

Parotid pleomorphic adenoma with extensive squamous metaplasia and abundant keratin pearl formation: An unusual histopathological case report

Sarita Nibhoria¹, Shilpa Bairagi², Mamta Goyal³, Pounami Debbarma³, Jasleen Kaur³, Anna Nibhoria³, Sonam Juneja³, Shaifali Tiwari³, Kiranpreet Kaur³

¹ Professor, Department of Pathology, Guru Gobind Singh Medical College and Hospital, Faridkot, Punjab, India

² Assistant Professor, Department of Pathology, Guru Gobind Singh Medical College and Hospital, Faridkot, Punjab, India

³ Junior Resident, Department of Pathology, Guru Gobind Singh Medical College and Hospital, Faridkot, Punjab, India

DOI: <https://doi.org/10.66856/ijsr.2026.8.4.8014>

Abstract

Pleomorphic adenoma is the most common benign mixed neoplasm of salivary gland and is characterized by remarkable histomorphological diversity. The neoplasm is composed of varying proportions of epithelial and myoepithelial elements embedded within a stromal background. Although foci of squamous metaplasia may be encountered, extensive squamous metaplasia associated with abundant keratin pearl formation and keratin-filled cystic spaces is unusual and may mimic malignant salivary gland neoplasms. We report a case of 37 years old male with an unusual variant of parotid pleomorphic adenoma exhibiting extensive squamous differentiation, numerous keratin pearls and keratin-filled cystic spaces.

Keywords: Keratinising Pleomorphic adenoma, Squamous metaplasia, Parotid gland

Introduction

Pleomorphic adenoma (PA) is the most common benign mixed salivary gland neoplasm accounting for approximately 60–70% of all salivary gland tumors and about upto 80% of benign salivary gland neoplasms [1, 2]. The tumor mainly involves parotid gland followed by mandibular gland and minor salivary glands with palate being the common site among the minor salivary glands accounting for approximately 50-60% [2]. Pleomorphic adenoma most commonly involves the superficial lobe of the parotid gland accounting for approximately 75% and deep lobe accounting for approximately 25% [2]. It occurs more often in females, mostly in fourth decade of life, however it can occur in other age groups like children and elderly also [1, 2]. Histopathologically, pleomorphic adenoma is characterized by marked morphological variability. The stromal component illustrates considerable heterogeneity and may exhibit myxoid, chondroid, hyalinised, fibrous, lipomatous or osseous differentiation. [2, 3, 5] Focal squamous metaplasia is usually common; however, extensive squamous differentiation with abundant keratin pearl formation and keratin-filled cystic spaces is distinctly uncommon and may mimic malignant salivary gland neoplasms like squamous cell carcinoma or mucoepidermoid carcinoma [2, 5]. Clinically, patients usually present with asymptomatic, slow growing, well-circumscribed mobile mass. [2]

Case Report

A 37-year-old male presented with chief complaint of a painless swelling in the right parotid region which had gradually increased in size over a period of one year. There was no history of infection, trauma, weight loss or prior surgery. Patient is chronic alcoholic and smoker. Clinical examination revealed a firm nodular swelling in parotid region. Ultrasonography of local area revealed solid cystic lesion of right parotid. A provisional diagnosis of a benign parotid gland neoplasm was made. Per operative findings

revealed a well-defined lesion in the superficial lobe of parotid gland which was excised and sent for histopathological examination. Gross examination revealed a right parotidectomy specimen measuring 6×4.5×1cm. Cut section demonstrated a well-circumscribed grey-white solid lesion measuring approximately 3×1.5×1cm (Figure 1).

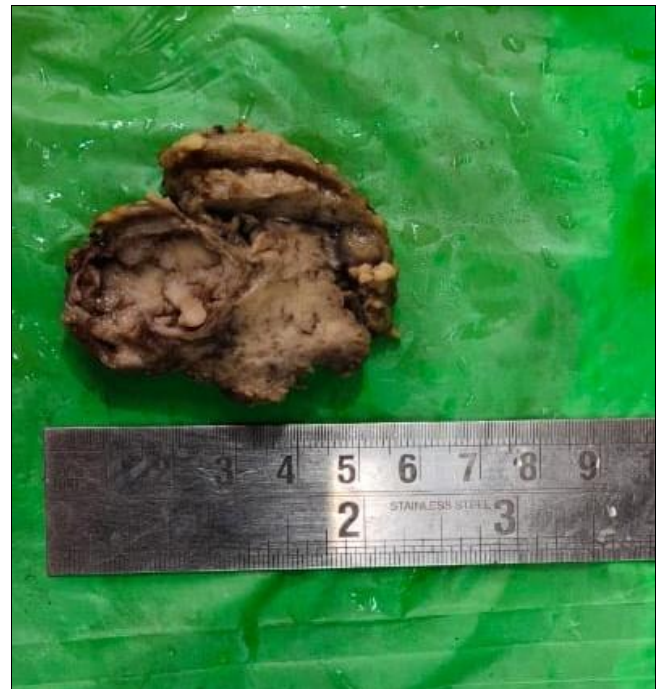


Fig 1: Gross photograph showing a well-circumscribed grey-white solid lesion.

Microscopic examination revealed a well-circumscribed salivary gland neoplasm composed of epithelial and myoepithelial cells arranged in nests, ducts, trabeculae and solid islands embedded within a fibromyxoid to hyalinized stroma. Multiple keratin-filled cystic spaces and extensive

squamous metaplasia were identified. No significant cytological atypia, necrosis or increased mitotic activity was identified (Figure 2).

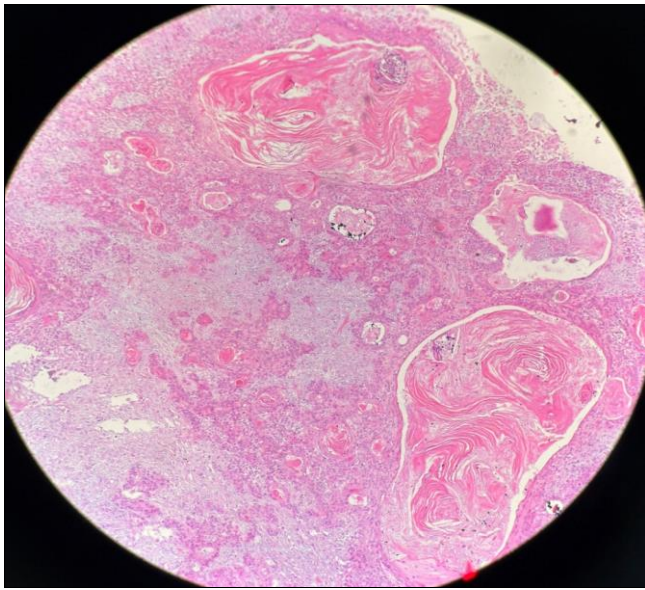


Fig 2: Hematoxylin and Eosin stained photomicrograph [10x] demonstrating extensive squamous metaplasia, keratin-filled cystic spaces and abundant keratin pearl formation within a fibromyxoid stromal background.

Discussion

Pleomorphic adenoma is characterized by marked histomorphological diversity attributable to the variable differentiation potential of epithelial and myoepithelial cells [1, 7]. The diverse morphological patterns seen in pleomorphic adenoma are attributed to the ability of neoplastic myoepithelial cells to produce abundant extracellular matrix, resulting in myxoid, chondroid, hyalinised, osseous or adipocytic stromal components. [2, 4]. Besides stromal variability, pleomorphic adenoma may also exhibit several epithelial metaplastic alterations including squamous, mucinous, sebaceous, oncocytic and clear-cell differentiation, highlighting the multidirectional differentiation potential of the tumour. [2, 4, 5].

Focal squamous metaplasia is usually common microscopic finding and is generally regarded as an incidental change without significant clinical implications. [4, 5].

In contrast, extensive squamous metaplasia accompanied by abundant keratin pearl formation and multiple keratin-filled cystic spaces is distinctly uncommon and has been described in isolated case reports and small case series. [4, 5, 6, 7].

The rarity of this histomorphological variant makes it diagnostically significant because the extensive keratinisation may obscure the classical biphasic morphology of pleomorphic adenoma and mimic a malignant salivary gland neoplasm. [4, 5, 6, 7].

There have been several case reports published describing similar morphology and highlighting the importance of diagnostic difficulty associated with extensive squamous metaplasia and keratinization.

Goulart *et al.* reported a case of minor salivary gland pleomorphic adenoma exhibiting extensive squamous metaplasia with multiple keratin-filled cystic spaces. The report concluded that the prominent squamous component could closely mimic squamous cell carcinoma or

mucoepidermoid carcinoma leading to an inaccurate diagnosis. Therefore, it is important to be aware of this possibility to distinguish it from malignant lesions and to avoid unnecessarily aggressive therapy of the patients [4].

Tandon *et al.* published a similar case report describing a keratinizing pleomorphic adenoma of hard palate with extensive squamous metaplasia and keratin-filled cystic spaces, further demonstrating that keratinization may occasionally become a dominant component of an otherwise benign pleomorphic adenoma closely mimicking the malignancy resulting in potential diagnostic pitfall [5].

Sankari *et al.* also reported a case of keratinizing pleomorphic adenoma as a rare variant in an unusual location (left cheek) highlighting the importance to recognize the underlying architecture of pleomorphic adenoma before interpreting the prominent squamous component as malignancy [8]. The present case demonstrates a similar morphological pattern, with extensive squamous differentiation and numerous keratin pearls within a lesion that retains epithelial and myoepithelial elements embedded in fibromyxoid to hyalinized stroma.

Recently Hosur *et al.* reported two cases of pleomorphic adenoma with extensive squamous metaplasia and keratinizing cysts, specifically emphasizing the diagnostic and clinical pitfalls associated with this morphological pattern [6]. This observation closely resembles the present case, in which the diagnosis was supported by the presence of a well-circumscribed lesion, epithelial and myoepithelial elements, fibromyxoid to hyalinized stroma and absence of significant cytological atypia, necrosis or increased mitotic activity.

Singh *et al.* reported a case of pleomorphic adenoma with extensive squamous and adipocytic metaplasia that mimicked low-grade mucoepidermoid carcinoma on aspiration cytology. Histomorphologically, the lesion exhibit closely packed glands with a double epithelial layer, chondromyxoid stroma, keratin pearl formation, squamous metaplasia and adipocytic metaplasia, without mitosis or necrosis [13]. This report is relevant to the present case as it illustrates how extensive squamous metaplasia may produce a misleading cytological or histological impression of mucoepidermoid carcinoma.

Varsha *et al.* have reported a case of keratinizing pleomorphic adenoma of the buccal mucosa, exhibiting extensive keratinization as an unusual histopathological presentation [7]. The report further supports the occurrence of this uncommon metaplastic variant in minor salivary glands and emphasizes the potential for extensive keratinization to become a dominant histological feature.

Gaskin *et al.* reported a case of 49-year-old man with a submandibular gland pleomorphic adenoma exhibiting extensive squamous metaplasia. Histopathological examination showed pleomorphic adenoma with extensive squamous metaplasia, highlighting the diagnostic pitfall created by this unusual variant [10]. This case report is particularly relevant to the present case report supporting the occurrence of extensive squamous metaplasia in both major and minor salivary glands.

Li *et al.* reported a case series and literature review of pleomorphic adenoma with squamous metaplasia, emphasizing the diagnostic pitfalls that may arise on FNAC when there is prominent squamous component [9]. Their observations strengthen the importance of correlating cytological findings with the overall histomorphology and

identifying the characteristic epithelial, myoepithelial and stromal components of pleomorphic adenoma.

Donohoe *et al.* described a case of pleomorphic adenoma with squamous metaplasia that resembled metastatic oral squamous cell carcinoma ^[11]. This report emphasizes that extensive squamous metaplasia may closely mimic primary squamous malignancy or metastasis and that architectural assessment is necessary for correct diagnosis.

Lin and Chiang recently reported a case that showed pleomorphic adenoma with extensive squamous metaplasia and lipomatous change while conventional tubular or glandular structures and myoepithelial components were retained within the stromal background. This further highlights the ongoing recognition of this rare morphological pattern. The benign nature of the lesion was further supported by the absence of significant cytological atypia and the overall histomorphological and immunohistochemical findings ^[12].

The principal differential diagnosis is squamous cell carcinoma of the salivary gland ^[7]. Although both lesions may demonstrate extensive keratinization and keratin pearl formation, squamous cell carcinoma typically exhibits infiltrative growth, marked cytological atypia, increased mitotic activity and tissue destruction ^[7]. These features were absent in the present case.

Second important differential is Mucoepidermoid carcinoma, particularly the low-grade variant because both lesions may demonstrate squamoid epithelial differentiation and cystic architecture ^[7]. However, the 'biphasic architecture' - the presence of characteristic epithelial-myoeplithelial architecture within a fibromyxoid stroma and absence of mucous cells strongly favored pleomorphic adenoma ^[2, 7].

Carcinoma ex pleomorphic adenoma was excluded because evidence of extracapsular invasion, significant pleomorphism, increased mitotic activity or necrosis was not identified ^[7].

Infarcted or metaplastic pleomorphic adenoma may exhibit squamous metaplasia, however infarction-related necrosis was not observed ^[2, 4, 7].

Warthin tumour with squamous metaplasia was also considered; however, because there was no oncocytic epithelium and lymphoid stroma this possibility was excluded ^[7].

Recognition of this rare variant is essential because its prominent keratinizing component can mimic a malignant neoplasm of salivary gland thus potentially resulting in aggressive treatment ^[2, 6].

Conclusion

Pleomorphic adenoma with extensive squamous metaplasia and abundant keratin pearl formation is a rare histological variant that may closely mimic malignant salivary gland neoplasms. Recognition of the characteristic biphasic architecture, supportive stromal features and absence of malignant cytological findings is essential for accurate diagnosis and appropriate patient management thus preventing aggressive treatment.

References

1. Speight PM, Barrett AW. Salivary gland tumours. *Oral Dis*,2002;8(5):229-40.
2. Goldblum JR, Lamps LW, McKenney JK, Myers JL, editors. *Rosai and Ackerman's Surgical Pathology*. 11th ed. Philadelphia (PA): Elsevier, 2018.

3. WHO Classification of Head and Neck Tumours. 5th ed. Lyon: International Agency for Research on Cancer, 2022.
4. Goulart MCV, Freitas-Faria P, Goulart GR, Oliveira AM, Carlos-Bregni R, Soares CT, *et al.* Pleomorphic adenoma with extensive squamous metaplasia and keratin cyst formations in minor salivary gland: a case report. *J Appl Oral Sci*,2011;19(2):182-8.
5. Tandon A, Jaiswal R, Siddiqui S, Bordoloi B. Keratinizing pleomorphic adenoma: an unusual case report. *J Oral Maxillofac Pathol*,2018;22(1):69-72.
6. Hosur MB, Puranik RS, Dandagi SG, Patil VM. Pleomorphic adenoma with extensive squamous metaplasia and keratinizing cysts: diagnostic and clinical pitfalls – a report of two cases and review of literature. *J Oral Maxillofac Pathol*,2024;28(4):689-93.
7. Sharma S, Mehendiratta M, Chaudhary N, Gupta V, Kohli M, Arora A. Squamous metaplasia in pleomorphic adenoma: a diagnostic and prognostic enigma. *J Pathol Transl Med*,2018;52(6):411-5.
8. Sankari SL, Dhanavelu P, Sangeetha Priya P, Jimson S. Keratinizing pleomorphic adenoma: a rare variant in an unusual location. *J Oral Maxillofac Pathol*,2021;25(3):557.
9. Li JJX, Ng JKM, Lau EHL, Chan ABW. Aspiration cytology of pleomorphic adenoma with squamous metaplasia: a case series and literature review illustrating diagnostic challenges. *Diagn Cytopathol*,2022;50(2):64-74.
10. Gaskin DA, Reid A, Gaskin P. Pleomorphic adenoma with extensive squamous metaplasia: the first well-documented case involving the submandibular gland. *Hum Pathol Rep*,2022;27:300600.
11. Donohoe E, Courtney R, Phelan S, McCann PJ. A case report of pleomorphic adenoma squamous metaplasia resembling metastatic oral squamous cell carcinoma. *Adv Oral Maxillofac Surg*,2021;2:100074.
12. Lin HP, Chiang CP. Pleomorphic adenoma with extensive squamous metaplasia - a case report. *J Dent Sci*,2026;21(1):668-70.
13. Singh A, Phulware RH, Ahuja A, Gupta A, Kaushal M. Pleomorphic adenoma with extensive squamous and adipocytic metaplasia mimicking as low-grade mucoepidermoid carcinoma on FNAC. *Indian J Otolaryngol Head Neck Surg*,2022;74(2):2132-5.