

CASE REPORT

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# Recurrent pleomorphic adenoma of submandibular gland — a rare entity

Ishan Chauhan<sup>1\*</sup> and Komal Garg<sup>1</sup>

## Abstract

**Background** Among benign salivary gland tumors, pleomorphic adenoma is one of the most common tumor involving submandibular gland, but it is rare to encounter recurrent pleomorphic adenoma in submandibular gland after submandibular gland excision.

**Case presentation** We present a case of 22-year-old female patient who presented to us with recurrent pleomorphic adenoma of submandibular gland region after 10 years of removal of submandibular gland, diagnosis was established with the help of ultrasound and fine-needle aspiration cytology, and thereafter, patient was taken up for surgical excision under general anesthesia. Histopathology confirmed the diagnosis.

**Conclusions** This case report highlights the importance of special caution in removal of submandibular gland along with its capsule especially in pediatric population where due to small facial structures, chances of capsular breach are high, which can result in tumor seedling and future recurrence of tumor.

## Main points

- Pleomorphic adenoma is commonly seen in submandibular region, but it is rare to encounter recurrence of this tumor after complete excision of submandibular gland after many years.
- This case report highlights the importance of complete excision of submandibular gland along with its capsule which is sometimes missed in pediatric age group due to small facial structures.

**Keywords** Pleomorphic adenoma, Recurrence, Submandibular gland, Excision

## Background

Salivary gland tumors though relatively rare account for approximately 5% of all head and neck tumors, while 0.5% of such tumors can be malignant [1]. Pleomorphic adenoma is a benign salivary gland tumor involving mainly major salivary glands; among this, it mainly involves parotid gland in about 63% cases, submandibular gland involvement can occur in about 10% cases, while rarely, it can occur in sublingual gland in about 0.1% of

cases [2]. Recurrence of this tumor can occur due to spillage of tumor cells intraoperatively, or it may occur due to incomplete removal of the gland. Recurrence of such tumors is at very slow rate so patient may present after 10–20 years of primary surgery [3].

## Case presentation

A 22-year-old female presented to us with complain of swelling in left side of the neck since last 1 year. History of left submandibular gland excision was present 10 years back. On examination, swelling of size 2.5 cm × 1.5 cm was noticed in left submandibular region which was mobile, non-tender, and not bimanually palpable. Patient underwent ultrasound neck which showed heterogeneously hypoechoic lobulated mass of size 3 cm × 1.5 cm

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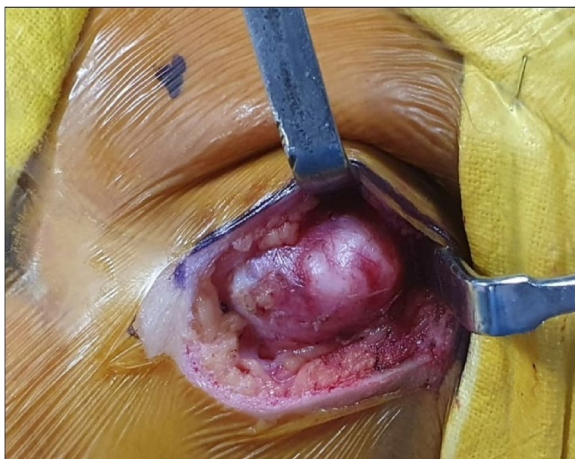
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with raised vascularity in left submandibular region, and left submandibular gland was not visualized. Fine-needle aspiration cytology was suggestive of pleomorphic adenoma. Patient underwent surgical excision under general anesthesia; intraoperatively, two nodules of size 3.5 cm × 2 cm and 1 cm × 1.5 cm were found lying in the submandibular bed which were removed and sent for histopathological examination (Figs. 1, 2 and 3). There was scarred tissue in the submandibular bed which was dissected carefully to preserve marginal mandibular nerve, lingual nerve, and hypoglossal nerve. Submandibular gland was found to be absent; histopathology confirmed the diagnosis of pleomorphic adenoma. Patient was followed up after 1 month; she improved without any complication. Thereafter, patient was advised to report if there is any such recurrence in future, as this tumor is known for recurrences, but patient did not report for any such complaint since then for 6 months till date.

### Discussion

Pleomorphic adenoma also known as benign mixed tumor is most common benign salivary gland tumor and involves both major salivary glands and minor salivary gland. Major salivary gland includes parotid gland, submandibular gland, and sublingual gland. It comprises both epithelial and mesenchymal elements and arises from cells of salivary gland tissue. The tumor is encapsulated when it arises in major salivary glands but not in case of minor salivary gland involvement [2].

Pleomorphic adenoma can be found among all age group, but it is usually common in age group of 30 to 60 years old. Gender wise, it is more common in females with female: male ratio of 2:1 [4]. It usually presents as swelling near angle of mandible when it involves submandibular gland which is bimanually palpable. Recurrence



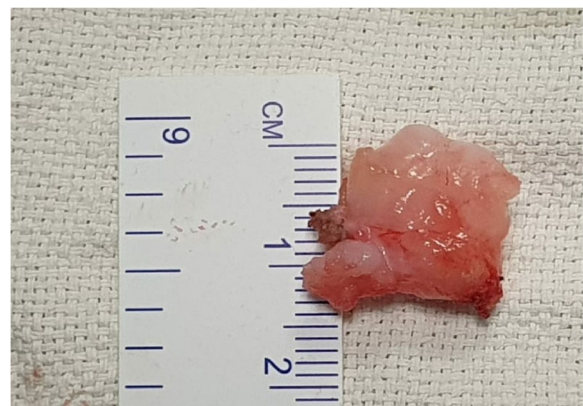
**Fig. 1** Tumor nodule lying in submandibular bed



**Fig. 2** Surgically excised first nodule

of this condition has been reported in 1–5% of cases, while in children it is much higher which may be due to small size of facial structures in children, which may lead to increased risk of violating the tumor capsule and resultant tumor seeding [5]. A rare recurrence of pleomorphic adenoma in submandibular region is attributed to its multicentric behavior. Seeding of tumor during surgical procedure leads to its multicentric behavior. Epithelial remnants of submandibular gland trapped in local lymph nodes explain multicentric behavior in recurrent pleomorphic adenoma [6]. In our case, patient was a child when she got operated first time so recurrence might have occurred, possibly due to violation of tumor capsule and resultant tumor seeding during previous surgery due to which we encountered two tumor nodules during surgical exploration this time.

Diagnosis is made with the help of radiological investigation and pathological investigation (FNAC or incisional biopsy). Preoperative imaging plays an important role in further management and helps us in guiding for fine-needle aspiration cytology if lesion is too small. Ultrasound examination (USG) neck is considered as first-line



**Fig. 3** Surgically excised second nodule

imaging modality because of its high sensitivity; it is also quick, noninvasive and free from radiation, and also easily available [7]. MRI seems the best diagnostic imaging modality as it is superior in differentiating soft tissue and detect deep tissue extension. MRI can also be helpful in detecting perineural spread, any extracapsular involvement, and nerve involvement and can also differentiate edema [8]. Fine-needle aspiration cytology helps in investigating salivary gland lesions but can lead to seedling of tumor along the needle. Incisional biopsy should not be indicated because of risk of fistula [9]. In this case, diagnosis was established using USG and FNAC, though USG could detect only one nodule of size 3 cm × 1.5 cm.

Recurrence of pleomorphic adenoma can occur due to many reasons comprising like if tumor capsule is not completely removed, perforating pseudopodia of tumour, and if capsule gets ruptured during the surgical procedure leading to seedling of tumour cells or incomplete excision of the gland [10]. In our case following second surgery, patient improved without any complications; however, long-term follow-up will be required.

## Conclusion

This case report highlights the importance of complete excision of submandibular gland along with whole of its capsule in cases where we encounter pleomorphic adenoma of submandibular gland. This is particularly important in case of children where facial structures are small and chances of capsular breach are high causing tumor seedling, which at later stages can cause recurrence of tumor.

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## Authors' contributions

IC laid the idea of writing this paper, and IC along with KG performed surgery of the patient. KG wrote the discussion and collected the data. IC did the manuscript writing and final editing of this article. All the authors read and approved the final version of the manuscript.

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## Consent for publication

An informed written consent for publication had been obtained from the patient.

## Competing interests

The authors declare that they have no competing interests.

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