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Journal of Health and Allied Sciences NU

Article in Press

Case Report

Benign Mixed Tumour Diagnosed over Fine Needle Aspiration Biopsy: A Case Report

Urvashi Shetty[1], Gopinath Thilak[1], Vinisha S. Pousya[1], Yashi Aggarwal[1]

[1]Department of Oral and Maxillofacial Pathology and Oral Microbiology, Attavar Balakrishna Shetty Memorial Institute of Dental Sciences, NITTE (Deemed to be University), Mangalore, India

ABSTRACT

Pleomorphic adenoma (PA) accounts for 50-70% of all salivary gland neoplasms. As the name suggests, it shows varied histological morphology. This entity accounts for about 65% of all salivary gland tumours and is well known for its exceptional ability to grow to enormous sizes if left untreated. Surgical resection is the most preferred treatment; however, the multi-nodular presentation of the tumor and incomplete resection are two challenging factors that may lead to recurrence. Optimal diagnostic measures for PAs can reduce the risk of recurrence and transformation to malignancy. We present a case of recurrent PA of the parotid gland in a 30-year-old female patient who was diagnosed 5 years ago, treated, and is now diagnosed with the same by a fine-needle aspiration biopsy.

Keywords: Biopsy, Parotid gland, Pleomorphic adenoma, Parotidectomy, Recurrence

INTRODUCTION

Pleomorphic adenoma (PA) represents the most common neoplasm of the salivary glands, accounting for approximately two-thirds of all cases. Commonly arising in the parotid gland with a predilection for females in their third to sixth decades of life, PA clinically presents with an asymptomatic, slow-growing, and unilateral mass. It can eventually become enormous in size, weighing several kilograms.[1]

PA is named for its diverse microscopic appearance, characterized by mix of epithelial, and myoepithelial cells within a variable stroma that appears chondroid, myxoid, or osteoid.[2] A delicate and imperfect pseudo capsule surrounds the tumour, which is delineated by minute finger-like extensions into the surrounding parotid tissue. This capsule is responsible for isolating the tumour nodules from the surrounding structures.[3] Rupture of the capsule and subsequent spillage of tumour content during excision is the prime reason associated with the recurrence of these tumours.[4] Recurrences are often extensive, bearing a demanding risk of facial nerve damage and re-recurrence. Initial management should be optimal and necessary to reduce the recurrence rate and transformation to malignancy.[5] This article reports a case of plemorphic adenoma (PA) of the parotid gland, which had exhibited characteristic clinical and histological features and was surgically treated 5 years ago and subsequently recurred. The recurred PA was diagnosed over a fine needle aspiration biopsy (FNAB).

CASE REPORT

A 30-year-old female patient reported to a private clinic with a complaint of progressive swelling in the left backside of the ear for 6 months associated with pain. The patient gave a history of similar swelling 5 years ago, which was histopathologically diagnosed as a PA. She was operated on. On extraoral examination, a diffuse swelling measuring approximately 2.0 x 2.0 cm in size was noted on the left side of the face near the left ear. The swelling was firm with no discharge [Figure 1].

Since the lesion had recurred and was associated with progressive swelling and pain, this created an alarm. FNAB (Fine needle aspiration biopsy) was performed, and the fluid was sent immediately for cytopathological evaluation.

***Corresponding author:** Dr. Yashi Aggarwal, Department of Oral and Maxillofacial Pathology and Oral Microbiology, Attavar Balakrishna Shetty Memorial Institute of Dental Sciences, NITTE (Deemed to be University), Mangalore, India. yashiaggarwal7474@gmail.com

Received: January 14, 2025 **Accepted:** March 21, 2025 **EPub Ahead of Print:** May 29, 2025 **Published:** *** **DOI:** 10.25259/JHS-2024-7-16-R1-(1486)

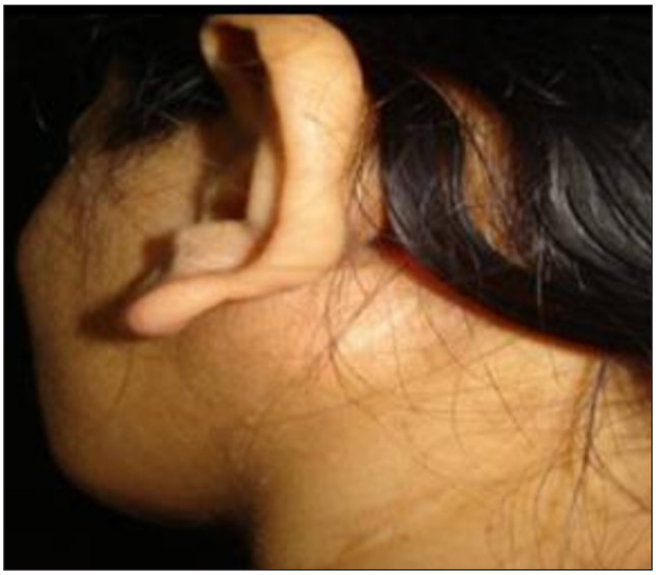

Figure 1: Diffuse swelling measuring approximately 2.0 x 2.0 cm in size was noted on the left side of the face near the left ear.

CYTOPATHOLOGICAL EXAMINATION

Three smears were prepared and stained with a PAP stain. They were highly cellular and had aggregates of spindle to ovoid-shaped cells with prominent nuclei, which were suggestive of myoepithelial cells and ductal cells that are seen in the background of a myxomatous-like matrix. No cytologic atypia was noted. This led to our differential diagnosis of recurrent PA, Carcinoma ex PA, and epithelial myoepithelial carcinoma [Figure 2]. A biopsy was advised.

SURGICAL INTERVENTION

Revision surgery was indicated, following which the lesion was surgically excised, and the excised tissue was sent for further pathological evaluation.

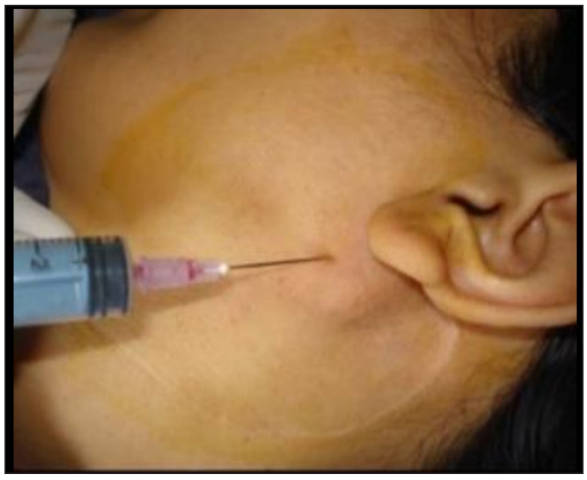

Figure 2: The clinical site of fine needle aspiration cytology.

HISTOPATHOLOGICAL EXAMINATION

The excised tissue pathology revealed a well-defined lesion surrounded by an incomplete fibrous capsule, separating the tumour from the adjacent glandular structures. The mass of the tumour was predominantly myxoid with components of epithelium, which was seen in various forms like ducts, cystic structures, small cellular nests, and anastomosing cord formations. Some chondroid stroma was also seen in dispersed areas. The cells were epithelioid and spindle-shaped with scanty pale cytoplasm, and the nucleus was vesiculated. The ductal structure was filled with eosinophilic material [Figure 3]. In summary, the examination revealed myoepithelial and ductal cells embedded within a myxoid stroma, with occasional areas showing chondroid-like differentiation. The major differential diagnoses considered included carcinoma ex pleomorphic adenoma and epithelial-myoepithelial carcinoma. Both entities were ruled out due to the absence of definitive invasion into surrounding tissues and the lack of cytological atypia, such as an increased nuclear-to-cytoplasmic ratio, variations in nuclear and cellular shape, or the presence of comedo necrosis. Taking into account the cytological features and correlating with the clinical history, a diagnosis of recurrent pleomorphic adenoma was made.

DISCUSSION

PA is a benign salivary gland neoplasm characterised by architectural diversity with an admixture of myoepithelial cells and ductal cells present against a myxoid to the chondroid

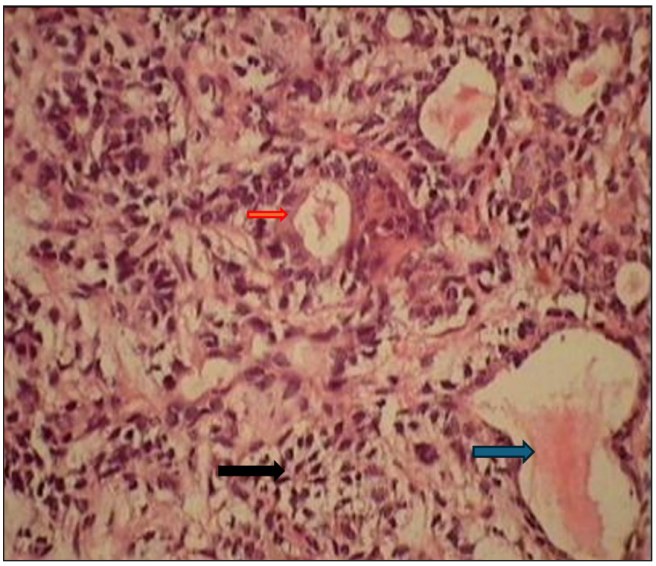

Figure 3: Photomicrograph shows ductal and myoepithelial cells arranged in a tubular pattern against a myxoid background (represented by an orange arrow). The ductal structure was filled with eosinophilic material (represented by a blue arrow). The myoepithelial cells are spindle to ovoid in shape (represented by a black arrow) and stained with Papanicolaou stain. 40x magnification.

stroma. It is caused by trans-location or change in intra-chromosomal arrangements of the PLAG1 on 8q12 gene. In about 50% of the cases, this rearrangement was noted, and in about 10-15% of cases, HMGA2 on 12q14.3 gene was noted.[6] It is a benign yet true neoplasm that occurs predominately in the superficial lobe of the parotid gland and commonly presents as a swelling on the ramus of the mandible in front of the ears. According to the literature, the age of occurrence of PA is 30 – 60 years, with a slight predilection towards females (2:1).[4,7] It is generally asymptomatic and may appear as a firm, irregular, and nodular lesion. The tumour will become bizarrely large if left untreated and could cause extrinsic facial nerve compression.[7] Few cases of PA transforming into malignancy have been reported in the literature.

The diagnostic approach for tumour confirmation includes Computed Tomography (CT) and Magnetic Resonance Imaging (MRI). However, MRI can yield more accurate results pertaining to identifying the tumour margins and precise location in relation to surrounding tissues.[8] FNAB has been used to differentiate malignant and benign tumours.[9] It is one of the most cost-effective methodology used for identifying palpable masses of the salivary gland. In literature, many studies support this evidence. One of the studies conducted by Fazilet Altin et al. revealed that FNAB can be utilized as a diagnostic tool in pre and post-operative biopsies of parotid gland tumours. They found out that the diagnostic accuracy of FNAB when detecting malignancy was 86.52%. The sensitivity and specificity were 68.96% and 89.63%.[10] Currently, USG-guided FNAB is preferred. A study conducted by Fiková A et al.[11] in 2024 highlighted the utility of FNAB in the diagnosis and management of parotid PA. They used USG-guided FNAB for diagnosing PA. They recorded a sensitivity of 88.83%, a specificity of 96.23%, and an accuracy of 92.31%.[11]

PAs have pseudocapsules with microscopic finger-like protrusions called pseudopodia, which extend to the adjacent normal tissue. There are high chances of tumour spillage because of these extensions. Hence, incisional biopsy of PA is avoided. Enucleation of PA is not preferred as it is a procedure in which normal tissue margin cannot be appreciated much, and PA cases enucleated have a recurrence rate of 45%. Violation of the tumour during enucleation could lead to the dispersion of numerous new nodules throughout the surgical field and perhaps beyond the scar line in unaffected tissue. The recurrence of PA was assessed and concluded to be less than 0.4% after total parotidectomy and between 2% and 5% after superficial parotidectomy.[12]

In recurrent cases of PA, due to the occurrence of nonspecific changes in the anatomy of the postsurgical field, it becomes difficult to positively identify the areas of residual tumour by radiographic imaging or other diagnostic methods, posing difficulty for the diagnostician in identifying the residual tumour.[12,13]

MRI is the hallmark for diagnosis of salivary gland tumours and their recurrence. Another useful diagnostic tool is the examination of the tumour contrast in conjunction with the assessment of the apparent diffusion coefficient.[13] According to some publications, RPA frequently manifests as multifocal tumours, including both the tumour bed and the surrounding areas, which are easily distinguished on T2 sequences.[14]

The usefulness of adjuvant radiation in RPA is debatable as in many studies it has shown that it can significantly correct the spillage of tumour especially during revision surgeries or can be utilized to identify residual tumour. It should be remembered that it is challenging to justify radiation therapy, and it should be kept as a last resort for insufficient surgery in initial as well as RPA to prevent the recurrence of a benign tumour with a slight preference for younger individuals.[15]

CONCLUSION

The diagnosis of salivary gland tumours is often challenging due to ambiguous clinical, radiological and overlapping histopathological features. These tumours exhibit a wide range of morphological patterns. Distinguishing between benign and malignant lesions is essential for proper management and accurate prognosis assessment. The key to successful therapy is a strong clinical suspicion and adequate tumour clearance with a cuff of surrounding discardable normal tissues. FNAB can be used in conjunction with MRI to help distinguish between benign and malignant tumours. Adjuvant radiation therapy remains a subject of debate in the management of pleomorphic adenoma. It may be helpful in cases with recurrent multifocal PA, but the resection must be as thorough as possible.

Ethical approval: Institutional Review Board approval is not required.
Declaration of patient consent: The authors certify that they have obtained all appropriate patient consent.
Financial support and sponsorship: Nil.
Conflicts of interest: There are no conflicts of interest.
Use of artificial intelligence (AI)-assisted technology for manuscript preparation: The authors confirm that there was no use of artificial intelligence (AI)-assisted technology for assisting in the writing or editing of the manuscript and no images were manipulated using AI.

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How to cite this article: Shetty U, Thilak G, Pousya VS, Aggarwal Y. Benign Mixed Tumour Diagnosed Over Fine Needle Aspiration Biopsy: A Case Report. J Health Allied Sci NU. doi: 10.25259/JHS-2024-7-16-R1-(1486)