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RESEARCH ARTICLE

LIPOMATOUS PLEOMORPHIC ADENOMA- A RARE ENTITY

[1]Dr. Subhasish Burman, [2]Dr. S.K.Majumdar, [3,*]Dr. Siddhartha Mishra, [4]Dr. Chandmani Tigga, [5]Dr. Mustafizur Rahaman and [6]Dr. Divya Chadda

[1]Associate Professor, Dept. of Oral and Maxillofacial Surgery, Dr. R. Ahmed Dental College and Hospital
[2]Professor and HOD, Dept. of Oral and Maxillofacial Surgery, Dr. R. Ahmed Dental College and Hospital
[3,4,5]Third Year M.D.S. Student, Dept. of Oral and Maxillofacial Surgery, Dr. R. Ahmed Dental College and Hospital
[6]Third Year Post Graduate Trainee, Dept. of OMFS, DR. R. Ahmed dental College and Hospital

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ABSTRACT

Pleomorphic adenoma (PA) is the most common salivary gland tumour of major and minor salivary glands accounting for about 40-70% of all salivary gland tumour. The commonest intraoral site is palate followed by buccal mucosa and lip. Histologically pleomorphic adenoma consisting of cells exhibiting the ability to differentiate to epithelial cells and mesenchymal cells. Avery few cases of lipomatous pleomorphic adenoma a rare clinical entity characterized by presence of lipomatous tissue has been reported in literature. Total surgical removal of the lesion is the treatment of choice and low recurrence rate has been found after complete removal of this tumour. In this present case report a lipomatous pleomorphic adenoma of palate in a 24 year old patient is described.

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INTRODUCTION

Salivary gland tumours are less common, incidence rate is about 3% of all head and neck tumours (Rajendran, Mcllveen, 1987). The pleomorphic adenoma (PA) or mixed benign tumour is the most common entity and accounts for 40–70% of all tumour (Shafer, 1983). Parotid gland is most commonly affected among the major salivary gland, and palate is the most common site of the minor salivary glands affected. The presence of tumour in the palate can cause difficulties in chewing, phonetics and breathing according to the severity of tumour extension (Soto *et al*., 2014). Clinical presentations of intra oral pleomorphic adenoma are mostly painless, firm mass in the posterior palatal mucosa. If the mucosa is ulcerated and the ulceration is not attributable to trauma or biopsy, the mass should be considered a malignancy. Radical resection is indicated for these tumours as inadequate resection leads to local recurrence (Clauser *et al*., 2004). The term "pleomorphic" refers to heterogeneous components of the tumour which may have chondroid, myxoid, hyaline, osseous and lipomatous tissue.

Corresponding author: **Dr. Siddhartha Mishra,**
Third Year M.D.S. Student, Dept. of Oral and Maxillofacial Surgery, Dr. R. Ahmed Dental College and Hospital.

The lipomatous pleomorphic adenoma is extremely rare entity (Korkmaz, 2002; Seifert *et al*., 1999; Kondo, 2009). Incisional biopsy and FNAC helps in diagnosing intra oral pleomorphic adenoma involving palate. Computed tomography (CT) scan and Magnetic resonance imaging (MRI) helps in determining the extension of the tumour. According to the extent of the tumour different types of maxillectomy (Simple, subtotal or total maxillectomy) is needed. We report a case of lipomatous variation of pleomorphic adenoma involving right side of palate in a 24 years old male patient. Subtotal maxillectomy was done in this case.

Case Report: A 24 years male patient came to our department with the chief complain of swelling in the right palatal region since two and half years .No associated pain was present, no rapid growth of the swelling was present. On intraoral examination a well defined firm swelling measuring about 5 cm x 4 cm was found in the right side of palate extending from mesial surface of right maxillary canine to right maxillary retromolar region and the swelling was crossing midline in mid palatal region (Fig.1). No surface ulceration or pigmentation was present. Adjacent teeth were not mobile and no paraesth esia was noted. No cervical lymphadenopathy was found. Incisional biopsy was done and it revealed lipomatous variation of pleomorphic adenoma.

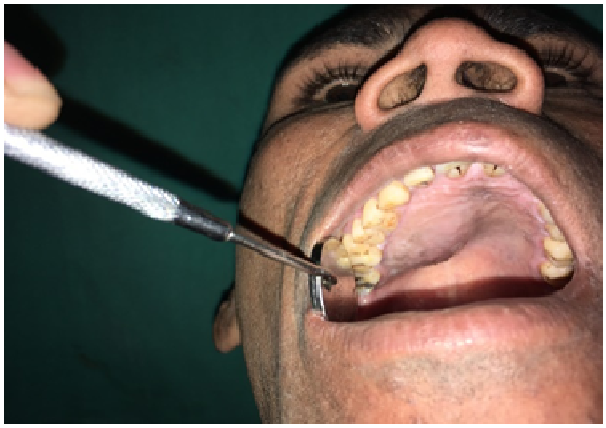

Fig.1. Pre Operative Intra Oral Photo Showing Palatal Swelling

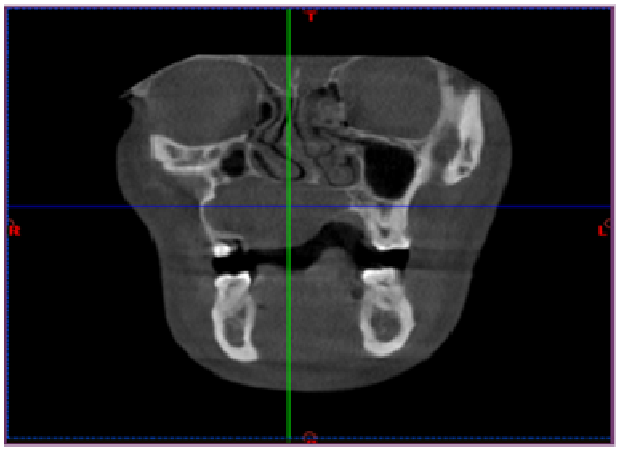

Fig. 2. Radiograph showing extension of the tumour

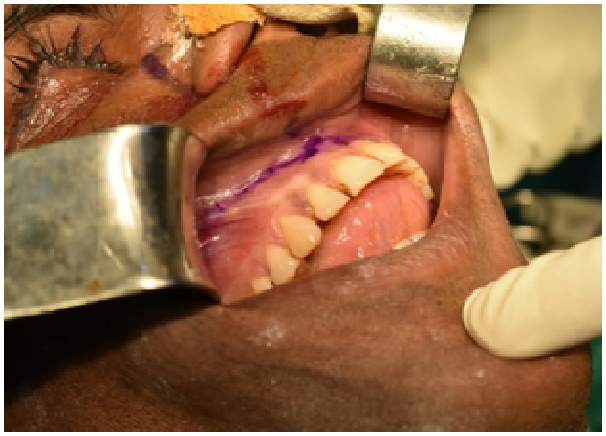

Fig.3. Intraoral Marking of The Planned Incision Line

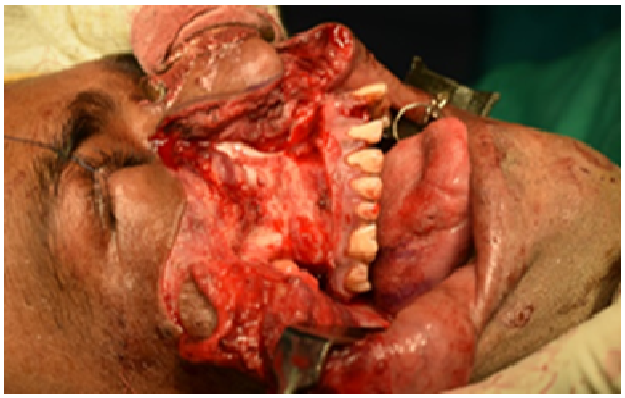

Fig. 4. After Complete Buccal Flap Exposure

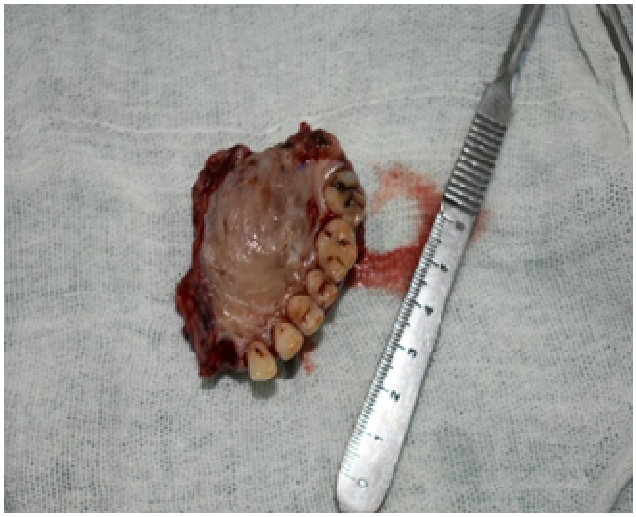

Fig.5. Resected Tumour

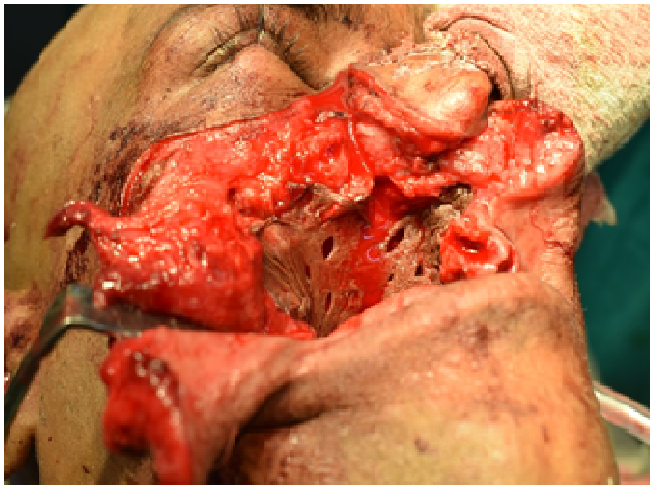

Fig. 6. Split Thickness Skin Grafting Placed In The Defect

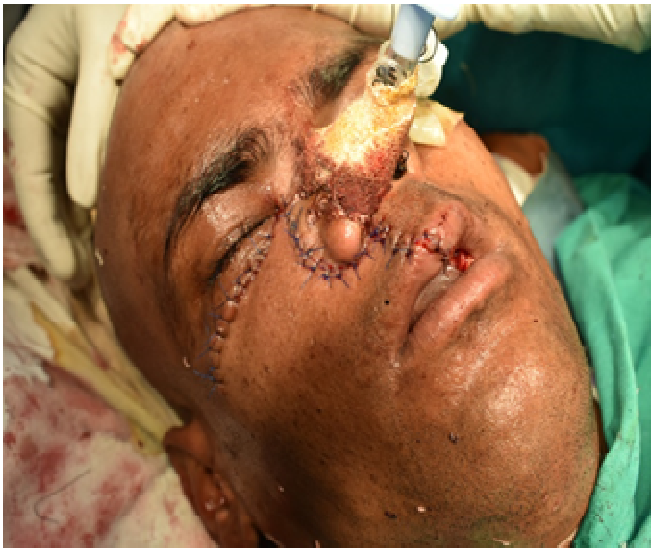

Fig. 7. Closure of the Wound

Contrast enhanced computed tomography and CBCT was done to evaluate the extension of the tumour (Fig.2). Radiographs revealed the tumour extended to involve all walls of maxilla and breach in continuity was found in the anterior wall of maxilla. No tumour mass was noted to invading maxillary sinus or orbital floor.

Necessary investigations was done including complete blood count , tests for coagulopathies ,liver function test, kidney function test, chest radiograph in PA view and ECG was done to assume any systemic comorbidity. All investigations were with in normal limits and pre anaesthetic check up for general anaesthesia was obtained. A modified weber-ferguson incision was given for subtotal maxillectomy after marking incision line (Fig.3). Incision was done with no 10 blade and scalpel, deeper tissue dissection was done with electrocautery blade. After flap elevation marking of the tumour mass along with 1cm free bony margin was done (Fig.4, Fig.5). Subtotal maxillectomy done with the help of sharp chisel. Split thickness skin graft was harvested from left thigh in aseptic condition and it was placed in the maxillary defect area (Fig.6) and was secured with the surrounding mucosa by absorbable suture. Temporary Obturator was placed and secured in the defect with interdental wiring. Cheek flap was closed in layers (Fig.7). Resected specimen was sent for histopathological examination. Histopathological evaluation confirmed the initial diagnosis of lipomatous variation of pleomorphic adenoma (Fig.8). Post-operative complication was peri-orbital oedema due to lymphatic obstruction (Fig.9), which gradually resolved. No facial nerve impairment was seen in postoperative period. After 3 months, the patient was recalled and the clinical examination showed complete epithelialization of the defect (Fig.10).

DISCUSSION

Pleomorphic adenoma is the most common benign neoplasm that occurs in the salivary gland, 84% occurs in the parotid gland, 8% in the submandibular gland and 4-6% in the minor salivary glands. The term 'pleomorphic' describes the embryogenic origin of these tumours i.e., a benign neoplasm consisting of cells exhibiting the ability to differentiate to epithelial and mesenchymal cells (Rajendran, ?). The PA may shows changes in the stromal and epithelial components, such as sebaceous, lipocytic, oncocytic and squamous metaplasia. A rare characteristic for PA is extensive lipomatous differentiation (Musayev *et al*., 2014), there are few cases reported in the literature (Korkmaz, 2002). The first "lipomatous pleomorphic adenoma" case in literature was reported in 1995 (Musayev *et al*., 2014) and the term was described by Seifert, characterizing the tumour as atypical with more than 90% of its area composed of adipose tissue (Seifert *et al*., 1999; Farina *et al*., 1997).

The histogenesis of LPA is not well established, with two possible mechanisms: metaplastic transformation of myoepithelial cells to adipocytes and entrapment of fat tissue during development (Korkmaz *et al*., 2002; Matsuzaka *et al*., 2003). In differential diagnosis lipoma, spindle cell lipoma, lipoadenoma, interstitial lipomatosis, lipomatous hepertrophy, sialolipoma, well-differentiated low-grade liposarcoma should be considered (Seifert *et al*., 1999; Siddaraju *et al*., 2009). Since the literature reports and the present case confirms that there is no standard symptoms of LPA, which complicates the differential diagnosis. When the LPA affects the parotid gland it is become difficult to diagnose the tumour by a computed tomography due to its lipomatous component that resembles the normal pattern of the gland tissue [6,8]. We should be more attentive to the recognition of the lesion in order to establish a diagnosis and proper clinical management. When the tumour affects the hard palate it becomes more difficult to diagnose as palate usually does not contain lipomatous components.

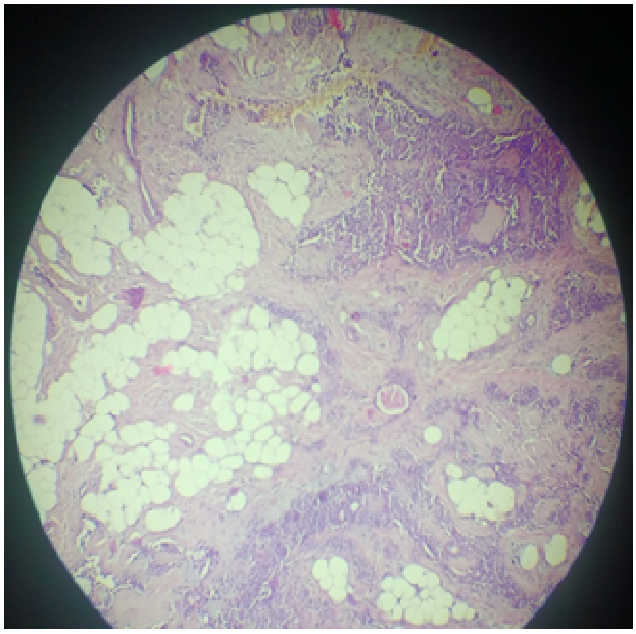

Fig.8. Photomicroghaph of the histology: Lipomatous tissue

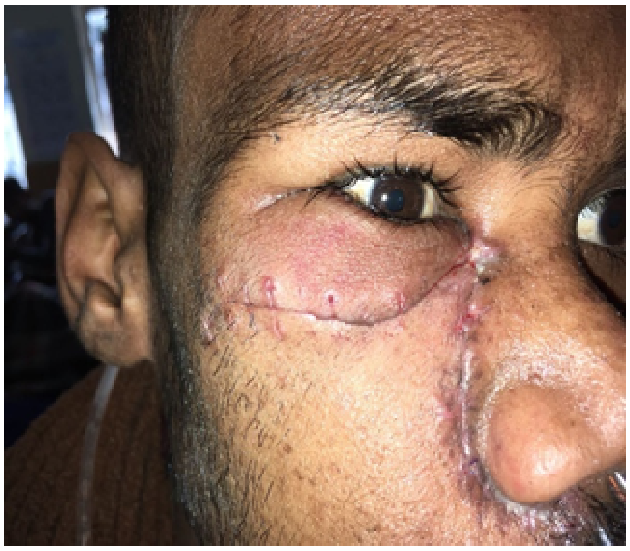

Fig. 9. 7thday post operative

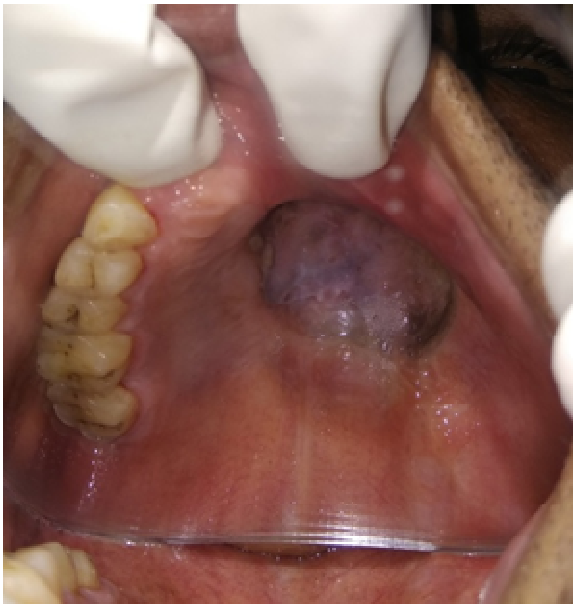

Fig.10. Complete epithelialization of the defect

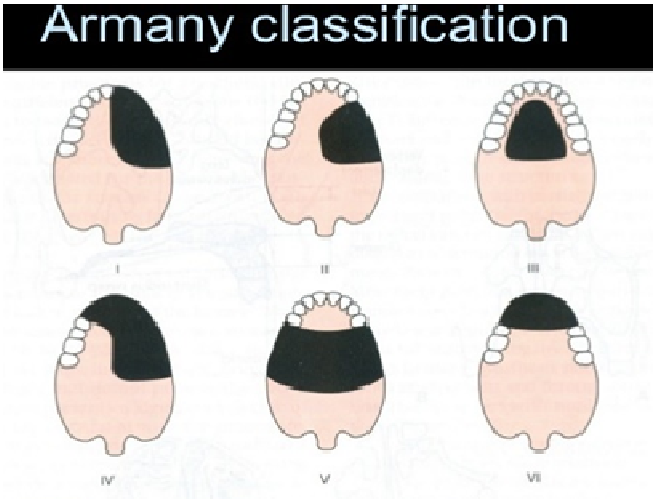

Fig.11. Armany classification of different maxillectomy defect

The fine needle aspiration (FNA) is indicated when there is presence of pain and swelling in the same one (DeMay *et al*., 2009; Simpek *et al*., 2009). There are different reconstruction options for maxillary defect such as Obturator with replacement of teeth, metallic mesh, pedicled flap, free flap. In pedicled flap temporalis myofascial flap is the most versatile flap. Different free flaps for maxillary soft and hard tissue defects are free fibula, radial forearm free flap, DCIA etc (Cordeiro *et al*., 2012). In our present case report the defect produced was class IV defect (Fig.11) according to Armany classification (Zubair Durrani *et al*., 2013). Definitive Obturator with replacement of the teeth was planned for reconstruction of the defect in the present case. After complete healing and epithelialization of the surgical area the definitive obturator will be fabricated.

Conclusion

The confirmation of this rare subtype of pleomorphic adenoma is essential to determine clinical management. Therefore, this lesion must be known to the pathologist in order to establish a differential diagnosis of lipomatous tumour. The elimination of malignancy is paramount important to manage this kind of lesion.

Conflict of Interest: None

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