Diagnostic Pitfalls in Salivary Gland Tumours : A Case Series

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ABSTRACT

Introduction

Salivary gland tumours are rare and heterogeneous neoplasms, accounting for 3–6% of head and neck tumours. Pleomorphic adenoma (PA) is the most common benign tumour and is often suspected clinically because of its typical presentation as a painless, slow-growing swelling. However, several other entities may mimic PA, creating diagnostic challenges.

Case Presentation

This case series includes four patients presenting with painless, slow-growing salivary gland swellings: case 1 of the parotid gland, case 2 of the submandibular gland, case 3 of the sublingual gland, and case 4 of the parotid gland, with clinical and imaging features suggestive of pleomorphic adenoma. All patients underwent radiological evaluation, fine-needle aspiration cytology (FNAC), and definitive surgical excision. Cytological interpretation was performed using the Milan System for Reporting Salivary Gland Cytopathology, which gave us the mirage of pleomorphic adenoma in case 1 and salivary gland neoplasm of unknown potential(SUMP) in the remaining three cases, but histopathologically all the four cases were reported differently as case 1 as myoepithelioma, case 2 and 3 as Adenoid cystic carcinoma(AdCC) whose grading was assessed using the MD Anderson grading system and case 4 as Basal cell adenoma.

Conclusion

This case series emphasises the diagnostic overlap between pleomorphic adenoma and other salivary gland tumours such as AdCC and myoepithelioma. FNAC and imaging have limitations in distinguishing these entities, and histopathology remains the gold standard for accurate diagnosis and management. Awareness of this overlap is crucial, particularly in submandibular and sublingual gland tumours, where the probability of malignancy is higher.

Keywords

Salivary Gland Tumours; Pleomorphic Adenoma; Adenoid Cystic Carcinoma; Myoepithelioma; Basal Cell Adenoma; Diagnostic Dilemma; Histopathology

Salivary glands are exocrine glands in the head and neck that produce and secrete saliva into the oral cavity. Their tumours are uncommon, constituting approximately 3–6% of all head and neck neoplasms.[1] Despite their rarity, they demonstrate a remarkable histological spectrum, with the World Health Organization (WHO) recognizing more than 20 distinct benign and malignant subtypes.[2] The parotid gland is most frequently affected, followed by the submandibular, sublingual, and minor salivary glands. A general rule is that the smaller the salivary gland, the higher the likelihood of malignancy.[3]

Among benign neoplasms, pleomorphic adenoma (PA) is the most common, accounting for about 60% of cases. It typically presents as a slow-growing, firm, mobile, and painless swelling, which often leads clinicians to suspect PA initially. However, several other tumors—including adenoid cystic carcinoma (AdCC), myoepithelioma, mucoepidermoid carcinoma, and basal cell adenoma—can present with similar clinical and radiological features. This overlap creates a diagnostic dilemma, particularly since fine-needle aspiration cytology (FNAC) and imaging may have limited ability to distinguish PA from these mimics. Consequently, histopathological examination remains the gold standard for definitive diagnosis.

Among malignant tumours, AdCC warrants special attention due to its paradoxical clinical course—slow progression but aggressive behaviour, characterised by

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perineural invasion, frequent recurrence, and late distant metastasis, most commonly to the lungs and bones. In contrast, myoepithelioma is a rare benign neoplasm derived from myoepithelial cells. While less aggressive than PA, it carries a risk of recurrence if incompletely excised.

Given these challenges, salivary gland tumors that clinically mimic pleomorphic adenoma but prove to be different entities histologically underscore the importance of thorough evaluation and tailored surgical management.

In addition to conventional diagnostic modalities, structured reporting and grading systems are crucial for the evaluation of salivary gland neoplasms. The Milan System for Reporting Salivary Gland Cytopathology (MSRSGC) standardises FNAC reporting into categories with defined risks of malignancy, such as Neoplasm: Salivary gland neoplasm of uncertain malignant potential (SUMP – Milan IVB), which carries an intermediate risk and often warrants excision for definitive diagnosis. Furthermore, the MD Anderson (MDA) histologic grading system for adenoid cystic carcinoma stratifies tumours based on the percentage of solid components—Grade I lacking solid areas, Grade II showing <30% solid, and Grade III demonstrating ≥30% solid—providing important prognostic information.

The integration of structured diagnostic systems improves the accuracy of characterising salivary gland lesions.

In this case series, we present four patients with classical features suggestive of pleomorphic adenoma; however, the final diagnoses varied. These cases highlight diagnostic pitfalls in salivary gland pathology and reaffirm the indispensable role of histopathology in accurate classification and management planning.

Case 1 : Myoepithelioma of Parotid Gland

A 62-year-old male presented to our department with a painless, slow-growing swelling in the preauricular region of two years' duration. On clinical examination, a firm, ovoid swelling measuring approximately 5 × 3 cm was noted in the parotid region, extending superiorly up to the zygomatic arch, inferiorly just below the angle of the mandible, and posteriorly up to the mastoid process, with a smooth surface and well-defined margins (Figure 1A). No facial nerve weakness was observed. Ultrasonography

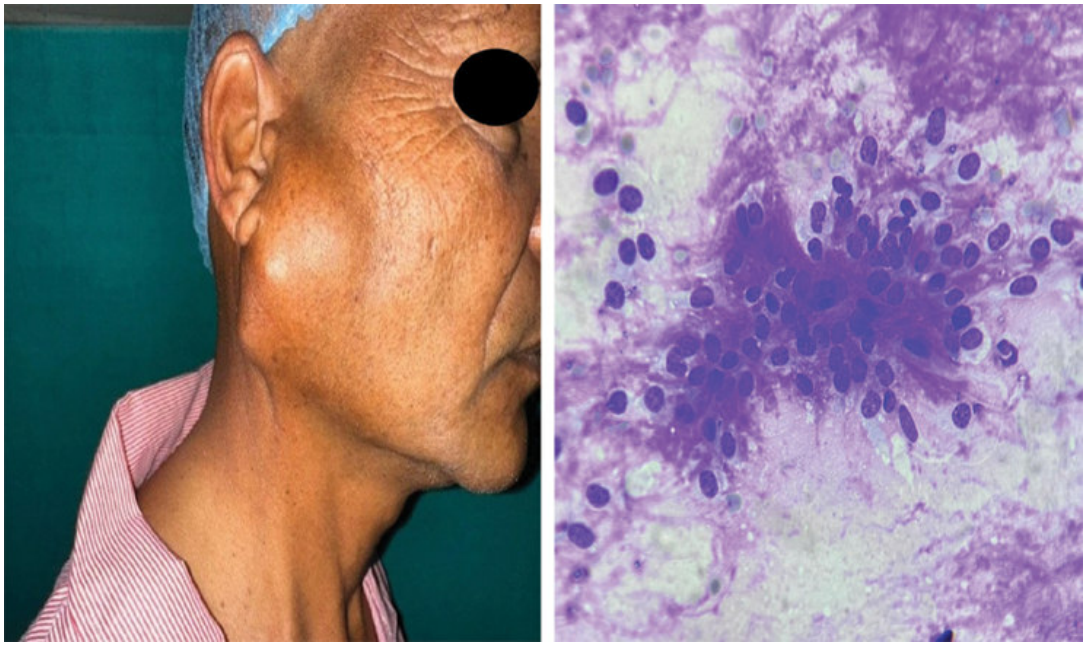

Fig. 1. Showing preoperative clinical image (a) and cytopathology; H&E staining at 40X (B) which is suggestive of pleomorphic adenoma of parotid gland

and contrast-enhanced magnetic resonance imaging were suggestive of pleomorphic adenoma. Fine-needle aspiration cytology revealed sheets and cohesive clusters of ductal epithelial cells admixed with round to spindle-shaped myoepithelial cells in a scant chondromyxoid, feathery stroma with a hemorrhagic background. No cellular atypia was noted, and the findings were suggestive of pleomorphic adenoma (Figure 1B).

Total parotidectomy was performed. Histopathological examination revealed a tumor composed of sheets of round to polygonal cells with round to oval nuclei, vesicular chromatin, and a moderate amount of eosinophilic to clear cytoplasm. Mild intratumoral edema and microcystic changes were observed, along with infrequent small acinar structures. No mitotic activity or necrosis was identified. The initial impression was that of a primary low-grade, myoepithelial-rich salivary gland neoplasm, with differential diagnoses including myoepithelioma and epithelial–myoepithelial carcinoma (Figure 2). Immunohistochemistry demonstrated positivity for CK, p40, SOX10, and CK7, confirming the diagnosis of myoepithelioma. On follow-up, the patient developed Grade IV facial nerve weakness, which subsequently improved with facial physiotherapy and conventional medical management.

Case 2 : Adenoid cystic carcinoma of the submandibular gland

A 32-year-old female presented to our department with a painless, slow-growing swelling in the right submandibular region of two months' duration. Clinical examination revealed a solitary, firm to hard swelling measuring approximately 4 × 3 cm, located below the angle of the mandible in the submandibular fossa and palpable bimanually, with no associated cranial nerve weakness. Ultrasonography and contrast-enhanced computed tomography demonstrated a well-defined lesion measuring 3.6 × 2.8 cm, suggestive of pleomorphic adenoma of the submandibular gland (Figure 3A). Two fine-needle aspiration cytology samples were obtained: the first, reported from an outside centre, was consistent with pleomorphic adenoma, while the second, evaluated at our institution, was categorised as a salivary gland neoplasm of uncertain malignant potential (SUMP) corresponding to Milan System category IVB, carrying an estimated

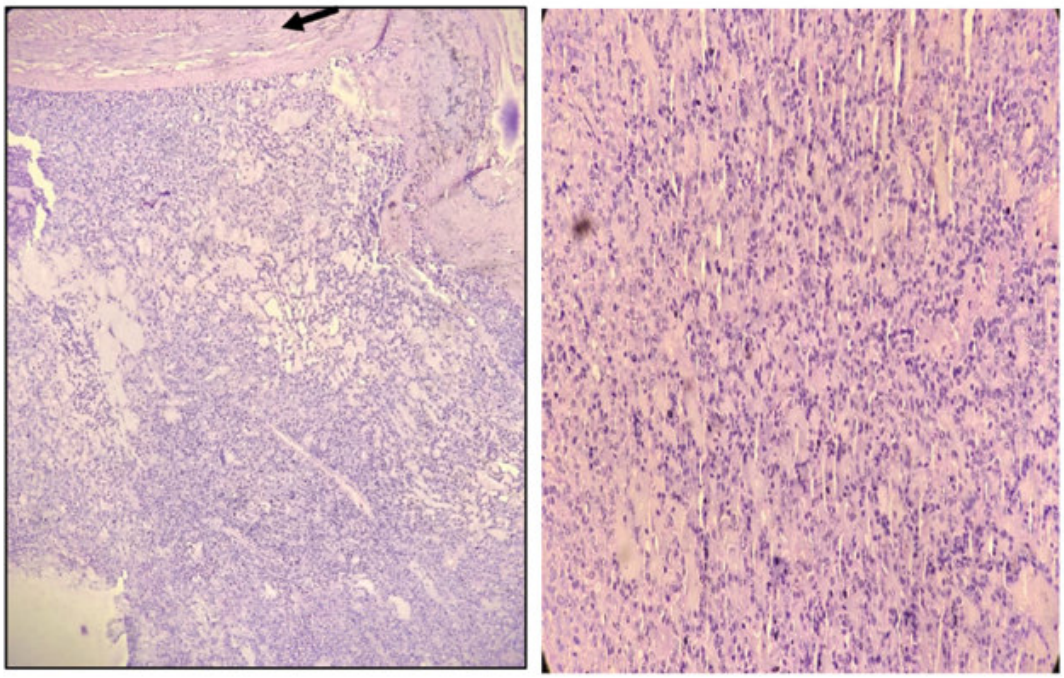

Fig. 2. Showing histopathology of myoepithelioma in 10X and 40X (H & E Staining)

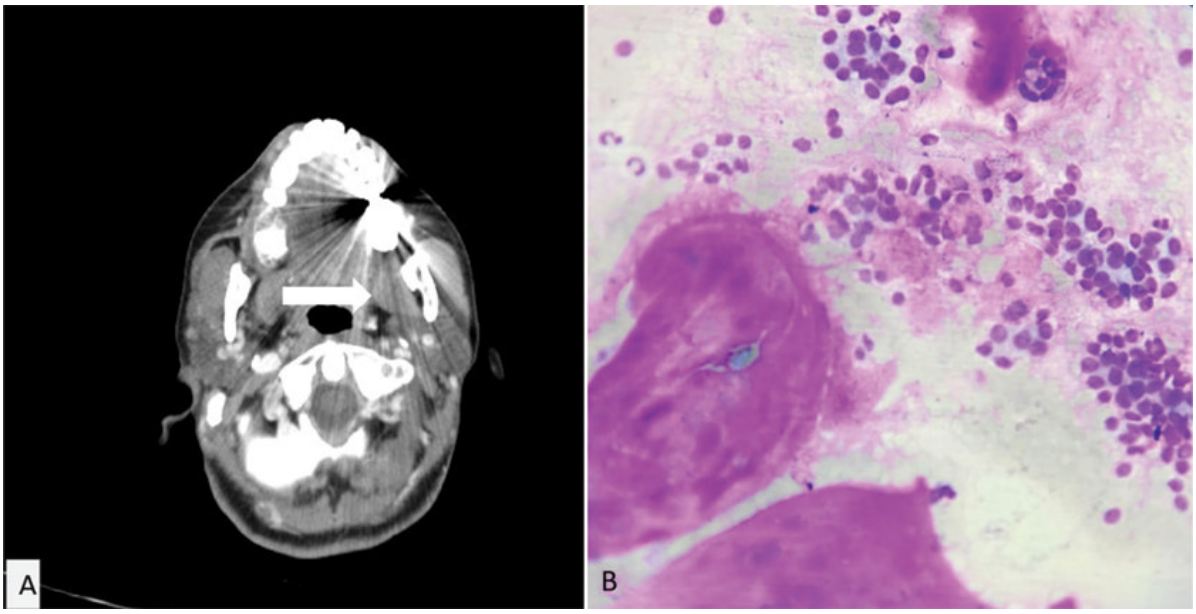

Fig. 3. Showing preoperative radiologival image of tumor (A) and cytopathology image; H & E staining (B) at 40X suggestive of milan category IVB

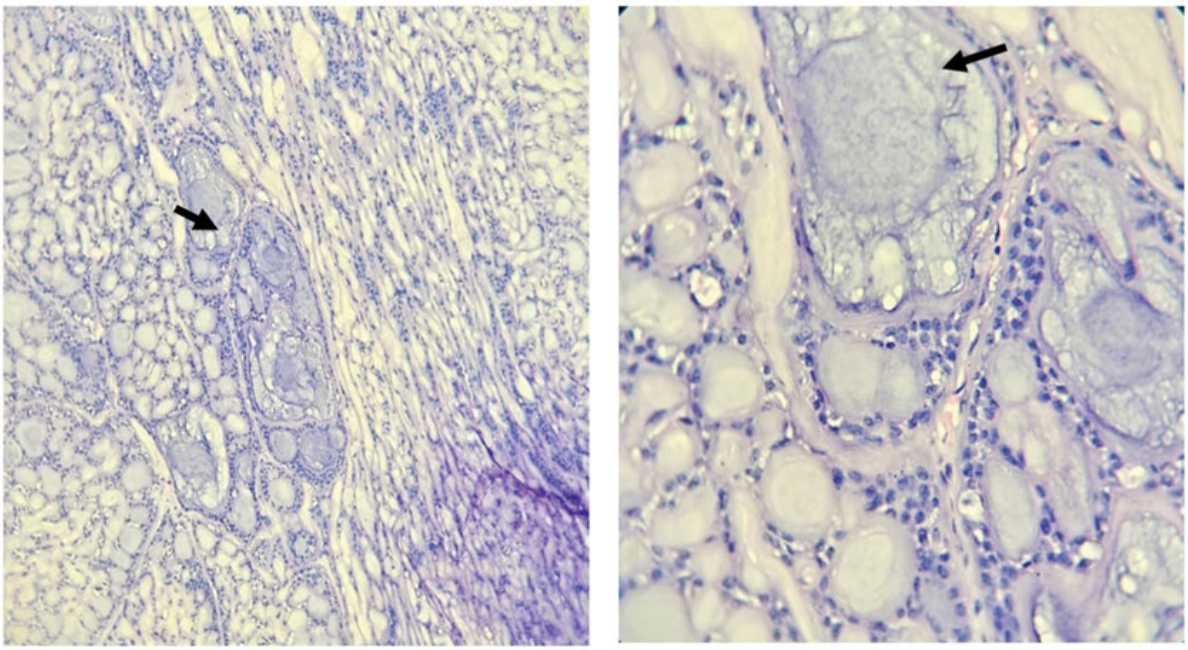

Fig. 4. Histopathology of Adenocystic Carcinoma of submandibular gland (MD Anderson Grade 1) at 10X and 40 X [H & E Staining]

35% risk of malignancy. This categorisation guided the decision to proceed with complete surgical excision for definitive diagnosis (Figure 3B).

Submandibular gland excision with meticulous nerve preservation was performed. Histopathological examination revealed tumor cells arranged predominantly

in a cribriform architecture, with only occasional tubular and solid components. The tumor cells were predominantly myoepithelial, exhibiting numerous hyaline globules and extensive stromal hyalinization. Focal areas showed myoepithelial cells with clear cell change. There was no significant pleomorphism or mitotic activity, and no evidence of necrosis, perineural invasion, or lymphovascular invasion was identified. The overall features were consistent with adenoid cystic carcinoma of the submandibular gland, classified as MD Anderson Grade I (Figure 4). MD Anderson Grade I denotes the absence of solid areas and is associated with the most favorable prognosis in adenoid cystic carcinoma grading.On follow-up, contrast-enhanced computed tomography of the chest was performed to evaluate for distant metastasis. Metastatic disease was detected, following which the patient underwent radiotherapy as part of the management for metastatic involvement.

Case 3 : Adenoid cystic carcinoma of the sublingual gland

A 26-year-old male presented to our department with a painless, slow-growing swelling in the floor of the mouth for a duration of two months. On clinical examination, a solitary, hard, fixed, and tender swelling measuring approximately 6 × 2 cm was noted in the floor of the mouth, extending deep into the submental space. Ultrasonography and contrast-enhanced magnetic resonance imaging were suggestive of pleomorphic adenoma (Figure 5A). Fine-needle aspiration cytology revealed cohesive clusters and sheets of basaloid cells, at times forming acinar structures, along with singly scattered cells demonstrating a high nuclear-to-cytoplasmic ratio. Based on these findings, the lesion was categorised as a salivary gland neoplasm of uncertain malignant potential (SUMP), corresponding to Milan System grade IVB. The cytomorphological features were suggestive of a cellular neoplasm with basaloid characteristics (Figure 5B).

Sublingual gland excision with meticulous nerve preservation was performed. Histopathological examination revealed a biphasic malignant salivary gland tumour composed of ductal and myoepithelial cells arranged in multiple nodules exhibiting lobular, cribriform, and focally solid architectural patterns, separated by thick fibrous hyalinized septae. Perineural invasion was identified. Although the exact proportion of the solid

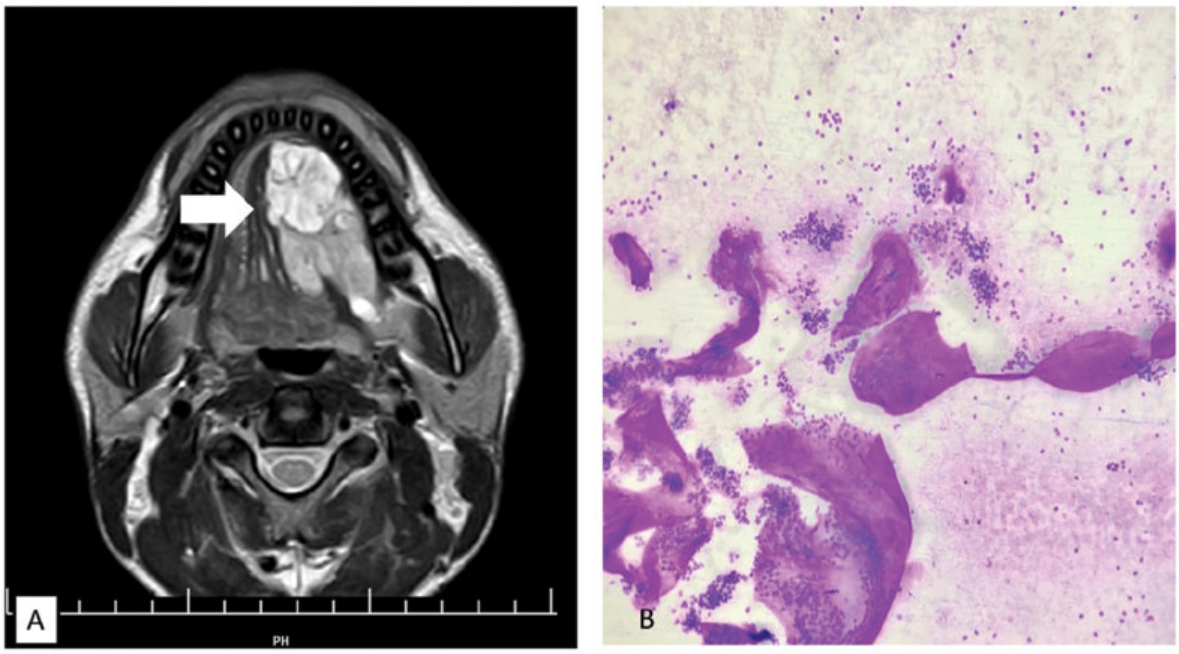

Fig. 5. Preoperative radiological image (A) and cytopathology {40X}; H & E Staining (B)

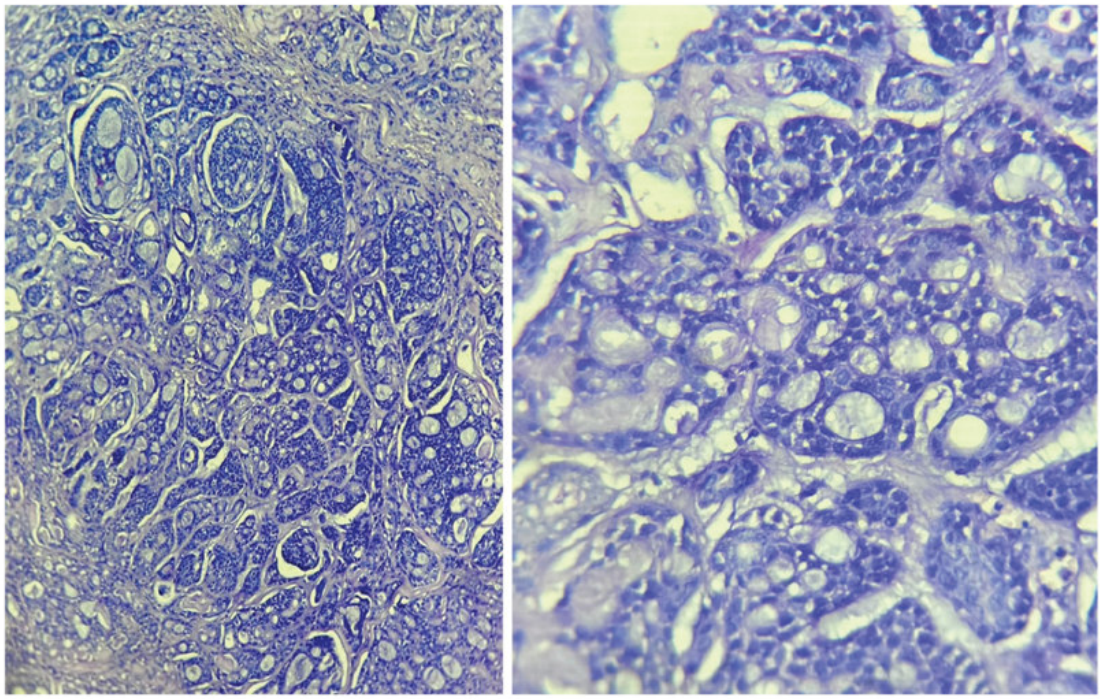

Fig. 6. Showing a histopathological image of the sublingual gland, Adcc at 10X and 40X (H & E Staining)

component was not quantified, the presence of solid areas indicates that the lesion is at least MD Anderson Grade II and may qualify as Grade III if the solid component comprises 30% or more. The overall histomorphological features were consistent with adenoid cystic carcinoma, cribriform type, with perineural invasion (Figure 6).On follow-up, the patient was sent to the Oncology Department for further management.

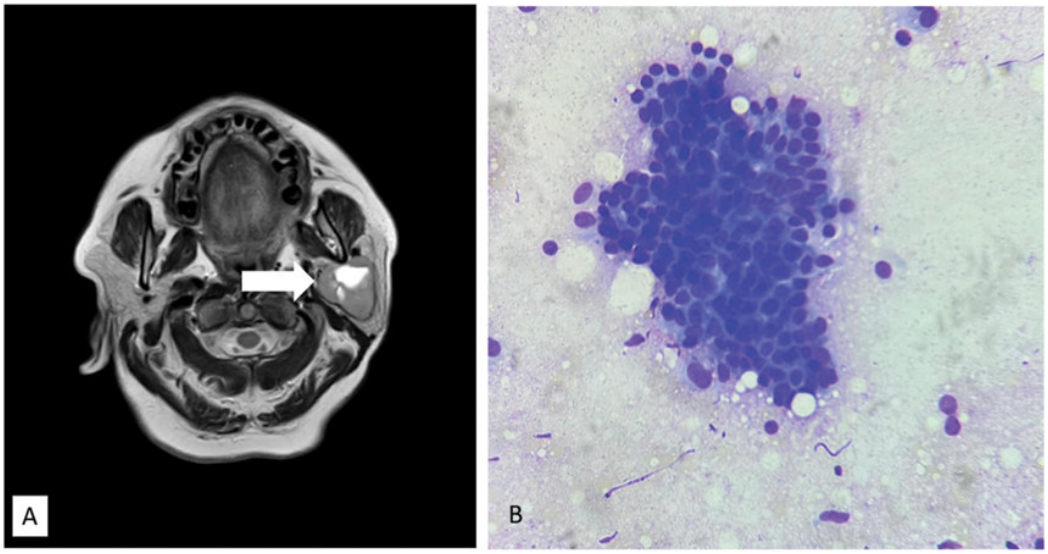

Fig. 7. Showing preoperative radiological image (A) and Cytopathology {40X}; H & E staining (Cellular basaloid Neoplasm) of Tumor (B)

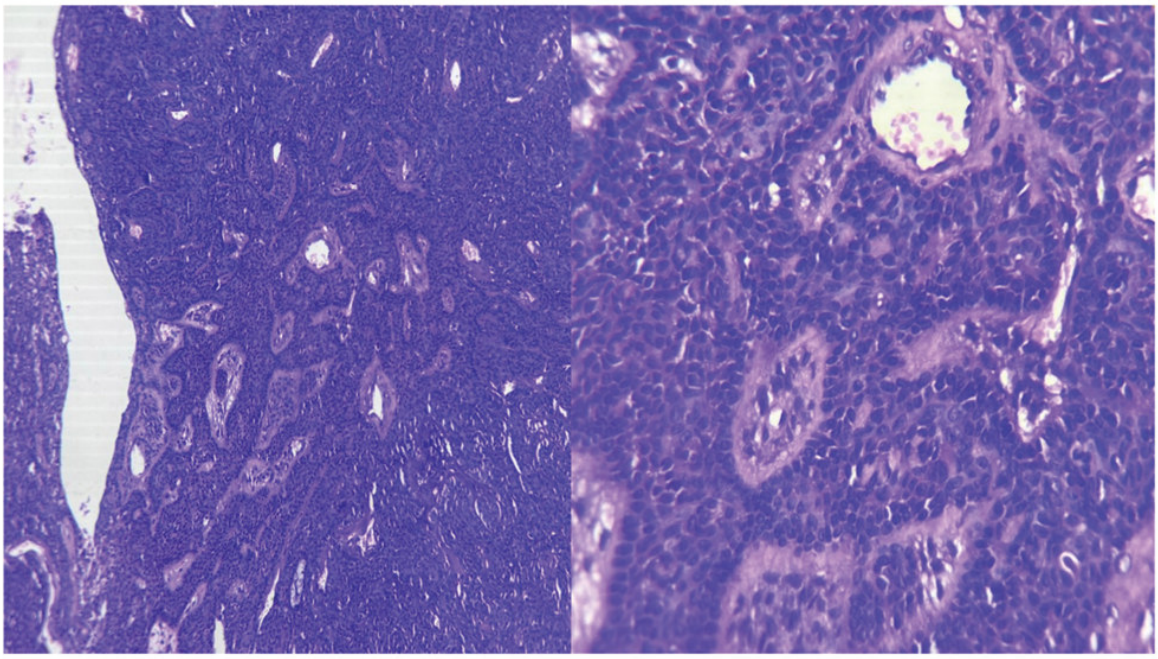

Fig. 8. Showing histopathology image at 10X and 40X respectively suggestive of basal cell adenoma (H & E Staining)

Case 4 : Basal cell adenoma of the deep lobe of the parotid gland

A 64-year-old female presented to our department with a painless, slow-growing swelling in the left parotid region of three years' duration. On clinical examination, a solitary, firm to hard swelling measuring approximately 4.5 × 3.5 cm was noted in the infra-auricular region of the parotid gland, with a smooth surface and well-defined margins. No facial nerve weakness was observed. Ultrasonography and contrast-enhanced magnetic resonance imaging were suggestive of pleomorphic adenoma of the gland (Figure 7A). Fine-needle aspiration cytology was reported as a salivary gland neoplasm of uncertain malignant potential (SUMP), characterised as a cellular basaloid neoplasm (Figure 7B).

Total parotidectomy with meticulous facial nerve preservation was performed. Histopathological examination revealed a well-circumscribed, encapsulated, and cellular tumor composed of basaloid cells arranged in solid and trabecular patterns, exhibiting round nuclei and scant cytoplasm within a fibrocellular stroma. The overall features were suggestive of basal cell adenoma (Figure 8). On follow-up, the patient developed Grade II facial nerve weakness. The weakness improved with facial physiotherapy and conventional medical management, with subsequent recovery noted.

Discussion

Salivary gland tumors present a unique diagnostic challenge due to their clinical and histological diversity. Although pleomorphic adenoma (PA) is the most common benign neoplasm, accounting for nearly 60% of all salivary gland tumors,[1,4] several other tumours can closely mimic its presentation. In our case series, all four patients presented with features highly suggestive of PA but were ultimately diagnosed as adenoid cystic carcinoma (AdCC) of the sublingual and submandibular glands, myoepithelioma of the parotid gland and Basal cell adenoma of deep lobe of parotid gland on histopathology.

This clinical overlap is well-documented in the literature. Eveson and Cawson[3] observed that tumors of the submandibular and sublingual glands carry a higher likelihood of malignancy compared to the parotid. Similarly,

Seethala[5] emphasized that AdCC and other malignant tumors may present as slow-growing, painless swellings, often indistinguishable from PA preoperatively. FNAC, while valuable, has shown variable diagnostic accuracy; in an extensive review, Stewart et al. [7] reported sensitivity rates ranging from 70% to 90%, with misdiagnosis frequently occurring in differentiating PA from AdCC and myoepithelioma.

Among malignant tumours, AdCC is particularly notorious due to its indolent yet aggressive behaviour. It demonstrates a high propensity for perineural invasion and local recurrence, with long-term survival affected by late distant metastasis to the lungs and bones.[8,9] Our two cases of AdCC underscore the importance of maintaining a high index of suspicion for malignancy, particularly in tumors arising in the sublingual and submandibular glands, where the probability of malignancy may exceed 50%.[11]

Myoepithelioma, on the other hand, represents a rare benign tumor composed exclusively of myoepithelial cells. It accounts for less than 1% of all salivary gland neoplasms.[12] Clinically, it is almost indistinguishable from PA, and definitive diagnosis relies solely on histopathological and immunohistochemical evaluation. Though benign, recurrence can occur if the tumor is incompletely excised, warranting complete surgical removal with clear margins.

Basal cell adenoma (BCA) involving the deep lobe of the parotid gland is a rare pathological entity, accounting for approximately 1–3% of all salivary gland neoplasms[15]. While the majority of benign parotid tumours occur in the superficial lobe, recent retrospective data suggest that the deep lobe may be more prone to developing BCAs than previously thought; studies report that up to 37.9% of BCAs are located in the deep portion,[16] which is similar to our case. Clinically, deep lobe BCAs often present as asymptomatic, slow-growing masses that may remain undetected until they displace the lateral oropharyngeal wall or the parapharyngeal space.[17] Preoperative diagnosis via Fine Needle Aspiration Cytology (FNAC) is notoriously difficult due to the "basaloid" overlap with more aggressive malignancies like adenoid cystic carcinoma or basal cell adenocarcinoma; however, MRI findings of a well-circumscribed mass with substantial enhancement and a higher frequency of cystic degeneration (found in 45.5% of deep lobe cases) can be suggestive.[16,18] Surgical management typically necessitates a total parotidectomy or a deep lobe excision with meticulous facial nerve dissection, as the tumour's medial position requires the nerve to be fully mobilised for safe removal[19]. While the prognosis for most subtypes is excellent, the membranous variant requires long-term surveillance due to a reported recurrence rate of up to 25% and a higher risk of malignant transformation.[20]

The present case series highlights three essential points:

1. Diagnostic Dilemma – Pleomorphic adenoma remains the leading preoperative clinical suspicion, but malignant tumors (especially AdCC) may mimic its presentation.
2. Limitations of FNAC and Imaging – These tools are helpful for initial assessment but may not reliably distinguish PA from histological mimics.
3. Indispensable Role of Histopathology – Final diagnosis rests on comprehensive histopathological examination, which dictates prognosis and management.

The application of a standardised classification system enhances diagnostic accuracy and management planning. In cases 2 and 3, FNAC was categorised as MILAN system Category grade IV (SUMP), which carries an intermediate risk of malignancy of approximately 35%.[13] This justified surgical excision for definitive diagnosis.

Furthermore, grading of adenoid cystic carcinoma using the MD Anderson system provides valuable prognostic information. Case 2 demonstrated MD Anderson grade I, lacking solid components and therefore associated with comparatively favourable outcomes. In contrast, case 3 exhibited cribriform morphology with focal solid areas and perineural invasion, indicating a more aggressive tumour – at least MD Anderson grade II, with potential to be grade III if solid areas exceed 30%.

Incorporating both the MILAN and MD Anderson grading systems framework for cytologic interpretation, histopathologic stratification, and clinical decision-making, reinforcing their importance in evaluating salivary gland neoplasms.

Our findings are consistent with the literature, which emphasises the need to correlate clinical, radiological, cytological, and histopathological data to ensure accurate diagnosis and optimal treatment planning.

Conclusion

Salivary gland tumours encompass a broad spectrum of benign and malignant neoplasms, many of which can present with features mimicking pleomorphic adenoma. This case series demonstrates the diagnostic challenges posed by such mimics, including adenoid cystic carcinoma, myoepithelioma and basal cell adenoma. While clinical presentation and FNAC may suggest PA, histopathology remains the gold standard for definitive diagnosis.

Clinicians should exercise caution in assuming a diagnosis of pleomorphic adenoma, particularly in submandibular and sublingual gland swellings, where the risk of malignancy is significantly higher. A multidisciplinary approach that combines careful clinical evaluation, imaging, cytology, and histopathological confirmation is essential to ensure accurate diagnosis and guide appropriate surgical management.

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