Case Report

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Giant Pleomorphic Adenoma of Parotid Gland: A Rare Cytological Diagnosis

Pathology Section

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ABSTRACT

Pleomorphic adenoma is the most common salivary gland neoplasm, which affects both major and minor salivary glands. It is also the most common neoplasm arising in the parotid. In minor salivary glands, it most commonly involves the palate and rarely involves the lips, cheek and gingiva. It is also called Benign Mixed Tumour (BMT), since it is composed of a variable amount of epithelial and mesenchymal elements. Diagnostic modalities include Ultrasonography (USG), Computed Tomography (CT) and Magnetic Resonance Imaging (MRI). MRI provided the most precise structural information of the tumour as well as surrounding structures. A 60-year-old female presented with a massive right-sided infra-auricular swelling, since 20 years. On Contrast Enhanced Computed Tomography (CECT), a huge lobular exophytic lesion with thin enhancing septation and few calcific foci was visualised in the right parotid region. The Fine Needle Aspiration Cytology (FNAC) yielded the diagnosis of Pleomorphic Adenoma (PA). Giant PA with an indolent course is a rare entity in itself. This is a case of giant PA of the parotid gland growing insidiously over a period of 20 years.

Keywords: Benign, Infra-auricular, Salivary gland

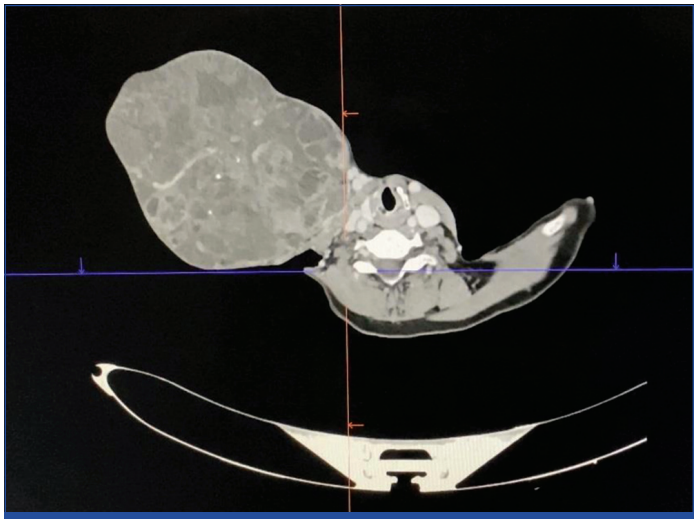
CASE REPORT

A 60-year-old female presented in the Outpatient Department (OPD) of Ear, Nose and Throat (ENT) with a right-sided infra-auricular mass, which was gradually increasing in size. It was not associated with any pain or local rise of temperature. Patient first noticed the swelling 20 years back and took homeopathic treatment for the same. There was no history of previous examination and diagnosis. There was no other significant medical and surgical history. She did not report of similar condition in any of the family members.

On clinical examination, a large bosselated swelling was present in the right infra-auricular region. It was firm to hard, non tender and measured 30 cm superoinferiorly and 28 cm anteroposteriorly [Table/Fig-1]. Mobility of the lesion was limited. The overlying skin and right ear lobe were stretched. No palpable cervical lymph nodes or signs of facial nerve deficits were noted. Oral examination showed unremarkable features. As the swelling was present from 20 years, a provisional diagnosis of benign salivary gland neoplasm was considered. A differential diagnosis of pleomorphic adenoma and myoepithelioma was considered.

[Table/Fig-1]: An oval bosselated mass measuring 30×28×20 cm in right parotid region.

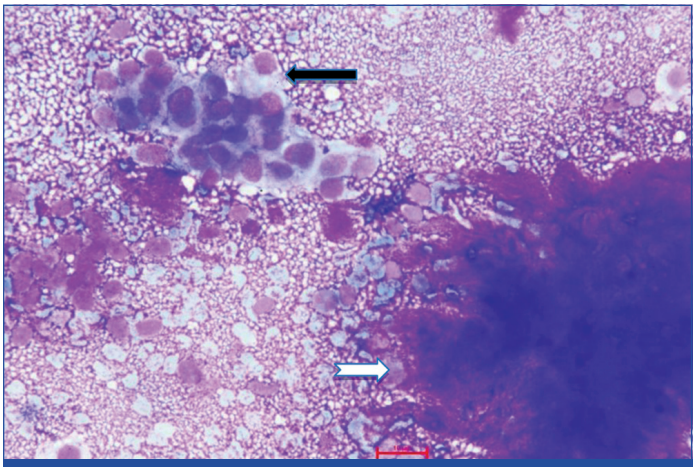
The CECT of the mass showed a large lobular exophytic lesion of size 30×28×20 cm with thin enhancing septation and few calcific foci (along posteroinferior aspect) in the right parotid gland region [Table/Fig-2]. The right parotid gland could not be observed separately from the lesion. Medially the mass was extending in the submandibular space displacing the right submandibular gland anteriorly, growing into the masticator space compressing the parapharyngeal fat medially with narrowing of pharyngeal lumen causing compression and displacement of pterygoid muscles anteriorly. The lesion was also compressing the right internal jugular vein, displacing the internal carotid artery posteriorly. The external carotid artery branches were observed traversing the lesion. It was displacing the ramus of the right mandible laterally with mild anterior displacement of the temporomandibular joint. Posteroinferiorly, it was compressing the right sternocleidomastoid muscle.



[Table/Fig-2]: The CECT (coronal plane) showing a large lobular exophytic lesion of size 30×28×20 cm with thin enhancing septation and few calcific foci in the right parotid region.

The FNAC was advised for cytological diagnosis. Under aseptic conditions multiple passes were taken from different parts of the lesion using 22 gauge needle. Smears were cellular and showed singly scattered as well as clusters of round to polygonal cells having bland nuclear features and abundant pale eosinophilic cytoplasm

in a background of extensive chondromyxoid ground substance [Table/Fig-3]. Features of cellular atypia were not observed. A final diagnosis of PA was given involving the whole parotid. Based on the diagnosis, complete surgical excision (total parotidectomy with preservation of the facial nerve) of the lesion was performed in the ENT Department and the patient was relieved of her symptoms. Patient was followed-up for six months postsurgery and no recurrence is reported as of now.



[Table/Fig-3]: Round to oval cells with pale cytoplasm (black arrow) and abundant chondromyxoid ground substance (white arrow). (May Grunwald Geimsa stain, 40X).

DISCUSSION

The PA is the most common benign salivary gland neoplasm. PA forms 45-74% of all salivary gland tumours and most commonly affects the parotid. It typically involves the lower pole and superficial lobes of parotid [1]; however, approximately 10% of cases arise from deep lobe and can expand intraorally to involve the parapharyngeal space [1,2]. It generally presents as a slowly progressing swelling, asymptomatic, without involving the facial nerve [3].

The first case of giant PA in medical literature was reported by Spence in 1863. Short DW and Pular P, reviewed massive PAs and reported a 2.3 kg adenoma [4]. Schultz-Coulon HJ, did a review of 31 cases of giant PAs [5]. Predominance of female cases (64.5%) was reported, with age ranging from 20-40 years, and weighing 1-27 kg [5]. PA can occur at any age; however, usually affects middle aged adults between 30-60 years. There is slight female predominance [6]. In this case, a 60-year-old female presented with the lesion evolving over a period of 20 years. The average size of a PA varies from 2-6 cm [7]. Alkindi M et al., reports a giant PA measuring 10×7×8 cm [8]. In the present case, the tumour grew to attain a huge size of 30 cm in the largest dimension. It usually presents as an asymptomatic slowly growing, firm, mobile, lobulated mass [7]. The consistency may vary from rubbery to firm depending on the presence of cystic or mucoid degeneration and chondroid or osseous tissue. Ulceration does not usually occur. Involvement of facial nerve and pain are rare [3]. Cosmetic deformity usually prompts the patient to seek treatment.

In the past PA was also known as mixed tumour, endothelioma, enclavoma, endochroma and branchioma [9]. It is considered as a benign neoplasm. Exact aetiology of PA continues to be unknown. Exposure to radiation increased the risk of developing PA. PA is a genetically heterogeneous neoplasm having different chromosome aberrations, translocations and gene mutations [10]. Most commonly it involves chromosomes 3, 8, 9, and 12 [11]. Deregulations of specific genes like PLAG1 encoding for zinc finger protein related to control of Insulin-like Growth Factor 2 (IGF II) expression have been detected. Simian virus 40 (SV 40) may also be a cofactor in the development or progression of PA [10].

Initial imaging modalities include USG, CT and MRI. In the present case, CT was performed for initial imaging and FNAC was performed for cytological diagnosis. FNAC is the preferred investigation for its diagnosis. It is considered a reliable procedure which can guide the surgeon and also help in choosing the correct surgical approach [12]. FNAC, whether blind or guided, is a rapid and cost-effective modality for early diagnosis of PA. Incisional biopsy is linked to recurrence and is contraindicated [3]. The incidence of malignant transformation in PAs range from 1.9-23.3% [13]. The risk increases in cases with long duration of evolution, recurrence and advanced age [3,14]. The transformation risk is 1.6% in tumours of age less than five years and it increases to 9.5% in cases with more than 15 years of evolution [15]. Clinical features associated with malignant transformation are ulceration, spontaneous bleeding, superficial and deep tissue invasion, onset of a facial nerve deficit, changes in consistency, more rapid growth and pain [3,16].

The treatment for PA is surgical excision, either superficial or total parotidectomy with recurrence being the least following total parotidectomy [17]. In the present case, the patient underwent total parotidectomy with preservation of the facial nerve. The [Table/Fig-4] is showing the comparison of present study with different studies [3,17].

S. No.	Author's name and year	Place of study	Age of subjects	Site of occurrence	Clinical and histopatho-logical findings	Diagnosis and treat-ment
1.	Sergi B et al., (2008) [3]	Rome, Italy	36-42 years	Left parotid, right submandibular, parapharyngeal space	3-6 cm, deep lobe. FNAC findings consistent with PA.	PA. Facial nerve preserving total parotidectomy
2.	Dhir P et al., (2014) [17]	Punjab	35 years	Left sided facial swelling	Asymptomatic, 2×1.5 cm. FNAC: mixture of spindle cells and round cells i.e., myoepithelial and ductal cells in clusters as well as individually in a fibromyxoid background	PA. Superficial parotidectomy.
3.	Present Study	Patna	60 years	Right infra-auricular	30×28×20 cm. FNAC: Round to oval cells with pale cytoplasm and abundant chondromyxoid ground substance	PA. Total parotidectomy with facial nerve preservation

[Table/Fig-4]: Comparison of present study with different studies [3,17].

CONCLUSION(S)

An indolent nature of the swelling, classical cytomorphological findings on FNAC and lack of recurrence were the clues to this diagnosis. The uniqueness of this case was that despite the huge size of the swelling, it came out benign. However malignant transformation can still be seen in elderly patients; therefore, they should be evaluated cautiously.

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