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(CASE REPORT)

A rare case of a pleomorphic adenoma of the submandibular gland

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Abstract

We report the case of a 27-year-old female with a painless slowly growing mass that had developed over 2 years in the right submandibular region. Physical examination revealed a firm painless submandibular mass. Fine-needle aspiration and computed tomography (CT) scan concluded a pleomorphic adenoma of the submandibular gland. The patient underwent a right total submandibular sialoadenectomy. Histopathological examination confirmed the diagnosis of pleomorphic adenoma arising from the submandibular gland. Transient palsy of the marginal mandibular division of the facial nerve was noted post-operatively, which recovered with supportive management. Pleomorphic adenoma of the submandibular gland represents a diagnostic and therapeutic dilemma in young adults. Preoperative evaluation, including MRI/CT scan and fine-needle aspiration, is recommended in all cases. Total submandibular sialoadenectomy is the standard recommended treatment to minimise recurrences and avoid malignant transformation.

Keywords: Sialoadenectomy; Submandibular Gland; Pleomorphic Adenoma; Benign Salivary Gland Tumours

1. Introduction

Pleomorphic adenomas are the most common benign salivary gland tumours [1]. They can occur in all age groups but are most commonly seen between the third and sixth decades with a female preponderance [1]. They occur most frequently in the parotid glands (>80%), but are also seen in the submandibular gland and hard palate. Pleomorphic adenoma presents as a painless, well-defined mass with gradual progression over the years and can reach enormous proportions [1]. Occasionally but rarely, they may progress to malignancy especially in recurrent cases [1, 2]. Another concern arises in the case of low-grade mucoepidermoid carcinomas which may bear resemblance to pleomorphic adenomas in imaging [1]. This necessitates prompt and complete removal of the tumour, in the form of excision of the tumour with a cuff of surrounding normal tissues to include the pseudopods from the tumour capsule. Recent studies suggest the use of immunohistochemistry, where luminal cells express CK7 and myoepithelial cells express p63, S100, SOX10 and SMA [1].

2. Case presentation

A 27-year-old female presented with painless, progressively increasing swelling in the right side of the neck for 2 years. The patient had no other complaints and was otherwise stable. The swelling has been progressively growing since it was first noticed to a size of approximately 4cmx3cm. The patient's history was otherwise unremarkable. On examination, there was a solitary non-tender swelling was noted in the right submandibular region, with a smooth surface well-defined margins and uniformly firm consistency. The skin over the swelling was normal. There were multiple Level II B non-tender, mobile, lymph nodes palpable on the right side, the largest measuring 1.5x1cm, with a firm consistency. Contrast contrast-enhanced CT scan of the neck revealed a well-defined, heterogeneously enhancing soft tissue lesion with few macrocalcifications in the right submandibular region, along the superior border of the right

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submandibular gland (FIGURE 1). The lesion was in close approximation with the gland and was noted to be compressing the gland inferiorly. The adjacent bone was intact. Few enhancing lymph nodes were noted in the right level IA,

IB II and V region, and the fatty hilum was not well demarcated. Fine needle aspiration and cytology showed features suggestive of ? pleomorphic adenoma.

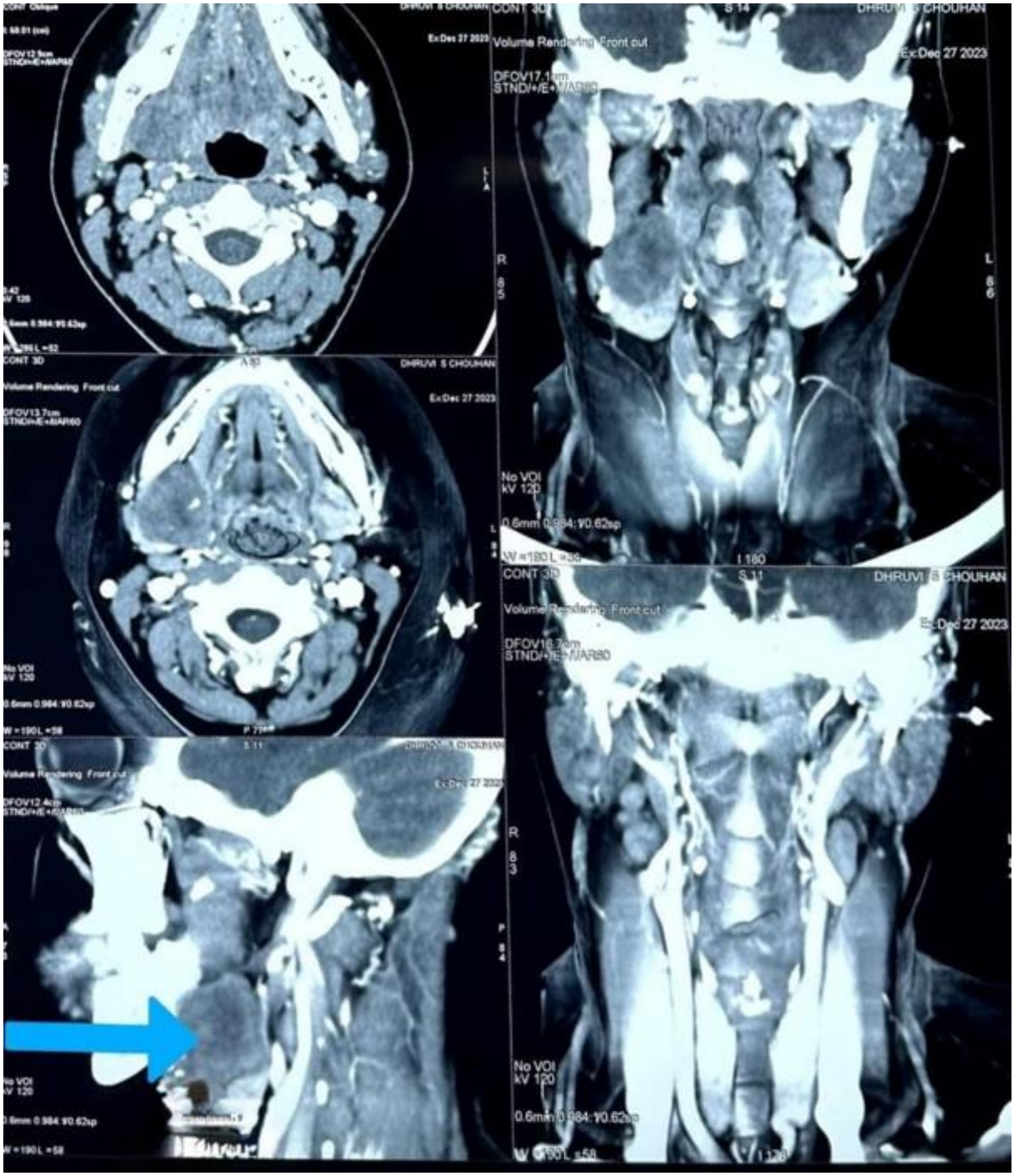

Figure 1 Computed tomography of the submandibular region showing a heterogeneous lesion in the right submandibular gland. The blue arrow points towards the lesion in the sagittal view of the scan.

2.1. Treatment

Based on the clinical, radiological and cytological findings, a diagnosis of pleomorphic adenoma was made and the patient was posted for right submandibular sialoadenectomy. Intra-operatively, the tumour had a variable consistency

with multiple hard level I and II lymph nodes. An intraoperative decision to perform an excision biopsy of the doubtful lymph nodes was taken. The excised specimen demonstrated the tumour with atrophy of the associated submandibular gland, probably secondary to pressure effects (FIGURE 2). Primary closure of the wound was done.

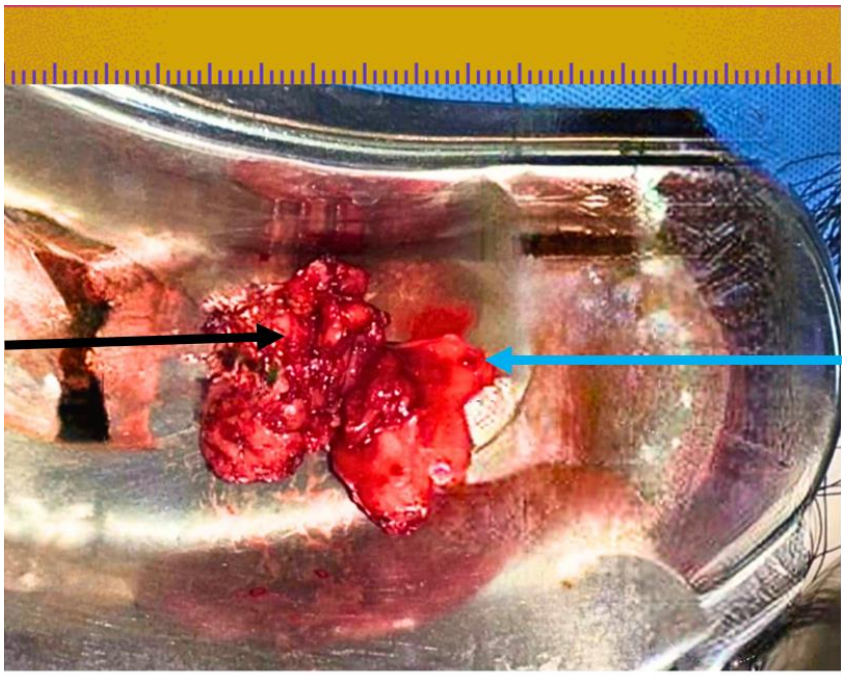

Figure 2 Resected specimen of right submandibular sialoadenectomy. The blue arrow points towards the tumour and the black arrow points towards the atrophied right submandibular gland.

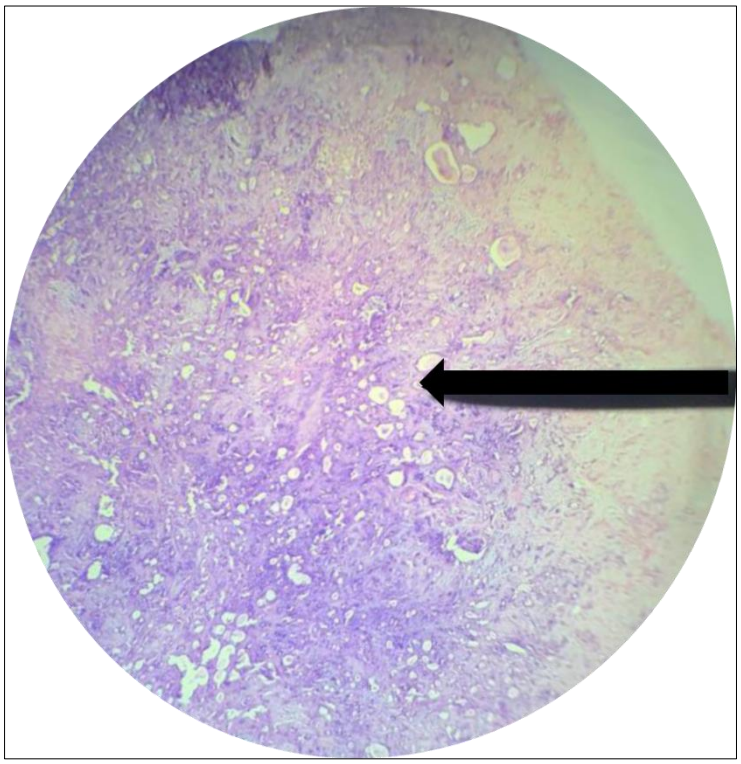

Figure 3 The figure shows dual cellular morphology with epithelial and stromal components, with a benign appearance. This is consistent with pleomorphic adenoma.

2.2. Outcome and follow-up

Postoperatively, the patient developed transient lower motor neuron facial palsy involving the marginal mandibular nerve. The palsy improved with symptomatic treatment in the next 4 weeks. The histopathological report of the resected tumour showed partially encapsulated tissue consisting of a dual population of predominantly epithelial cells in a myxoid stroma. The cell population is benign. No significant atypia/pleomorphism/ mitosis/necrosis was noted (FIGURE 4). Histology is consistent with pleomorphic adenoma of the submandibular gland with reactive lymphadenitis (FIGURE 3). Immunohistochemistry could not be performed as the patient and guardian were unwilling for the same.

3. Discussion

Pleomorphic adenomas of the submandibular gland are rare in the young population, accounting for less than 5% of all salivary gland tumours. Patients with this condition usually present with painless, gradually growing swelling in the submandibular region [2], seldom associated with pressure effects. There is usually no lymph node or nerve involvement [2, 3]. Ultrasonography is usually the first line of investigation but has a low specificity and sensitivity [2]. Thus, the investigation of choice is usually Magnetic resonance imaging or Computed tomography, as done in our case. Fine needle aspiration under ultrasonographical guidance diagnoses the condition downright [2]. These tumours are treated with excision of the tumour with or without removal of the involved gland. Though a rare and predominantly benign entity, removal of the tumour is advised as standard treatment. One of the reasons is due to its close cytological resemblance with low-grade mucoepidermoid carcinoma.

4. Conclusion

Pleomorphic adenoma (PA) of the submandibular gland is rare. They can grow to significant sizes if left untreated due to a lack of awareness or fear of surgery. – Regardless of their size, adenomas can be surgically removed successfully without any morbidity or residual effects. These are the most common salivary gland tumours and may, in some cases, become malignant.

Compliance with ethical standards

Disclosure of conflict of interest

No conflict of interest to be disclosed.

Statement of informed consent

The patient has signed informed consent to include personal details for publication.

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