masseter muscle hypertrophy, sialadenitis, etc.

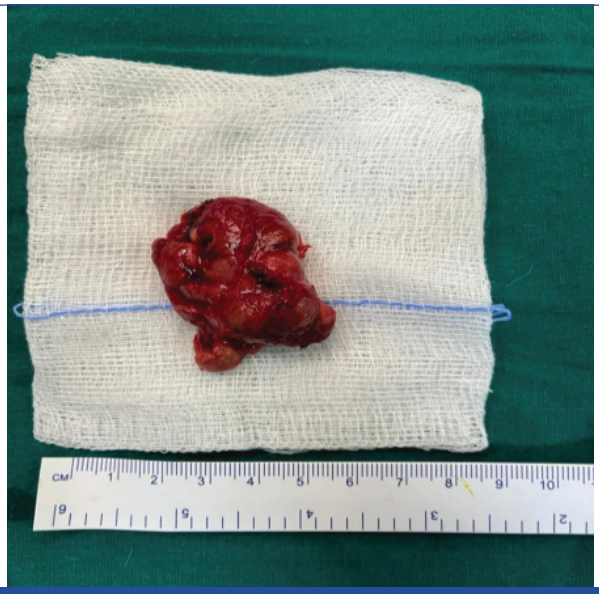
Examination of the throat revealed no abnormalities. Nasal examination demonstrated a Deviated Nasal Septum (DNS) toward the right side. Otoscopic evaluation showed an intact tympanic membrane on the right side, while the left tympanic membrane displayed Grade I pars tensa retraction with a tympanosclerotic patch.

The USG of the neck revealed a well-defined hypoechoic lesion in the right submandibular gland measuring 18 × 12 mm, demonstrating posterior acoustic enhancement and mild internal vascularity on colour Doppler. Both lobes of the submandibular gland showed heterogeneous echotexture. FNAC findings were suggestive of a PA originating from a level IV A lymph node. Though the patient had a clinically positive right submandibular lymph node, involvement of level IV lymph node is rare and might indicate towards metastatising PA. CT of the neck demonstrated an enlarged lymph node in the right parotid region, indicative of an inflammatory or infective aetiology [Table/Fig-2]. No biochemical investigations were done. Risk of malignant transformation is less than 5%. The patient underwent complete surgical excision of the right submandibular gland. Intraoperatively, the mass was well encapsulated, congested on gross examination [Table/Fig-3], histopathological examination {Haematoxylin & Eosin (H&E)} demonstrated an encapsulated tumour with lobulated architecture, containing duct like epithelial

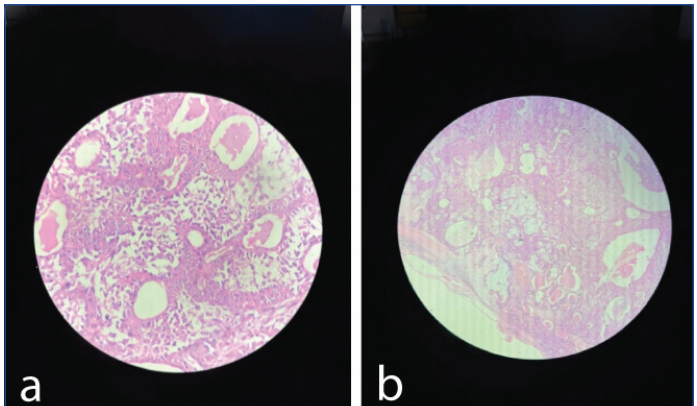
structures, myoepithelial cells, myxoid stromal background with cystic spaces, confirming the diagnosis of PA [Table/Fig-4a,b]. The postoperative period was uneventful with no signs of nerve injury, wound complications. Patient was followed up for six months and there were no signs of recurrence.



[Table/Fig-2]: CT film showing enlarged lymph node in the right infra parotid region.



[Table/Fig-3]: Intraoperative specimen from right submandibular region. Gross specimen measuring 5 × 3.5 × 2 cm, externally congested.



[Table/Fig-4]: a) Histopathological slide showing encapsulated tumour with lobulated architecture, multiple cystic spaces and mixed stromal background (H&E, 40x). b) Histopathological slide showing duct-like structures with myoepithelial cells and myxoid stroma (H&E, 100x).

DISCUSSION

The PA is the most commonly encountered benign neoplasm of the salivary glands, comprising 90% of all benign salivary gland tumours [1,2]. It is characterised by a complex histomorphology, comprising both epithelial and myoepithelial components arranged in varying patterns within a mucopolysaccharide-rich stroma containing mucoid, myxoid, or chondroid elements [2,3]. The differential diagnosis includes other salivary gland lesions such as basal cell adenoma, mucoepidermoid carcinoma, adenocarcinoma, and lymphoma, which must be considered during clinical and histopathological evaluation.

The CT and Magnetic Resonance Imaging (MRI) are considered the gold standard imaging modalities for evaluating lesions of major and minor salivary glands [1]. Adjunctive techniques such as ultrasound-guided or FNAC, though useful, are non-definitive. For lesions of considerable size, an initial diagnostic incisional biopsy may be warranted to establish a histopathological diagnosis. Definitive surgical management is most effectively achieved via a direct submandibular approach, which provides optimal anatomical exposure and access for complete excision [4].

In a case report by Rai S et al., it was noted that the occurrence of PA in the submandibular or sublingual glands is relatively uncommon [5]. Da Silva LP et al., conducted a large retrospective study highlighting the epidemiology of salivary gland tumours [6]. Khanal P documented a case involving a well-encapsulated lesion composed of both epithelial and mesenchymal elements in a patient with PA [3]. Rocha JC et al., described management with partial sialoadenectomy [7], while Susana EN et al., discussed overall management strategies [8]. An analysis by Dombrowski ND et al., evaluating paediatric cases of PA estimated the annual recurrence risk postsurgery to be approximately 0.45% per year of follow-up [9]. Furthermore, Rachida B et al., reported a rare case of PA arising from the submandibular gland in a 10-year-old girl [10].

If left untreated, PA carries a risk of malignant transformation, as noted in the literature. Additionally, the tumour may continue to increase in size, potentially progressing to a giant PA due to its painless and slow-growing nature, often leading to delayed patient presentation. Although cases of malignant transformation in PA are uncommon, the neoplasm is generally associated with an excellent prognosis. In light of these findings, the present case report describes a histopathologically confirmed PA arising from the submandibular gland.

On the histological scale, tumours are classified in terms of their morphological diversity, including glandular regions, myxomatous areas, and other solid regions. It is important to note that this variation in histopathological diagnosis reflects the biphasic composition of epithelia and mesenchyme within a tumour, which explains both the complexity and nomenclature of the tumour [7]. Clinically, these tumours typically present as slow-growing, painless, and firm masses and may remain asymptomatic for prolonged periods [3]. However, despite their benign nature, these tumours carry a recognised risk of recurrence and malignant transformation if inadequately excised.

The submandibular gland, being less accessible than the parotid, presents a diagnostic challenge in early identification of neoplasms [3,8]. In the modern era, imaging modalities such as USG, CT, MRI, and Contrast-Enhanced Computed Tomography (CECT), along with FNAC, are key diagnostic tools in preoperative evaluation. These tools are important to know the lymphovascular invasion of the tumour, nerve involvement and to plan the surgery accordingly [7,8]. Complete surgical excision remains the gold standard for treatment, offering both diagnostic confirmation and definitive management.

The purpose of the present case report was to describe a rare presentation of PA of the submandibular gland. The authors focused on the clinical, radiological, and histopathological features and

discuss the importance of early detection; as well as appropriate surgical intervention to prevent recurrence and achieve optimal outcomes.

CONCLUSION(S)

The PA of the submandibular gland, though rare, is a significant entity in the differential diagnosis of neck masses. Early and accurate diagnosis, aided by imaging techniques such as CT and MRI, is crucial in guiding appropriate management. Surgical excision remains the gold standard for treatment and provides definitive resolution, with a low-risk of recurrence if adequately removed. Although malignant transformation is rare, the benign nature of PA generally results in an excellent prognosis when surgically managed. The present case underscores the importance of early detection and timely intervention in ensuring favourable outcomes for patients with submandibular gland tumours.

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