

CASE REPORT

Cytodiagnosis of Pleomorphic Adenoma of Submandibular Gland with Cystic Degeneration: A Common Lesion with an Uncommon Presentation - A Case Report

Pritam S Khairkar¹, Sarika More², Ashish Pandey³, Megha Pandey⁴, Sawke GK⁵

1,2- Associate Professor (Pathology), Chirayu Medical College and Hospital, Bhopal, India

3,4- Assistant Professor (Pathology), Chirayu Medical College and Hospital, Bhopal, India

5- Professor (Pathology), Chirayu Medical College and Hospital, Bhopal, India

Abstract

Pleomorphic adenoma (PA) is the most common tumor to arise in the parotid gland but can be seen in other salivary glands also. Generally the tumor is solid and easily diagnosed but cystic degeneration can pose a diagnostic challenge. Such cystic degeneration in Pleomorphic adenoma can lead to erroneous decisions concerning diagnosis and treatment. We present a similar rare case of pleomorphic adenoma of the submandibular gland with cystic degeneration, along with the cytohistopathological findings and discussion of some pitfalls in the diagnosis.

Key Words: Adenoma, Cystic degeneration, Pleomorphic adenoma, Submandibular gland neoplasms

Key messages: PA is usually solid but can rarely undergo cystic degeneration. Cytological diagnosis may be difficult in such cases. Careful attention to aspiration of any solid nodule should be paid which will clinch a correct diagnosis.

Address for correspondence: Dr. Pritam S Khairkar. Associate Professor (Pathology), Chirayu Medical College and Hospital, Bhopal, Indore Highway, Bhopal. M.P. India. Mobile No.+91 7738247822 & +91 8806119469. Email: dr_pritam2006@yahoo.co.in, dr.sarikamore@rediffmail.com, ashishpandey_789@yahoo.com, drmeghashukla@yahoo.com

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Introduction

Pleomorphic adenoma (PA) is the tumor that commonly arises in the parotid gland. It can also be seen in submandibular and other minor salivary glands. Generally the tumor is totally solid but can rarely undergo cystic degeneration imposing diagnostic challenges^[1]. We present a case of pleomorphic adenoma of submandibular gland with cystic degeneration and discuss its clinical, cytological and histopathological features.

Case Report

A 45 year old male presented with left submandibular oval mass since 15 yrs. On examination, the mass measured about 5.5 x 4 cms and was solid in the anterior part and cystic in the posterior part. The patient was subjected for FNA. The cystic part of the mass yielded

0.5ml turbid fluid while the solid part yielded a small amount of mucoid aspirate.

The aspirate from solid part showed epithelial cells arranged in sheets, clusters and at places in acinar & irregular branching patterns. The cells had a bland round to oval nuclei with moderate amount of cytoplasm. Background showed plenty of individual scattered cells with plasmacytoid appearance. Fragments of fibrillary chondromyxoid material were seen interspersed between the cell clusters. Spindle cells were also present in the fragments of chondromyxoid material and the epithelial cell sheets (Fig.1B).

Smears from the cystic part showed few small clusters of macrophages, epithelial cells and lymphocytes against a necrotic background (Fig 1A).

Based on the cytological findings a diagnosis of Pleomorphic adenoma with cystic degeneration was made and excision of the mass was advised.

The mass was excised and sent for histopathological examination.

Figure- 1(A): FNA smear from cystic part showing few small clusters of macrophages, epithelial cells & lymphocytes.

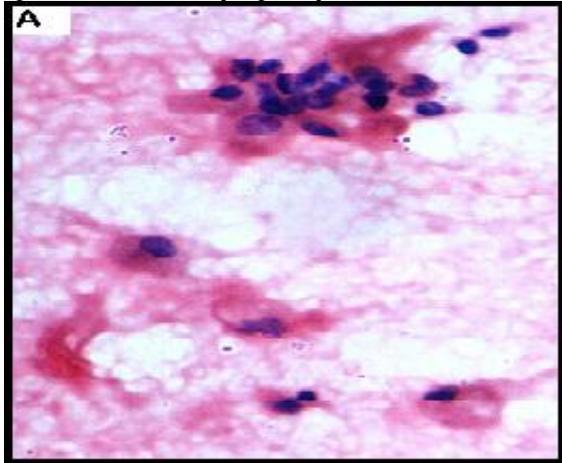
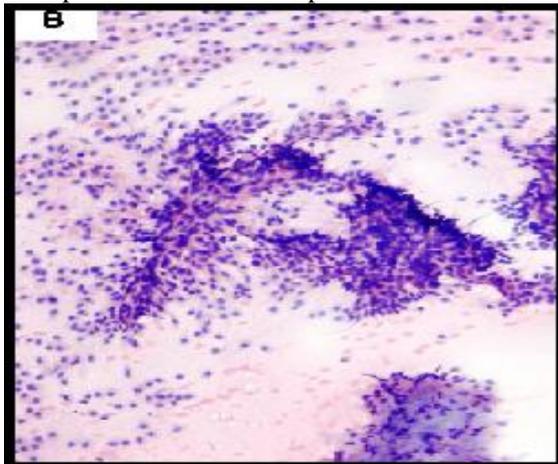


Figure- 1(B): FNA smear from solid part showing fibrillary chondromyxoid material interspersed between the epithelial cell sheets.



On gross examination, the mass measured 5 x 4 x 2.5 cms. Cut section showed solid and cystic areas. The solid part was grey white, firm in consistency with tiny myxoid areas and the cystic part contained necrotic material. The wall of the cystic part was grey white, firm and measured 3mm in thickness. Normal salivary gland tissue was seen attached to the mass (Figure-2).

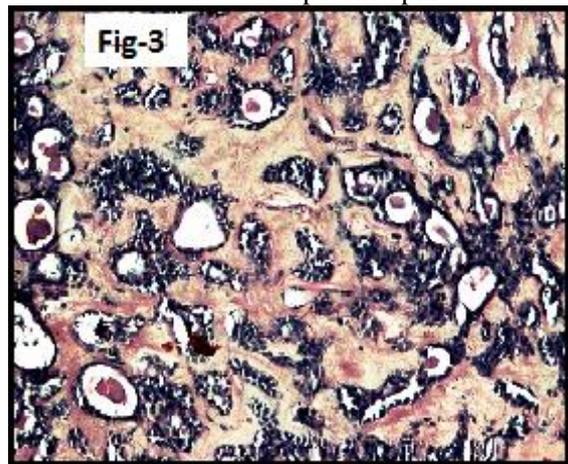
The histopathological examination of the solid part revealed a tumor showing classical features of pleomorphic adenoma. Sections from the cyst wall were highly cellular and showed few tubules along with chondromyxoid areas suggestive of pleomorphic adenoma. (Figure-3).

The center of the cystic area showed necrotic material & hemorrhage.

Figure- 2: Showing the solid whitish part and the cystic part with necrotic material



Figure- 3: Section from solid part showing classical features of pleomorphic adenoma



Discussion

A wide variety of neoplastic and non-neoplastic lesions of the salivary glands may be partially or completely cystic. Neoplasms which commonly present with a cystic component include Warthin's tumor, mucoepidermoid carcinoma and mucinous adenocarcinoma^[2]. Cystic change in PA is very rare.

The cytodiagnosis of PA is not difficult in typical cases^[3]. However gross cystic degeneration of PA compounds diagnostic problem. Such a tumor cannot be distinguished from a mucous cyst, basal cell adenoma and low grade mucoepidermoid carcinoma, if it completely lacks the features of a solid tumor^[2].

After aspiration of cyst contents careful attention should be paid if any solid part or nodule is palpable and should be aspirated^[4]. Multiple sampling usually provides some diagnostic tissue fragments. The aspiration of mucoid fluid from cystic part may suggest mucous cyst because of sparse cellularity in the aspirate^[3].

Epithelial metaplasia is common in PA and goblet cells can be seen sometimes. If a PA with cystic degeneration shows these features with background of mucoid material a false diagnosis of mucoepidermoid carcinoma (MEC) can be made^[3,5]. Low grade mucoepidermoid carcinoma can be cystic and may show squamous metaplasia. Clues to the diagnosis of the same will be provided by mucoid aspirate with mucoid background and mucus cells with cytoplasmic vacuoles.^[6] Identification of three types of cells: intermediate, mucus-producing, and squamous cells in the smear are most predictive of mucoepidermoid carcinoma. Rupani et al suggested the importance of stringy mucin as the stringy nature helps to differentiate it from mucin seen in pleomorphic adenoma.^[7] Cohen et al in their study had emphasized on the overlapping epithelial groups in MEC.^[8] Batrani et al suggested a close scrutiny for fragments of chondromyxoid stroma, since in its absence and with the presence of squamous, mucinous or sebaceous metaplasia, which can be seen in PA, the lesion may be misinterpreted as mucoepidermoid carcinoma on cytology.^[9] Therefore if any solid nodule is present after cyst aspiration, it should always be aspirated because the diagnostic material is present in the solid portion of the tumor^[3]. In the present case the diagnosis of PA was possible only from the aspirates from the solid nodule.

Conclusion

Pleomorphic adenoma, a common lesion of salivary gland can rarely present with cystic degeneration. Therefore, a cystic lesion should be completely aspirated and a thorough search and aspiration from the solid part/nodule should be carried out in order to obtain appropriate diagnostic tissue material. This would not only help the cytopathologist in coming to a correct diagnosis but at the same time will also prevent in missing out the lesion and making a false

positive diagnosis of the conditions with cystic degeneration.

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