

Heterotopic sites of pleomorphic adenoma

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ABSTRACT

The majority of the tumours occurring in minor salivary glands are malignant with only 18% being benign. Among the benign tumours, pleomorphic adenoma is the most common. Although it presents more commonly in the parotid and submandibular glands, it can occur at any site where salivary glands reside. Only 6.4% of cases of pleomorphic adenoma occur in minor salivary glands, e.g. palate, cheeks, lips, para-pharyngeal space, etc. It occurs in females in the fourth or fifth decade. It presents as an asymptomatic and slow-growing mass. The treatment of choice is wide local excision due to the high rate of recurrence and malignant potential. We present rare case reports of two patients. The first case is of a young 18 years female presenting with recurrent neck (anterior triangle) swelling and the second case is of a 46-year-old female presenting with lower lip swelling. In both cases, it was a slow-growing mass and asymptomatic. Cytological examination of the lesion revealed the diagnosis of pleomorphic adenoma. It was completely excised with no post-operative complication. Pleomorphic adenoma, although a common benign neoplasm of major salivary glands, should always be kept as a possibility at the locations of minor salivary glands. As pleomorphic adenoma can transform into a malignant counterpart, therefore early diagnosis and treatment of the lesion can prevent further complications. The prognosis of pleomorphic adenoma is excellent. However, a close follow-up of the patient is required due to the high rate of recurrence.

Keywords: Benign, lower lip, neck, pleomorphic adenoma

Introduction

Salivary gland tumours are rare, constituting only 2–6.5% of all head and neck tumours.^[1] The tumours of minor salivary glands are uncommon and constitute only 10–25% of all salivary gland tumours. In the minor salivary glands, malignant transformation is more common.^[1,2] Of the benign tumours of the salivary glands, pleomorphic adenoma tops the list.^[1] It is also known as a benign mixed tumour. The parotid gland accounts for 85% of cases of pleomorphic adenomas.^[3] However, these tumours can also present unusual locations, for example, minor salivary glands of the lip, para-pharyngeal space, the cheek, the floor of the mouth, heterotopic salivary gland tissues, etc.^[3,4] The heterotopic rests of salivary gland

tissue can be found in various places, including the pituitary, thyroglossal duct, mandible, neck, etc.^[4]

Here, we discuss two cases of pleomorphic adenoma occurring at heterotopic rests of salivary tissue in the anterior triangle of the neck corresponding to level II lymph nodes and the minor salivary gland of the lower lip.

Case Reports

The first patient is a 28-year-old with slow-growing swelling in the anterior triangle of the neck. The swelling is along the anterior border of the sternocleidomastoid muscle at level II of cervical lymph nodes [Figure 1]. It was a small-circumscribed, mobile, well-circumscribed swelling. She gave a history of exactly swelling at the exact location 10 years back, which was excised surgically; however, no histopathology testing was done then. The clinical diagnosis of the current swelling was lymphadenopathy or keloid, and the patient was sent for fine-needle aspiration

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Received: 09-08-2022

Revised: 09-11-2022

Accepted: 01-02-2023

Published: 31-05-2023

Access this article online

Quick Response Code:



Website:
www.jfmprc.com

DOI:
10.4103/jfmprc.jfmprc_1591_22

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How to cite this article: Kaur J, Kaur M. Heterotopic sites of pleomorphic adenoma. *J Family Med Prim Care* 2023;12:1009-11.

cytology (FNAC) testing. Fine-needle aspiration (FNA) yielded thick mucoid material in the needle's hub. The smears were prepared from the mucoid material. Microscopic examination showed cellular smears with a biphasic tumour. The stromal component was seen as abundant chondromyxoid stroma, and the epithelial component was seen as monolayered sheets of benign-looking epithelial cells with bland round to oval nuclei and a moderate amount of cytoplasm [Figure 2]. Myoepithelial cells were also present in the background exhibiting spindle-shaped morphology. Based on these features, the diagnosis of pleomorphic adenoma was made. The swelling was excised successfully by the surgeon. Histopathologically, the features were consistent with pleomorphic adenoma [Figure 3]. The patient was followed up and is doing well without any recurrence.

The second patient is a 46-year-old female. She also presented with a slowly progressive asymptomatic swelling on the lower lip from the last ten months. It was around 1 × 1 cm in size, non-tender and had soft swelling. No other notable lesion was identified in or around the oral cavity. There was no history of trauma, and the dental history was unremarkable. The provisional diagnosis made by the clinician was mucocele. FNAC was done, and it yielded blood admixed aspirate. The smears were prepared, showing high cellularity comprising groups and clusters of benign ductal epithelial cells and myoepithelial cells. There was abundant chondromyxoid stroma in the background. Therefore, a diagnosis of pleomorphic adenoma was made. The patient was lost to follow-up.

Discussion

The most common benign tumour in the salivary glands is a pleomorphic adenoma, yet the exact aetiology is unknown. It is thought that these tumours originate from the reserve cells of intercalated ducts. The molecular studies show chromosomal alteration at 8q12 and 12q15.^[5] It can occur at any age and sex, yet females in their fourth to sixth decade are more commonly affected.^[1] The female/male ratio is 1.9:1.^[5] It occurs more typically between 7 and 16 years of age in children.^[6]

Pleomorphic adenoma can also occur at heterotopic salivary gland tissue (HGST)^[7] as seen in our case. HGST refers to salivary gland tissue present outside the major/minor/accessory salivary gland tissue.^[7] These ectopic islands can be present in various places like the pituitary, temporal bone, mandible, thyroglossal duct, and rarely in the neck.^[4,7] In the neck region, although rare, the heterotopic rests of salivary tissue can be found along the anterior border of the sternocleidomastoid muscle.^[4] Various theories have been put forward to explain the basis of HGST, for example, epithelial remnants from the brachial apparatus.^[8] Pleomorphic adenoma is the second most common tumour of HGST, the first being Warthin's tumour.^[7] Very few cases of pleomorphic adenoma at ectopic salivary tissue in the upper cervical region have been described so far.

In the case of minor salivary glands of lips, the propensity of pleomorphic adenoma is more on the upper lip than the lower lip. The upper lip tumours are more commonly benign, whereas



Figure 1: The patient presented with recurrent swelling at the anterior triangle of the neck

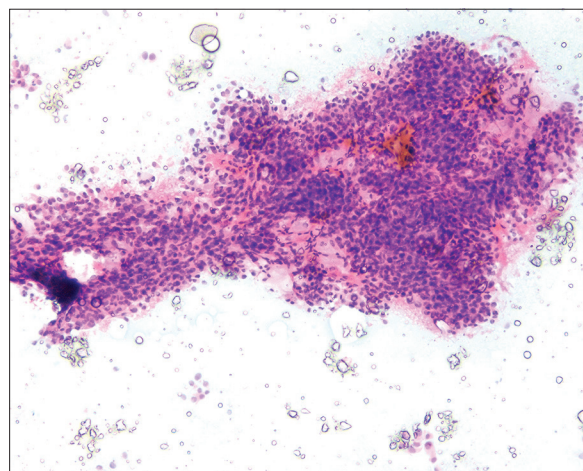


Figure 2: Cytopathological examination shows benign-looking epithelial cells with bland morphology (200×)

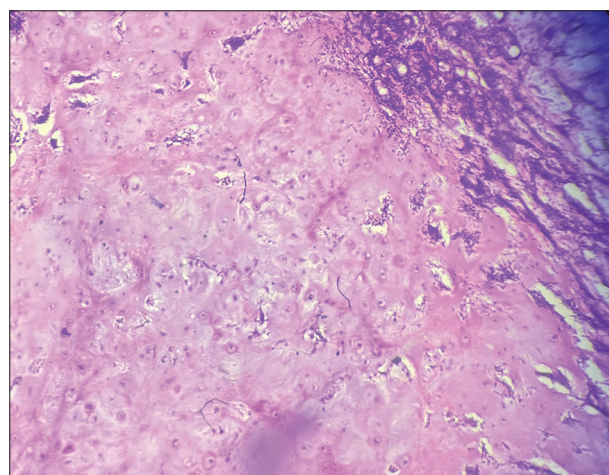


Figure 3: Histopathological examination shows abundant chondromyxoid stroma and few epithelial cells (200×)

lower lip tumours are more commonly malignant due to the difference in their embryonic development.^[9] In our case, this is

an exception, as we reported a benign pleomorphic adenoma on the lower lip. The incidence of pleomorphic adenoma on lips is seen a decade earlier and peaks in the third or fourth decade.^[5] Fine-needle aspiration cytology is a safe and reliable pre-operative test for diagnosing pleomorphic adenoma. However, FNAC may pose some diagnostic difficulties due to variations in the cellular composition of pleomorphic adenoma.^[10] According to a study by Vaidya *et al.*,^[11] FNAC has a sensitivity of 95.83% and a specificity of 97.05%. The study also showed that the diagnostic accuracy of FNAC in diagnosing pleomorphic adenoma is 96.55%.

Pleomorphic adenoma at heterotopic sites, like swelling at the anterior triangle of the neck and lower lip, can be kept as a differential diagnosis. The clinician should keep a high index of suspicion and adequately clear all the margins with a cuff of surrounding dispensable normal tissues, which is essential for preventing recurrence.^[9] Therefore, the complete wide local resection of the tumour with negative margins is the treatment of choice.^[2] Many have used post-operative radiation to prevent a recurrence.^[9] The patients should be kept on the long-term follow-up to see for any recurrence or malignant transformation of the swelling.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient (s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

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