

Special Article - Oral and Maxillofacial Surgery

A Case of Massive Recurrent Pleomorphic Adenoma of the Palate

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Abstract

Pleomorphic adenomas are the most common benign tumors arising from the cells of salivary gland tissue characterized by the presence of both epithelial and mesenchymal elements. About 10 percent of the pleomorphic adenomas occur in the palatal minor salivary glands. The present case is that of a 73 year female patient that reported with histopathological diagnosed case of a massive recurrent pleomorphic adenoma of the palate. The patient was reportedly operated elsewhere 20 years ago and had developed a diffuse enlargement of bilateral maxillae since 5 years. Computed tomography scan showed extensive bony destruction with tumor extension diffusely into bilateral maxilla, nasal and palatine bones inferiorly. Superiorly approximating the floor of the orbit and extending distally into the sphenoid bones and the cranial base. Case assessment conclusion was that of an inoperable tumor with areas of cranial extension. The patient was treated symptomatically and was kept on basic life support.

Background

Pleomorphic adenomas are the most common benign tumors arising from the cells of salivary gland tissue characterized by the presence of both epithelial and mesenchymal elements. Pleomorphic adenomas may occur at any age but occur more commonly in 3rd to 6th decade of life and are slightly more frequent in women (male-to-female ratio 1:1.39) [1]. 60 to 85 percent of pleomorphic adenomas occur in the parotid. The minor salivary glands of the palate account for only 10 percent of the pleomorphic adenomas. The site of predilection is the mucosa over the posterior hard palate and anterior soft palate. Most frequent presentation initially is a firm painless mass with intact overlying mucosa. Ulceration may be present secondary to trauma or previous biopsy. Ulceration not secondary to the above mentioned causes may be malignant. The tendency to recur and the accompanying risk of malignant transformation make the clinical diagnosis and treatment of pleomorphic adenomas important. The rate of malignant transformation reported in literature is 1.5% to 23%.

Case Presentation

A 73 year old female patient reported to the maxillofacial department with complaint of a large growth in the upper jaw bone. Clinical examination revealed severe facial deformity with expansion of bilateral maxillae and palatine bones, deformed nasal bones, left malar prominence and no regional lymphadenopathy. Few mobile teeth (two premolars on the right side and molars on the left side) were present in the enlarged maxilla. Anterior palate had areas of ulceration left over by extraction or by trauma from mandibular teeth. Indentations of mandibular teeth were present bilaterally (Figure 1-3). The patient was on assisted respiration by a nasotracheal intubation. She was poorly nourished, her weight was 33 kg, and she could not walk by herself. The patient reported of twice being operated for a pleomorphic adenoma of the palate. The first surgery was performed 20 years ago. The second surgery



Figure 1: Frontal view.



Figure 2: Left lateral view.

for a reported recurrence was performed 8 years back at the same centre. Histopathological examination following a deep multiple location biopsies were consistent with the previous reports, that is



Figure 3: Intra oral view.

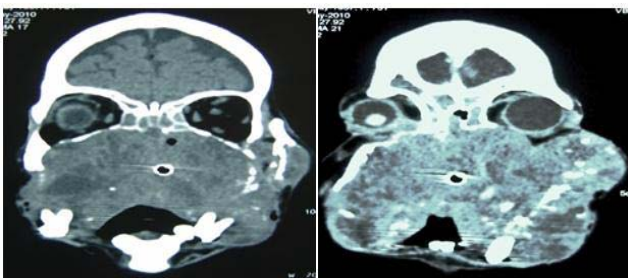


Figure 4: Coronal view computed tomography scan.

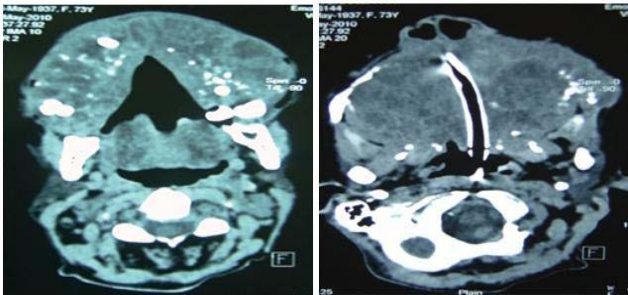


Figure 5: Axial view computed tomography scan.

of a pleomorphic adenoma. Since then the medical condition of the patient deteriorated and the recurrent enlarged mass at present state was considered to be inoperable after a thorough examination of the tumor extension using a computed tomography scan (Figure 4-8).

Investigations

A computed tomography scan was performed with axial, coronal, saggital and 3D reconstruction images to determine the extent of the tumor (Figures 4-8). Tumor arising from the maxilla, almost entirely occupying the alveolar process, bilateral maxillary sinuses and the nasal cavity. The scan revealed a massive tumor extending into bilateral maxillae, alveolar processes, palatine bones, infraorbital rims and laterally involving the whole of the zygomatic bones on the left side with extensions into the temporal and infratemporal spaces with incomplete involvement on the right side. The nasal bones were deformed with complete erosion of the nasal septum and involvement of the ethmoidal bones and sphenoid bones laterally. Saggital scans from the middle of the tumor mass showed tumor invasion into the skull base and the cranial cavity.

The mobile teeth causing pain were extracted and an incisional



Figure 6: Saggital view- computed tomography scan.



Figure 7: 3D Ct reconstruction scan.

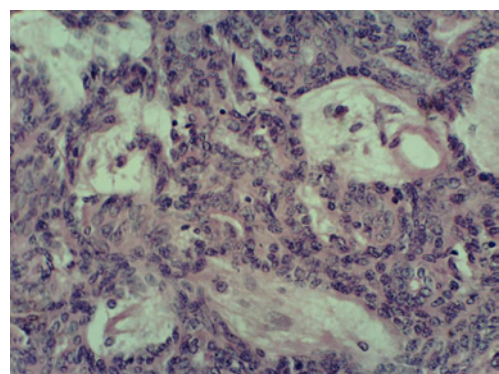


Figure 8: Microscopic view showing islands of ductal epithelial cells and hyalinized stroma.

biopsy specimen was taken for histopathological examination. The histopathological examination was conclusive of a recurrent pleomorphic adenoma.

Discussion

The palate is the most common site of minor salivary glands (10%), followed by the lip (4%), and unusual sites such as the sinuses, larynx,

epiglottis, and trachea have been reported. Depending on site, it may present with dyspnoea, dysphagia, acute airway obstruction, and obstructive sleep apnea. In the present case a severely compromised airway was managed by prolonged nasotracheal intubation. Both benign and malignant tumors arising in salivary glands generally grow slowly. They are painless and it may take a long time for patients to consult a doctor even if they have noticed the tumor. However tumor of the palate is often detected earlier than parotid tumors. Very few case reports of massive pleomorphic adenomas of the palate exist in English literature. Recent advances of diagnostic techniques and also an increase in the public awareness of disease has helped in limiting the disease [2]. Byars et al. [3] (1957) reported the first case of a recurrent pleomorphic adenoma that reported recurrence 5 years after treatment. Sasaki et al. [4] reported 11 cases in a review of massive tumors arising from the salivary glands, weighing more than 200 g between 1979 to 1998. Of the 11 tumors only one was from the palate. Galich [5] described a case of a 12-year-old girl who presented an ulcerating PA on the right hard and soft palate extending into the pterygoid fossa, which was tolerated for 18 months before drawing attention because of pain and bleeding. Passi et al. [6] reported a tumor of the soft palate that measured 3 x 4 cm which was managed by surgical excision. A confirmed pleomorphic adenoma of the palate is excised with 1 cm clinical margins which includes the overlying mucosa and periosteum of the palate. The rate of tumor recurrence after surgery for benign pleomorphic adenoma varies considerably in different clinical settings and seems to depend to a great extent on the surgical technique used. Maran et al. [7]. (Arch Otolaryngology 1984) underlined that the main causes of recurrence in a pleomorphic adenoma to be: rupturing of the capsule causing tumor implantation, incomplete excision of Islands of tumor tissue left behind and multicentricity of pleomorphic adenoma. It has been observed that the capsule characteristics such as incomplete capsule, capsular penetration, pseudopodia and satellite nodules maybe compromising a complete tumor resection. Buchman C et al. [8]. (Laryngoscope 1994) in their study emphasized that microscopically positive margins after resection of a pleomorphic adenoma were associated with an increased rate of tumor recurrence. Intraoperative tumor spill as a cause of postoperative recurrence has recently been questioned. The present case seems to be one of an incomplete excision of the primary tumor with a multifocal point of recurrence. The multifocal enlargement subsequently involved the bilateral maxillae, alveolar processes, palatine bones, infraorbital rims and laterally involving

the whole of the zygomatic bones on the left side with extensions into the temporal and infratemporal spaces. Henriksson et al. [9] reported cancer 1998 in their study of 197 patients; follow up of 9.7 years reported recurrence rate of 7% following capsule rupture as against a total recurrence of 4% when no capsule rupture occurred. Their study reported that the occurrence of pseudopodia that extend beyond the main lump of the tumor was a significant risk factor for local recurrence rather than the tumor spill.

Learning points/Take home messages

- Confirmation of lesion by FNAC/ biopsy taken from centre of lesion.
- Complete removal with recommended margin (1cm) of healthy tissues.
- Long term follow- up and early treatment of recurrence-adenoma may transform into carcinoma.

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