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Review Article

UNUSUALLY LARGE PLEOMORPHIC ADENOMA OF PALATE: A CASE REPORT AND REVIEW OF LITERATURE

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ABSTRACT

Pleomorphic adenoma (PA) is the most common benign mixed salivary gland neoplasm that accounts for 60% of all benign salivary gland tumors. It has a remarkable degree of morphological diversity. PA of minor salivary gland in the palate is a common entity. We report the case of a 40-year-old female who presented with a painless, slow growing swelling of the palate over the last 1 year. Computed tomography (CT) scan revealed an isodense structure in the palate causing bony erosion of the palatal region and perforating into the nasal cavity. Incisional biopsy report revealed PA of the palate. The entire tumor mass was excised along with the overlying mucosa followed by removal of the perforated bony structure. The Greater palatine vessels and nerve were dissected from the tumor mass and ligated. It was followed by chemical cauterization using carnoy's solution on the exposed bone to kill any remnants of tumor cell. The excised tumor along with excised palatal bone was sent for histopathology. A preformed stent encasing collagen and perio-pack was used to cover the defect. Histopathological examination confirmed the diagnosis of cellular PA of minor salivary gland. Patient has been on regular follow ups with no signs of recurrence and satisfactory healing.

INTRODUCTION

Pleomorphic adenoma (PA) is a most common benign salivary gland neoplasm with remarkable degree of morphological diversity[1]. It accounts for 60% of all benign salivary gland tumors and occurs in both major and minor salivary glands[2]. Parotid gland is the most commonly affected of the major group, and palate is the most common site of the minor salivary glands affected followed by lip, buccal mucosa, floor of the mouth, tongue, tonsil and reteromolar area[3].

Pleomorphic adenomas are considered as mixed tumours, composed of epithelial and myoepithelial cells. Epithelial cells typically form ductal structures. The stroma demonstrates varying degrees of myxomatous, hyaline, cartilaginous and osseous differentiation[1]. PA has a slight female predilection and clinically presents as well delineated painless firm mass and in most cases, does not cause ulceration of the overlying mucosa. Tumour when present on the palate causes difficulty in mastication and known to cause underlying bone erosion. When they are associated with minor salivary glands, they are more liable to be malignant (50%)[1].

Diagnosis is made on the basis of history, clinical examination, laboratory investigations and radiographs. Computed tomography (CT) and magnetic resonance imaging can be used to see the bony invasion, erosions, extensions, and surrounding areas of the lesion[4]. Fine needle aspiration cytology (FNAC) is a sensitive and minimally invasive diagnostic method implemented for determining the histological subtype[1,2,4,5]. The treatment of choice for pleomorphic adenoma of the palate is surgical excision followed by reconstruction for maintaining speech, mastication and esthetics of the patient[4]. This case report describes a case of mixed tumor, cellular pleomorphic adenoma in a minor salivary gland of the hard palate causing underlying bony erosion causing perforation into the nasal cavity.

Case Report

A 40 year old female patient reported to the Department of Oral and Maxillofacial Surgery with the chief complaint of non

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painful swelling over the palatal region since last 1 year. There was no preceding history of trauma .The swelling was slow growing and assymptomatic, no associated pain or parestheisa was reported. Patient had difficulty in speech and mastication. There was no regional lymphadenopathy, general physical and systemic examination was normal.

On clinical examination a dome-shaped swelling on the palate crossing the midline was seen (Fig ١). The swelling measured 4.5 cm × 2.5 cm in size. The mass had a bosselated surface and the overlying mucosa was pale with pinpoint erythema in the centre. The lesion was slightly lobulated, soft to firm, and nontender to palpation. . The patient was further subjected to routine blood and radiographic investigations including occlusal radiograph and CT scan. Occlusal radiograph revealed a well defined radio-opaque mass. Computed tomography (CT) scan revealed an isodense structure in the palate with signs of bony erosion of the palatal region (Fig ٢). Based on history and clinical examination, a provisional diagnosis of pleomorphic adenoma of the palate was given. The differential diagnosis included median palatal cyst, adenoid cystic carcinoma, mucoepidermoid carcinoma, lymphoma and myoepithelioma.

FNA biopsy was performed, which showed features suggestive of pleomorphic adenoma. Incisional biopsy was also done and was sent for histopathological examination. Microscopic examination of the incisional biopsy specimen stained with H&E showed features of pleomorphic adenoma.

Surgical Intervention

Pre-surgical preparation

An impression was made and a preformed acrylic stent was prepared.

Surgical

The tumor was excised using electrocautery inclusive of the palatal mucosa. On excision of the tumor, perforations on the palatal bone were seen, the weakend bony structures were removed and the edges were smoothened using acrylic trimming bur. The tumor was closely associated with greater palatine nerve and vessels. The tumor mass was dissected from the structures and the vessels were ligated. Due to the perforation of the palate there was a central region of exposure of nasal mucosa. The nasal mucosa was protected and Carnoy's solution was applied for 4 minutes on the remaining bone structure for the chemical cauterization of any remaining tumor cells. Hemostasis was achieved. The excised specimen was sent for histopathological diagnosis (Fig ٤&٣).

Post- surgical

After extubation and stabilization of the patient, the performed stent encasing collagen membrane overlying periopack was placed on the palate.

Histopathological Report

Grossly received a well encapsulated soft tissue specimen, greyish brown in colour, firm in consistency, measuring about 4x3.5 cm in dimension (Fig ٥). Histopathological examination of the specimen stained with hematoxylin and eosin showed a well encapsulated lesion surrounded by thick fibrous capsule clearly demarcated from the overlying parakeratinized stratified squamous epithelium. The lesion proper shows presence of numerous duct like structures with highly cellular areas consisting of sheets of ductal cells and plasmacytoid cells with eccentrically placed nucleus and glassy homogeneous eiosinophilic cytoplasm interspersed with scanty myxoid stroma. Few areas in the stroma also shows extrensive hyalinization. Areas of squamous metaplasiawith keratinization is also evident (Fig ٦). Histological features were confirmatory of cellular pleomorphic adenoma. The decalcified sections from the bone from the base of the tumour around the perforation into nasal cavity showed no evidence of tumour cell infiltration. The patient is followed up for 6 months and was observed for any sign of recurrence of the disease.

DISCUSSION

Pleomorphic adenoma (PA) is a most common benign salivary gland neoplasm with remarkable degree of morphological diversity[1]. It accounts for 60% of all benign salivary gland tumors and occurs in both major and minor salivary glands [4,7]. Parotid gland is the most commonly affected of the major group, and palate is the most common site of the minor salivary glands [1,2,4,6].

According to a study conducted by Spiro *et al*, including 2078 patients with salivary gland neoplasia, 20-40% of all salivary gland tumors arise from minor salivary glands. The mixed minor salivary tumors affects mostly patients in their fourth to sixth decades of life[4,9,10]. Though it is been reported to affect both the sexes, slight predilection for female gender with ratio of 2:1 has been reported. This current case of PA is reported in a 40 year old female patient which is in accordance to the literature.

Intra oral PA appears as slowly growing, painless mass. Pain, tenderness and ulceration are unusual. Although it is a benign tumor, it has a high recurrence rate and in a small number of cases, a benign PA may degenerate into a malignant tumor. When it occurs in oral cavity it usually lacks its characteristic fibrous capsule due to which it has a high recurrence rate. These tumors are also able to invade and erode adjacent bone, causing radiolucent mottling on the x-ray of the maxilla. In our present case CT imaging showed evidence of bony erosion of the palate caused due to pressure effect[2].

The case reported in this study presented a well-circumscribed, encapsulated lesion, presented with smooth surface initially, which got ulcerated due to intervention during FNAC and incisional biopsy[2,4]. Necrosis in PA can be divided into spontaneous and induced. Necrosis of the pleomorphic adenoma can occur following aspiration puncture as seen in our present case, which is attributed to the interruption of microvascular supply, arising from vigorous aspiration of the tissue[2].In some cases the tumour may induce pressure on the major or minor palatine arteries, which would leave the lesion without blood supply and consequently leading to necrosis[2,4]. The diagnosis of PA is established based on the history, clinical presentation and cytological and histopathological examinations. Cytological sampling procedures commonly include FNAC and core needle biopsy (thicker needle used when compared to FNAC). According to Feinmesser and Gay 90% accuracy of FNAC was reported, whereas Core needle biopsy is more invasive but is more accurate compared to FNA with diagnostic accuracy of 97% 2. In this reported case FNAC was performed and on cytopathological examination of the

smear showed presence of cellular areas consisting sheets of predominantly plasmacytoid like cells with eccentrically placed nucleus with pale glassy eiosinophilic cytoplasm suggestive of benign salivary gland neoplasm.

Histopathologically, PA is considered as mixed tumour because of its morphological diversity. It is an epithelial tumour of complex morphology, possessing epithelial and myoepithelial cells arranged in a variety of patterns which is embedded in a mucopolysaccharide rich myxomatous stroma. Mucous producing cells and keratinizing producing cells may or may not be seen. Myoepithelial cells may have an angular or spindle like presentations, they may also be presented as rounded cell with an eccentric nucleus, and they have a striking resemblance with plasma cell.[2,4,8,9]. The tumour will be well encapsulated. Formation of the capsule is as a result of fibrosis of surrounding salivary parenchyma. In the present reported case highly cellular stroma with areas of squamous metaplasia and epithelial pearls were evident.

Conventionally, the tumors of the hard palate are treated by wide local excision through the mucoperiosteum. Usually, pleomorphic adenoma does not recur after adequate surgical excision. Most recurrences can be attributable to inadequate surgical techniques such as simple enucleation leaving behind microscopic pseudopod-like extensions, or due to capsular rupture and subsequent tumour spillage during excision Hard tissue defect of the palate can be treated using obturators, and soft tissue defect can be left to heal[4,7,8,9]. In this case, we used a preformed stent with collagen membrane so as to enhance the healing and protect the exposed nasal mucosa.

The patient has been on regular follow-ups shows satisfactory healing and no signs of recurrence.

CONCLUSION

Pleomorphic adenoma of minor salivary gland of palate is a rare entity. Its symptoms include painless, slow growing mass on the hard palate. Diagnosis can be made on basis of histopathological examination. Treatment was done using wide excision, peripheral ostectomy and chemical cauterization using carnoy's solution. The defect was further covered using a pre-formed stent encasing collagen membrane. The patient has been on regular follow-ups and healing is satisfactory.

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