

PLEOMORPHIC ADENOMA OF MINOR SALIVARY GLANDS OF FACE AND ORAL CAVITY: A STUDY OF 34 CASES

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ABSTRACT

BACKGROUND

Pleomorphic adenoma of minor salivary glands is extremely rare (especially the sites other than aerodigestive tract) and very few isolated cases are reported, located in the larynx, pharynx, trachea, lacrimal glands and sinonasal tract. Here, we have carried out a study of 34 cases over a period of about 11 years where pleomorphic adenomas of minor salivary glands were noted mostly in sites other than aerodigestive tract such as cheek (7 cases), lips (9 cases), external area of nose (5 cases). Twelve cases were observed in soft palate and 1 case in parapharyngeal space.

The aim is to study the incidence and clinicopathological aspects of this rare entity.

MATERIALS AND METHODS

Total 34 cases of pleomorphic adenoma of minor salivary glands of face and oral cavity were studied during 11 years of period from January 2006 to November 2016. Age of the patients ranged from 17 years to 58 years. Twenty one cases were male and 13 were female. After clinical history taking, FNAC and histopathological examination of affected tissue were done except in parapharyngeal mass where only histopathological examination was performed.

RESULTS

Age of the patients ranged from 17 years to 58 years with a peak in fourth decade, having male predominance. On both FNAC and histopathological examination, the diagnosis of pleomorphic adenoma was confirmed. However, few histopathological features are seen in variable amount in different cases.

CONCLUSION

Pleomorphic adenoma of minor salivary glands is a very rare entity. We have encountered majority cases (21) of pleomorphic adenoma of minor salivary glands arising from other than aerodigestive tract out of total 34 cases in contrast to extremely rare incidence in these sites in other studies. So it can be regarded as a significant series for study.

KEYWORDS

Pleomorphic Adenoma, Minor Salivary Glands, FNAC, Histopathological Examination.

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BACKGROUND

Pleomorphic adenoma, the most common benign tumour of major salivary glands such as parotid and submandibular glands is also known as benign mixed tumour due to presence of both epithelial and stromal components in FNAC as well as histopathology. However, pleomorphic adenoma of minor salivary glands is a rare entity. Only 7% of pleomorphic adenoma occurs in minor salivary glands in comparison to 93% in major salivary glands.^{1,2} Pleomorphic adenomas of minor salivary glands are seen in upper aerodigestive tract such as nasal cavity (including nasal septum and lateral nasal wall), pharynx, larynx, trachea, lacrimal glands, hard and soft palate and parapharyngeal space.^{1,3,4,5,6}

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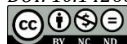
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However, it is extremely rare in the sites other than aerodigestive tract. As for examples, they can occur very rarely over skin, scalp, eyelid, external nose, cheek, lips, external ear and external auditory canal.^{1,5,7}

Here, we are presenting the cases of pleomorphic adenomas of minor salivary glands where highest number of cases is seen in soft palate (12 cases out of 34) but majority of cases are from sites other than aerodigestive tract (21 cases out of total 34). However, only one case is noted in parapharyngeal space.

Aims and objectives

- To study the incidence of pleomorphic adenoma of minor salivary glands of face and oral cavity.
- To study the clinicopathological aspects along with its variation of this rare entity.

MATERIALS AND METHODS

Total 34 cases of pleomorphic adenoma of minor salivary glands were studied during 11 years period from January, 2006 to November, 2016. All patients were from ENT Department of the institute. Age of the patients ranged from 17 years to 58 years. 21 cases were male, 13 cases were female.

After clinical history taking and relevant investigations of the patients, Fine Needle Aspiration Cytology (FNAC) was performed in all cases except in parapharyngeal space. In 33 cases, surgical removal of masses was done successfully after taking all appropriate preoperative management. The specimens were sent for histopathological examination. The remaining one case of parapharyngeal mass was detected by CT scan and subsequently surgical removal of mass was done in proper procedure. The surgically removed parapharyngeal mass was also sent for histopathological examination.

FNAC slides were stained with MGG (May Grunwald Giemsa) and Pap (Papanicolaou) stains. Staining of tissue sections from all masses were done by Haematoxylin and Eosin (H&E) stain. Special stains like PAS and Mucicarmine were carried out wherever required.

FNAC slides and histopathological sections were studied and corroborated in details to confirm the diagnosis.

Observations and results

Most of the patients were of young adult age group with a peak in fourth decade. The age of the patients ranged from 17 years to 58 years. (Table – 1, Chart – 1)

Age group (in years)	11 - 20	21 - 30	31 - 40	41 - 50	Above 50	Total	%
Male	2	3	13	2	1	21	61.76
Female	2	4	5	1	1	13	38.24
Total	4	7	18	3	2	34	

Table 1. Age and sex distribution of 34 cases of pleomorphic adenoma of minor salivary glands of face and oral cavity

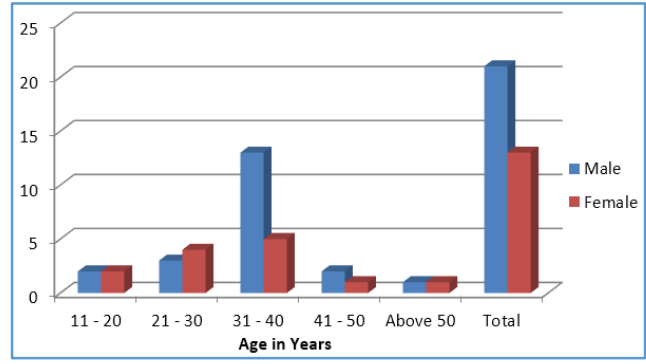


Chart 1. Age and sex distribution of 34 cases of pleomorphic adenoma of minor salivary glands of face and oral cavity.

All the patients presented with slow growing palpable lobulated mass with a history of 1 to 7 years. Though the singly highest incidence of pleomorphic adenoma of minor salivary glands was noted in soft palate (Figure-1) in our study (12 cases out of 34), most cases (21 cases out of 34) were in sites other than aerodigestive tract. Out of which 7 cases were noted in cheek (Figure-2), nine cases in lips (Figure – 3, 4, 5 & 6) and five cases over ala of the nose (Figure-7). Only 1 case was seen in parapharyngeal space (Figure-8 & 9). The Table-2 showed the distribution of sites of 34 cases of pleomorphic adenoma of minor salivary glands of face and oral cavity.



Figure 1. Pleomorphic adenoma of soft palate



Figure 2. Pleomorphic adenoma of cheek.

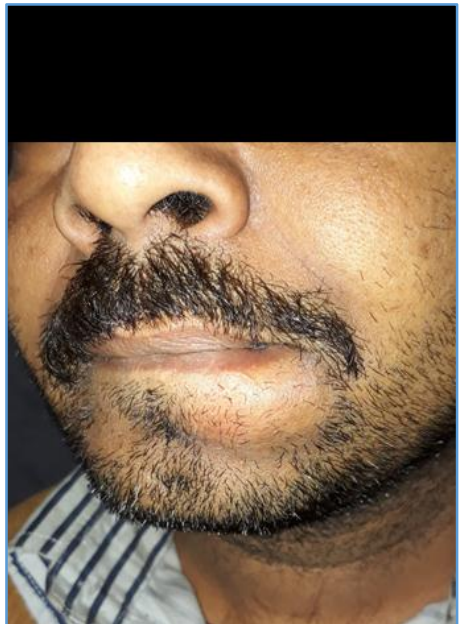


Figure 3. Pleomorphic adenoma of lower lip



Figure 4. FNAC of lower lip mass (Diagnosed as Pleomorphic Adenoma)



Figure 7. Pleomorphic adenoma of right ala of the nose



Figure 5. Pleomorphic adenoma of upper lip

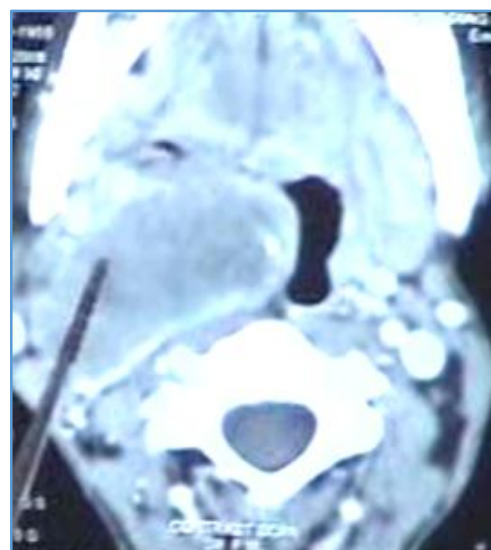


Figure 8. C T Scan of parapharyngeal mass



Figure 6. Per-operative gross photograph of pleomorphic adenoma of upper lip with glistening cut surface having mucoid material



Figure 9. Per-operative gross photograph of parapharyngeal mass (diagnosed as pleomorphic adenoma on histopathological examination)

Sites	No. of Cases	%
A. Sites other than aerodigestive tract	21	61.76
a. Cheek	7	20.59
i. Right	2	5.88
ii. Left	5	14.71
b. Lips	9	26.47
i. Upper	6	17.65
ii. Lower	3	8.82
c. External nose	5	14.71
i. Right ala of the nose	4	11.76
ii. Left ala of the nose	1	2.94
B. Sites within aerodigestive tract	13	38.24
a. Soft palate	12	35.29
b. Parapharyngeal space	1	2.94

Table 2. Distribution of sites of 34 cases of pleomorphic adenoma of minor salivary glands of face and oral cavity

On physical examination, no abnormalities in major salivary glands were noted. There were no cervical lymphadenopathy or other physical problems. FNAC of all 33 patients yielded more or less gelatinous material, smears from which showed discrete as well as poorly cohesive clusters and sheets of epithelial cells, embedded within pinkish or purple chondromyxoid stromal substance. The epithelial cells showed thin rim of pale blue cytoplasm and round to oval mostly uniform nuclei. The spindle shaped mesenchymal cells were seen in stromal matrix at places. In every cases, the diagnosis of pleomorphic adenoma was given on FNAC. (Figure – 10,11,12,13 & 14).

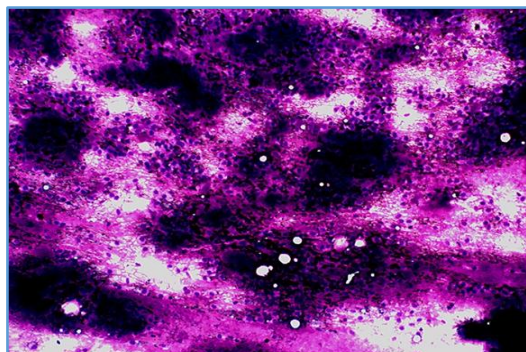


Figure 10. Microphotograph of FNAC smear of pleomorphic adenoma shows clusters of epithelial cell embedded within myxoid stroma (MGG x100)

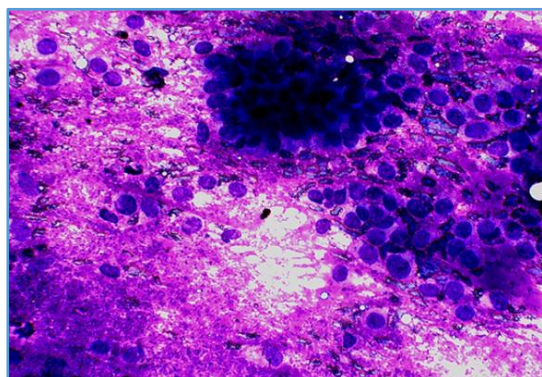


Figure 11. Microphotograph of FNAC smear of pleomorphic adenoma shows discrete as well as loose cohesive clusters of round to oval epithelial cells embedded within myxoid stroma (MGG x400)

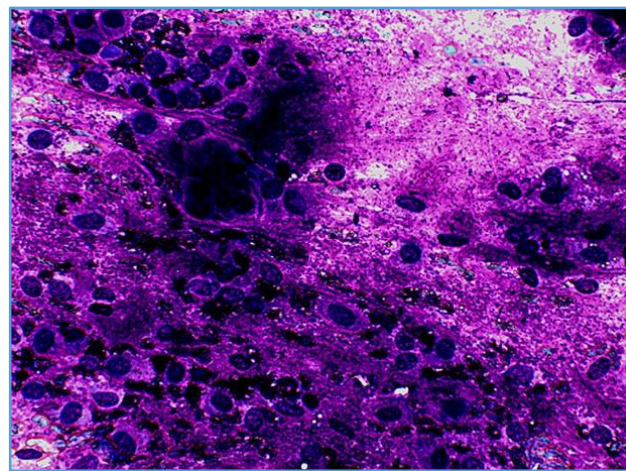


Figure 12. Microphotograph of FNAC smear of pleomorphic adenoma shows discrete epithelial cells submerged within chondromyxoid stroma (MGG x400)

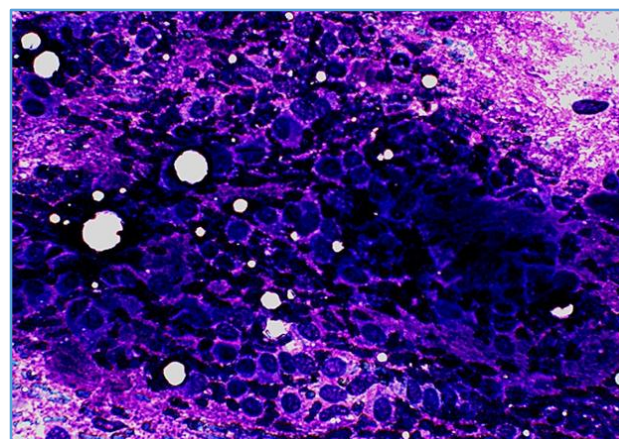


Figure 13. Microphotograph of FNAC smear of pleomorphic adenoma shows thin rim of bluish cytoplasm and round to oval nuclei of epithelial cells. Background is chondromyxoid. (MGG x400)

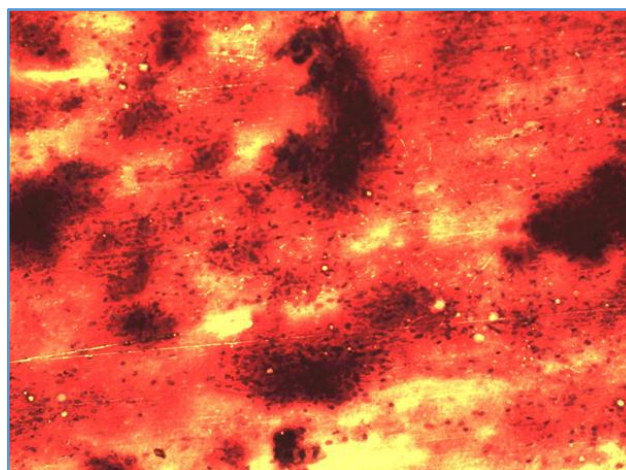


Figure 14. Microphotograph of FNAC smear of pleomorphic adenoma shows clusters of epithelial cells and spindle myoepithelial cells embedded within pinkish stromal substance (MGG x400)

Gross (Macroscopic) Examination of Specimens Revealed-

- The size of the masses ranged from 1.5 cm (from ala of the nose) to 5.5 cm in diameter (from upper lip).
- All the masses were well circumscribed having thin capsule except those in soft palate. There were no definite capsules over the masses from soft palate.
- Masses were lobulated in shape. Cut surfaces showed tan colour with focal brownish areas and appeared fleshy and rubbery. In few cases, mucinous or glistening areas were seen.

On histopathological examination, the sections showed in all cases, epithelial cells were arranged in anastomosing tubules, small cysts, cords and solid sheets, submerged in chondromyxoid stroma. The epithelial cells varied from columnar, cuboidal to flat. The epithelial cells were layered by myoepithelial cells at places which appeared as spindle, stellate and epithelioid cells. The duct lumens were mostly open. However, in few areas, lumens were filled with eosinophilic colloid like material. The diagnosis of pleomorphic adenoma was given beyond reasonable doubt on the basis of histopathological features. (Figure - 15, 16, 17, 18, 19 & 20).

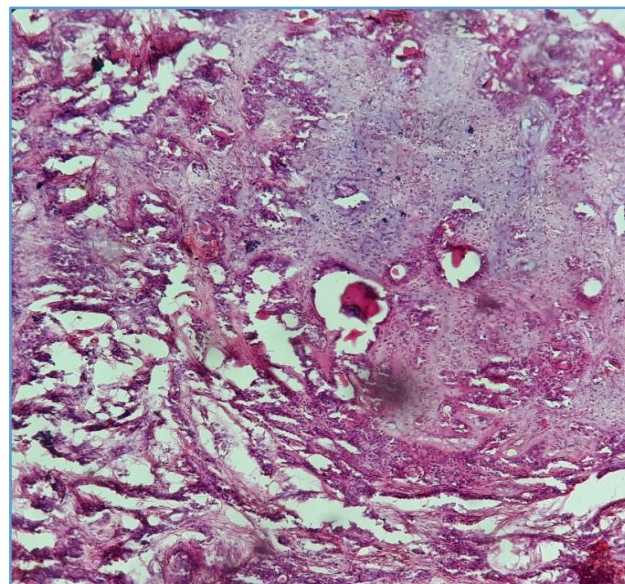


Figure 17. Microphotograph of histopathological examination of pleomorphic adenoma shows eosinophilic colloid like material in the duct lumen (H&E x100).

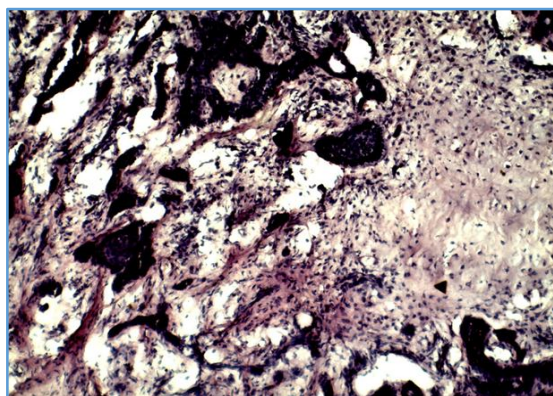


Figure 15. Microphotograph of histopathological examination of pleomorphic adenoma shows anastomosing tubules and cord like arrangement of epithelial cells in the background of chondromyxoid stroma (H&E x100)

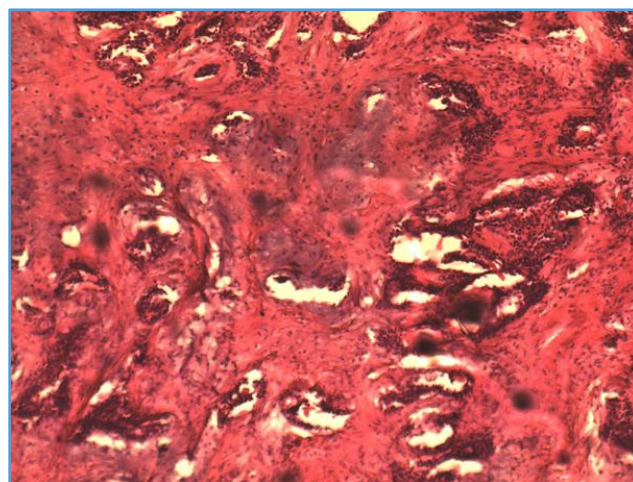


Figure 18. Microphotograph of classical features of pleomorphic adenoma on histopathology (H&E x100)

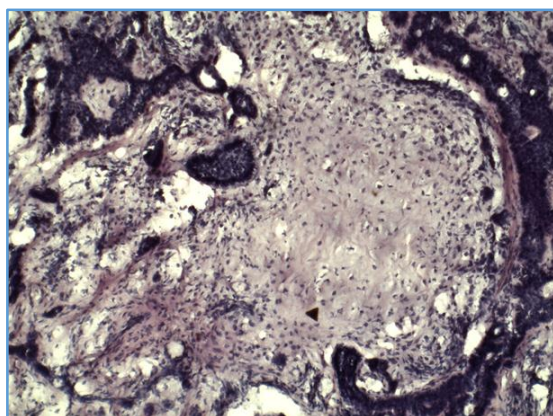


Figure 16. Microphotograph of histopathological examination of pleomorphic adenoma shows epithelial and myoepithelial cells in the background of chondromyxoid stroma (H&E x100)

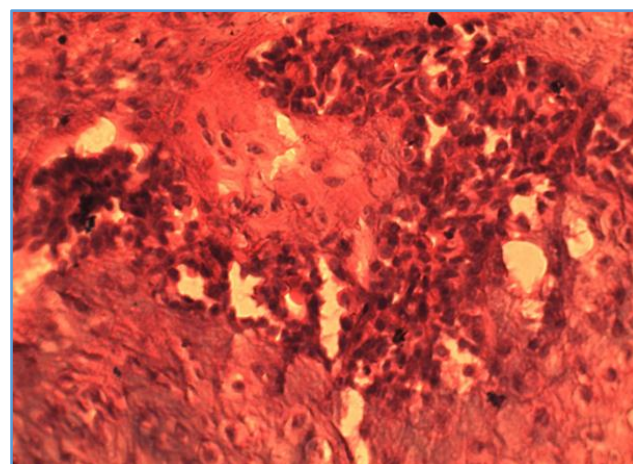


Figure 19. Microphotograph of high power view of epithelial cells in the background of chondromyxoid stroma in a case of pleomorphic adenoma on histopathological examination (H&E x400)

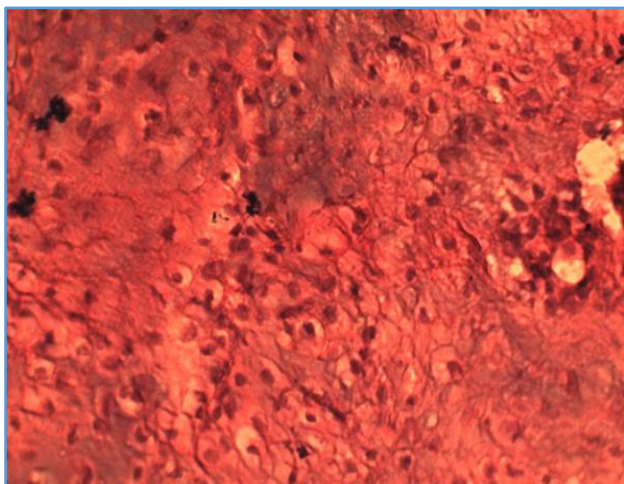


Figure 20. Microphotograph of high power view of chondromyxoid stroma in a case of pleomorphic adenoma on histopathological examination (H&E x400)

The diagnosis of pleomorphic adenoma on FNAC was corroborated by histopathological examination in all 33 cases. In 1 case, located in parapharyngeal space where only histopathological examination of surgically removed specimen was done and the same diagnosis of pleomorphic adenoma was given.

DISCUSSION

The present study shows that the incidence of pleomorphic adenoma of minor salivary glands of face and oral cavity is very low. Only 34 cases were noted over a period of about 11 years.

Age incidence is highest in fourth decade which is similar to other studies whereas regarding sex incidence male preponderance is noted in place of female of other studies.^{8,9} The incidence of pleomorphic adenoma of minor salivary glands in sites other than aerodigestive tract is extremely rare.^{1,10} However, in our study, the majority (21 out of 34) of the cases were reported from the sites other than aerodigestive tract. This finding is contrast to that of other study.

Pleomorphic adenoma of minor salivary gland is most common in palate, followed by lip.^{11,12} Similar findings are also noted in our series where highest (12) numbers of cases reported from palate, followed by nine cases in lips out of total 34 cases. However, in our study, all palatal pleomorphic adenomas are from soft palate only. So, it is a notable feature of our study.

There is record of only 126 cases of pleomorphic adenoma affecting minor salivary glands of cheek^{13,14}. In our study, there are only 7 cases of pleomorphic adenoma diagnosed from the masses of cheek over a period of about 11 years.

As per nasal involvement, pleomorphic adenoma is more common in nasal cavity than external nose.^{1,10} However, in our series, all the five cases were located in external nose (ala of the nose).

According to literatures, primary pleomorphic adenomas of parapharyngeal space may occur probably from displaced

or aberrant salivary gland tissue within a lymph node. An extensive study of literatures show only very few case reports of pleomorphic adenomas arising de novo in parapharyngeal space.^{6,15,16} We have encountered only one case of pleomorphic adenoma in parapharyngeal space.

Cytopathological features on FNAC smears of 33 patients out of total 34 cases were highly diagnostic of pleomorphic adenoma without any variation. In one case of parapharyngeal space only histopathological examination of surgically removed mass was performed and histopathological diagnosis of pleomorphic adenoma was given. Hence, there is 100% corroboration of FNAC and histopathological diagnosis.

CONCLUSION

Pleomorphic adenoma of minor salivary glands is a very rare entity with a peak incidence in fourth decade of life. In contrast to other study, male predominance is noted in our series. We have encountered majority cases (21) of pleomorphic adenoma of minor salivary glands arising from other than aerodigestive tract out of total 34 cases in contrast to extremely rare incidence in these sites in other studies. So it can be regarded as a significant series for study.

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