Pleomorphic Adenoma of Minor Salivary Gland in a 14 year Old Child

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Abstract Salivary gland tumours are rare in childhood, and almost all of them occur in parotid gland. Minor salivary gland tumours are even rarer, pleomorphic adenoma being the most frequently found tumour. Only seventeen cases of pleomorphic adenoma arising in the minor salivary gland tumour have been reported in children and adolescents. Pleomorphic adenoma of minor salivary gland represents about 45% of all the tumours of the minor salivary glands. Pleomorphic adenoma is slowly enlarging tumour indistinguishable from adenoid cystic carcinoma clinically, except for pain and ulceration, which is more common in the latter. Carcinoma arising from pleomorphic adenoma has been reported in 3% cases amongst the minor salivary gland tumours. This report presents a case of pleomorphic adenoma of minor salivary gland in a 14 year old female patient with a brief review of literature.

Keywords Pleomorphic adenoma · Minor salivary gland tumour · Cystic degeneration

Introduction

Salivary gland tumours are rare in children and the incidence differs from the adult counterpart. When salivary gland tumours do arise in children, they preferentially affect major salivary glands; however minor salivary gland tumours have also been reported. Pleomorphic Adenoma is a minor salivary gland tumor and indistinguishable from

the adenoid cystic carcinoma except for pain and ulceration of overlying mucosa, which is rarely seen in pleomorphic adenoma. Among the cases of pleomorphic adenoma of minor salivary gland reported in children, recurrences were seen in two cases [1].

Case Report

A 14 year old female patient reported with a chief complaint of swelling in palatal region for past 2 months.

Clinical Presentation

Examination revealed a single swelling on right posterior part of the palate extending anteroposteriorly from the right premolar region to the soft palate and mediolaterally from the right palatal mucosa to the midline of the palate. The swelling was 3 cm × 3 cm in diameter, nodular and fluctuant (Fig. 1). All the teeth in upper right quadrant were vital. Radiolucent shadow with well corticated border was seen on paranasal sinus view and occlusal view radiographs (Fig. 2). On aspiration of the swelling, a dark straw coloured fluid was obtained.

Diagnosis

The site of the lesion being posterior part of palate and the swelling being solitary and nodular were suggestive of minor salivary gland tumour. Other features such as straw coloured fluid obtained on aspiration were pointing towards the diagnosis of an odontogenic cyst or odontogenic tumour with cystic degeneration. Incisional biopsy and

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Fig. 1 Preoperative photograph

Fig. 2 PNS view showing radioluscent shadow with well corticated border on right posterior palate region

histopathological examination revealed the diagnosis of pleomorphic adenoma.

Management

Surgical excision and histopathological examination were planned. Crevicular incision was given in palate from upper right first molar to upper left first molar. Mucoperiosteal flap was raised. Encapsulated mass of tumour was separated from the overlying periosteum and underlying bone. The tumour was removed in toto (Fig. 3).

The flap was repositioned and sutured. Prior to the surgical procedure, an impression of maxillary arch was made and cast prepared with dental stone. The elevation on the cast was trimmed and a surgical stent was prepared. After the flap was repositioned, surgical stent was inserted. The excised mass was well encapsulated and had a glistening tan. The cut surface was rubbery. The specimen was

Fig. 3 Excised specimen

sent for histopathological examination. H&E section revealed the following findings.

1. Highly cellular stroma with fibrous encapsulation.
2. Bilayered duct with eosinophilic coagulum.
3. Myxochondroid areas along with areas of extensive hyalinization.
4. Areas of cystic degeneration with haemorrhage.

All the histopathological findings confirmed the diagnosis of pleomorphic adenoma (Figs. 4, 5, 6).

Discussion

‘Pleomorphic adenoma’ is a term suggested by Willis, because of its unusual histological pattern. It is not a mixed tumour, as it is not teratomatous or derived from more than one primary tissue. The morphogenic complexity is because of differentiation of the tumour cells into fibrous, hyalinised, myxoid, chondroid and even osseous tissue [2]. 45% of all tumours of minor salivary gland are pleomorphic adenoma [3].

Fig. 4 40× magnification showing bilayered ducts with eosinophilic coagulum in a cellular stroma (H&E stain)

Fig. 5 10× magnification showing nests of epithelial cells and areas of cystic degeneration

Fig. 6 20× magnification showing myxochondroid areas

The pleomorphic adenoma of intraoral accessory salivary gland seldom is allowed to attain a size greater than 1–2 cm in diameter [2]. In our case the tumour was 3 cm in diameter. The straw coloured fluid was due to cystic degeneration seen in the tumour.

Jansisyanont et al. [4], Neely et al. [5], and Gonz et al. [6] have reported that malignant tumours accounted for about 40–70% of all epithelial minor salivary gland tumours. In a series of 737 minor salivary gland tumours, Wang et al. [7] found 53.9% to be malignant. So any minor salivary gland tumour should be viewed with suspicion and any malignant transformation should be ruled out by histopathological examination. Minor salivary gland tumours range from benign pleomorphic adenoma to malignant mucoepidermoid carcinoma or adenoid cystic carcinoma. Clinical presentation of pleomorphic adenoma is similar to that of adenoid cystic carcinoma, except for pain and ulceration, which is seen in later [3]. Carcinoma arising from pleomorphic adenoma accounts for about 3 percent of salivary tumors [3]. Thus complete removal and thorough histopathological examination of any minor salivary gland tumour is mandatory and gold standard for treatment of pleomorphic adenoma.

Only six well-described cases of pleomorphic adenoma arising in the palate was reported in children till 1996 [8]. 11 additional cases were reported after 1996 (Table 1). Till now seventeen cases of paediatric pleomorphic adenoma gland of minor salivary gland have been reported in literature. Salivary gland tumours are rare in children and the incidence differs from the adult. When salivary gland tumours do arise in children, they preferentially affect major salivary glands.

In a retrospective analysis of 684 cases of salivary gland tumours in a single institution, Subhashraj [11] found 62% to be benign, 38% to be malignant; 61% to be in parotid gland, 22% in minor salivary gland and 17% in submandibular gland. In this retrospective analysis done between 1991 and 2006, the most common benign tumour was pleomorphic adenoma, accounting for 86% and the most common malignant tumours were adenoid cystic carcinoma (25%) and mucoepidermoid carcinoma (18%). Among the minor salivary gland tumours, 68% were seen in palate. A rare and unusual site for pleomorphic adenoma of minor salivary gland of upper lip was reported by Lotufo [12]. Intraoral salivary gland tumours were reviewed in two

Table 1 Paediatric pleomorphic adenoma of minor salivary gland reported in literature (post 1996)

Published Journal	No. of cases	Age/sex	Region	Treatment rendered	Recurrence
Med Oral Pathol Oral Cir Bucal 2009 Feb; 1:14(2):E73–75.	1	13 year/F	Palate	Wide excision	No
Int J Oral Maxillofac Surg 1996 June; 25(3):206–207.	1	16 year/M	Palate	Wide excision	No
Br J Plast Surg 2001 Apr; 54(3):245–247.	1	9 year/M	Palate	Wide excision	Yes
J.Oral Sci. 2008 Jun; 50(2); 225–228.	1	12 year/M	Upper lip	Complete excision	No
Int J Oral Maxillofac Surg. 2002 Jun; 31(3): 273–275.	2	Under 18	Palate	Surgical resection	No
Int J Oral Maxillofac Surg. 2002 Jun; 31(3): 273–275.	2	Under 18	Upper lip	Surgical resection	No
Int J Oral Maxillofac Surg. 2002 Jun; 31(3): 273–275.	2	Under 18	Tongue	Surgical resection	No
ASDCJ Dent Child 1998 May–Jun; 65(3):209–211	1	Not specified	Palate	Not specified	Not specified

Brazilian institutions by Jorge et al. [13] and five cases of pleomorphic adenoma in patients under 18 years of age were found. Four patients were female and one was male; two cases affected the palate, two the upper lip and one the tongue. All five patients were treated surgically and after long term follow-up, they found no recurrence. It was suggested that, in youngsters, pleomorphic adenoma seems to have similar biological characteristics as in adults, with low recurrence rates after surgical resection. A case of palatal pleomorphic adenoma in a 26 year old man was reported, which was treated with conservative enucleation of the lesion. There has been no recurrence in this case, after a follow up of 2 years. [14]

A case of palatal pleomorphic adenoma was reported with a rare histological feature of osteoid tissue [15]. Another case of a 9-year-old Japanese girl with pleomorphic adenoma of the buccal mucosa was described [16].

We have kept the patient under regular follow up and there is no recurrence noted after 2-years.

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Conflict of interests None

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