ORIGINAL RESEARCH PAPER
Volume - 9 | Issue - 7 | July - 2020 | PRINT ISSN No. 2277 - 8179 | DOI : 10.36106/ijsr
INTERNATIONAL JOURNAL OF SCIENTIFIC RESEARCH

OROPHARYNGEAL PLEOMORPHIC ADENOMA- A COMMON TUMOUR AT UNUSUAL SITE: CASE REPORT

Pathology

Dr. Vaishali Walke * Associate Prof, Pathology, Dept Pathology, Indira Gandhi Govt Medical, College. Nagpur. Maharashtra. *Corresponding Author

Dr. Sonali Datar Assistant Prof, Pathology, Dept Pathology, Indira Gandhi Govt Medical, College. Nagpur. Maharashtra

Dr. Joyce Thomas Senior Resident Pathology, Dept Pathology, Indira Gandhi Govt Medical, College. Nagpur. Maharashtra

Dr. Balwant Kowe Prof & Head, Pathology, Dept Pathology, Indira Gandhi Govt Medical, College. Nagpur. Maharashtra

ABSTRACT

Pleomorphic adenoma is the most common salivary gland tumour. It consists of cells with epithelial and myoepithelial differentiation, accompanied by variable amounts of characteristic chondromyxoid stroma. Usually they present as slow growing, solitary, unilateral, painless firm masses, commonly encountered in parotid gland while the infrequent sites are submandibular gland and minor salivary glands. Amongst minor salivary; oral cavity, retromolar region, tonsil, oropharyngeal and parapharyngeal regions are the rare locations. We report an unusual case of Pleomorphic Adenoma presenting as a large, oropharyngeal mass causing progressive dysphagia and dyspnoea in a young female. Here we discuss the morphological differential diagnosis at this anatomical location and the pointers to arrive at a correct diagnosis.

KEYWORDS

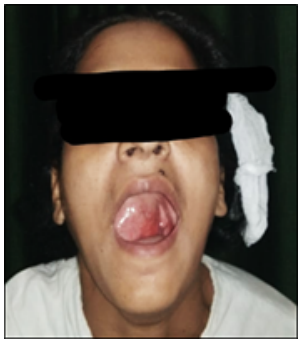
Pleomorphic Adenoma, Oropharyngeal Mass, Myoepithelioma, Parapharyngeal Mass, Minor Salivary Gland Tumour.

Introduction

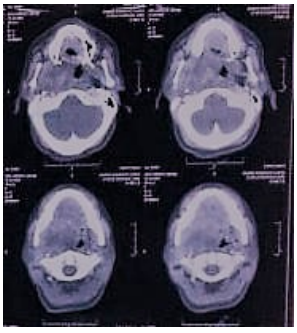
Pleomorphic adenoma (PA), is the most common benign tumour constituting up to two-thirds of all salivary gland neoplasms (73%). The most common sites are parotid gland (85%), minor salivary gland (10%), and the submandibular gland (5%). [1] Amongst minor salivary gland, the frequent locations of PA being soft palate (50%), lips (15%), cheek mucosa (12%), tongue (5%), floor of the mouth (5%); while the rare sites are tonsil, pharynx, retro molar area, gingiva and nasal cavity. [2,3] Also known as mixed salivary gland tumour, is commonly seen in female having maximum incidence in 4- 6th decade of life. [3] The signs and symptoms vary depending on its anatomical location. [4] Here we discuss a rare case oropharyngeal mass diagnosed as pleomorphic adenoma on histology. The aim is to create awareness about unusual presentations of this common tumour in order to avoid misdiagnosis thereby definitive treatment.

Case report:

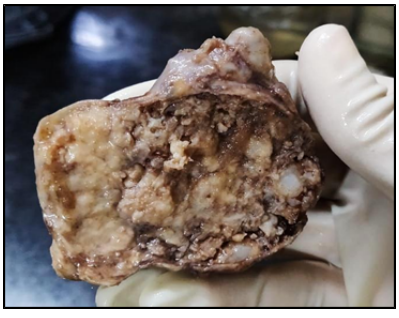
A 29-year young female attended ENT outpatient department with chief complaints of slowly growing mass in the oral cavity over a period of one year. She also gave history of dysphagia and difficulty in breathing of six months duration. Local examination revealed a mass of size 7x5x5 cm on the right side of oropharynx and protruding into ipsilateral oral cavity. The mass was soft to firm, pinkish white, non-tender with smooth surface. (Figure1; clinical photo)



There was no evidence of any cervical lymphadenopathy. Her general examination was within normal limits. The CT scan revealed a large, heterogeneously enhancing soft tissue mass involving right side of oropharynx and parapharyngeal space, anteriorly extending to right half of soft palate, tonsillar fossa and indenting base of tongue; laterally to right masticator space with loss of fat planes and pterygoid muscle, abutting submandibular gland and extending to floor of mouth causing mass effect over oropharyngeal airway. (CT Scan Figure 2)

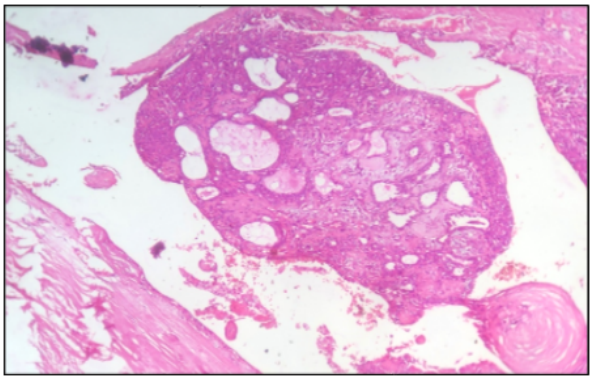


There was no evidence of bony erosion and the radiologist kept the possibility of a neoplastic process. The clinical impression was squamous cell carcinomas. Considering the neoplastic origin in mind, fine needle aspiration cytology (FNAC) was advised. On FNAC, 10 cc of thin yellow coloured fluid was aspirated, after which the swelling regressed partially. Fluid was centrifuged and smears were prepared which showed sheets and clusters of benign epithelial cells with bland nuclei were seen in a fluid background admixed with few inflammatory cells. A diagnosis of benign cystic lesion was offered. Surgical resection was done and we received excised partly capsulated, greyish pink, lobulated mass of size 5x5x2 cm. The cut surface was, mostly solid with few cystic areas, greyish white, soft to firm with foci of haemorrhage. (Gross: Figure 3)

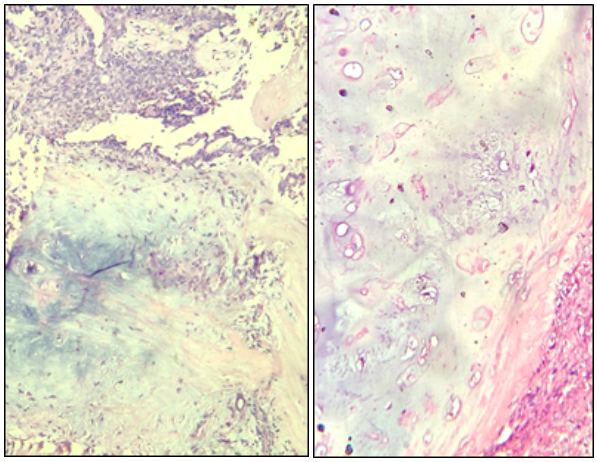


Haematoxylin and eosin (H&E) stained sections showed a well encapsulated tumour mass with compressed salivary gland tissue on one side. Tumour showed well define capsule and a biphasic pattern comprising of cellular and stromal components. Cellular element consisted of epithelial cells arranged in ducts, tubules and trabeculae lined by inner layer of ductal cells and outer myoepithelial. (HP: Figure 4)

PRINT ISSN No. 2277 - 8179 | DOI : 10.36106/ijsr



The surrounded stroma revealed spindle shaped myoepithelial cells embedded in chondromyxoid stroma and foci of cartilaginous differentiation. (Figure 5a& 5b)



These constellations of features together contributed to the diagnosis of pleomorphic adenoma. As complete resection of the mass was done no further surgical management was required for the patient. The post-operative period was uneventful and there was no evidence of recurrence post 6 months follow up.

Discussion:

Pleomorphic adenoma (PA) affect major salivary glands and in about 10-15% cases involve the minor salivary glands. Salivary gland tumors accounts for approximately 3-10% of all tumors arising in the head and neck region. [4] Soft palate is the most common site amongst minor salivary gland tumors (57%) followed by lips, buccal mucosa, tongue and floor of mouth; whereas rare sites are tonsil, pharynx, retro molar area, gingiva and nasal cavity. [2,3] Thus signs and symptoms of pleomorphic adenoma of minor salivary gland vary according to the anatomical site of origin. [4] In our case, the patient presented with a large unilateral, gradually increasing firm oropharyngeal mass. [3] The well circumscribed tumor mass without any evidence of infiltration into surrounding tissue and or necrosis favored a diagnosis of benign tumor. Looking at the clinical presentation of slowly growing but huge mass and radiological findings favouring neoplasm; the list of differentials ranging from benign neoplasms like pleomorphic adenoma, myoepithelioma, basal cell adenoma, schwannoma to malignant neoplasms like squamous cell carcinoma, mucoepidermoid carcinoma, adenoid cystic carcinoma, FDC Sarcoma were considered. [2,3,5] FNAC is an accurate pre-operative methodology for the diagnosis pleomorphic adenomas. However, in case of solid cystic swelling; when only cystic component is aspirated; it gives low cellular yield and the correct diagnosis may be missed. [1] The diagnosis of PA involves triangular approach which includes clinical examination, radiology and tissue sampling.[1] Histopathological picture is composed of mixture of epithelial and stromal component. Epithelial cells are arranged in cord-like, ductular or glandular patterns and stromal elements composed of fibrous, hyaline, myxoid, cartilaginous and or osseous areas. The amount of epithelium versus stroma varies tremendously from lesion to lesion. [6] Myoepithelioma the closest differential can arise in oropharynx; it was ruled out as the biphasic pattern comprising of chondromyxoid stroma and glandular differentiation a hallmark of PA was evident in our case. Myoepithelioma a monophasic tumour on the other hand is characterized by spindle or plasmacytoid to clear myoepithelial cells in diffuse sheets. [3] Basal cell adenoma another benign salivary gland tumour, also mentioned at this anatomical location, shows monomorphic population of basaloid cells with a distinct basement membrane like material; however, the classical chondromyxoid stromal component of mixed tumour is absent. [7] Ectomesenchymal chondromyxoid tumor (ECMT), a slowly growing, painless intraoral tumor exclusively seen on tongue and can morphologically mimic stroma rich Pleomorphic adenoma. Histologically it is a well circumscribed mass characterized by lobular proliferation of cells arranged in cords, strands and sheets of round to oval, polygonal to spindle cells with small uniform nuclei against a background of myxoid to chondromyxoid stroma. This tumor however lacks diagnostic ductal epithelial component of PA. [8] Schwannoma a benign nerve sheath tumour can also present as an oropharyngeal mass. Clinically it is indistinguishable from other benign neoplasm occurring at this site. Morphologically its an encapsulated mass having two distinct histologic patterns of Antony A and B areas which is in contrast with biphasic epithelial and myoepithelial components of PA. [9] The common malignant salivary gland tumour that can occur in oropharyngeal location is mucoepidermoid carcinoma. Histologically it shows characteristic three elements such as mucous cells, intermediate and epidermoid cells. All these diagnostic clues are absent in our case and morphology favoured PA. [10] Adenoid cystic carcinoma (ACC) although less common can occur in oropharyngeal region and may present as an asymptomatic mass. The three recognized histopathologic patterns of ACC are cribriform, tubular, and solid consisting of nests of basaloid epithelial cells containing multiple extracellular spaces. It is known for aggressive growth and characterized by perineural invasion [11] In case of large, oropharyngeal mass with dysphagia, squamous cell carcinoma (SCC) can be the most likely diagnosis. Histologically it is characterized by marked cellular pleomorphism, abnormally large hyperchromatic nuclei, increased N:C ratio, numerous typical and atypical mitotic figures with or without necrosis [5,10] However we may rarely encounter squamous metaplasia in PA. The squamous component here will be totally benign which will be seen against a background of epi -myoepithelial component and classical stroma.

Extra nodal Follicular Dendritic Cell Sarcoma (FDC) arising in oropharyngeal and parapharyngeal region is unusual and can mimic PA clinically. It shows fascicles, whorls and storiform pattern of spindle cells with elongated plump nuclei with coarse chromatin and nucleoli in some. The tumour cells are admixed with mononuclear cells a striking feature of FDC sarcoma. Thus, morphologically it is least likely to be miss diagnosed as a PA. The treatment of Pleomorphic Adenoma of minor salivary glands involves surgical excision with wide margins (1mm) due to occasional lack of encapsulation, mixing into normal host tissue and pseudopodia [3] Recurrence rate of PA of minor salivary glands is lower as compared to those arising from major salivary gland. [3] Malignant potential of PA ranges from 1.5% in first 5 years and 9.5% after 15 years. Malignant transformation must be suspected by the presence of clinical features such as pain, ulceration, fixity, spontaneous bleeding, and nerve palsy. [13] No features favouring malignancy were present in our patient and there was no evidence of recurrence post 6 months of surgery.

Conclusion:

Pleomorphic adenoma should be considered as one of the differential diagnosis even if unusual presentation as oropharyngeal mass is unusual. Its precise histopathologic diagnosis is essential as the treatment modality differs from its clinical mimics.

Abbreviations:

1. ACC: Adenoid cystic carcinoma
2. CT: Computer tomography
3. ENT: Ear nose throat
4. EMCT: Ectomesenchymal Chondromyxoid Tumour
5. FDCS: Follicular dendritic Cell Sarcoma
6. FNAC: Fine Needle Aspiration Cytology
7. H& E: Haematoxylin and eosin
8. PA: Pleomorphic adenoma
9. SCC: Squamous cell carcinoma

Legends:

Figure 1: Intra oral soft mass in right oropharynx protruding into the oral cavity. (Clinical Photo)

Figure 2: CT Scan: A large, heterogeneously enhancing soft tissue mass involving right side of oropharynx and parapharyngeal space and extending to floor of mouth causing mass effect.

Figure 3: Gross: A well encapsulated mass with variegated cut surface ranging from greyish white, brownish, translucent, fleshy, soft to firm nodular and cystic areas.

Figure 4: Tumour mass showing admixture of epithelial and stromal elements (HE;20X)

Figure 5a: Chondromyxoid stroma and myoepithelial component (HE; 40X)

Figure 5b: Inset Cartilaginous metaplasia (HE; 20X)

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