Dental Research Journal

Case Report

Pleomorphic adenoma of the cheek in a child: A case report

Kalenahalli Jagadishkumar[1], Mathod Ganeshrao Anilkumar[2], Halasahalli Chowdegowda Krishna Kumar[2], Rangaswamy Maggad[3]

[1]Departments of Pediatrics, [2]Pediatric Surgery, [3]Pathology, JSS Medical College, JSS University, Mysore, Karnataka, India

Received: January 2013
Accepted: July 2013

Address for correspondence:
Dr. K. Jagadishkumar,
Department of Pediatrics,
JSS Medical College,
JSS University, Mysore,
Karnataka, India.
E-mail: jagdishmandya@gmail.com

ABSTRACT

Salivary gland tumors are rare in children and, when they do arise, they mainly affect the major salivary glands. Minor salivary gland tumors are rare in children and are responsible for less than 10% of the cases. Pleomorphic adenoma is the most common tumor of the salivary glands. The most common sites of pleomorphic adenoma of the minor salivary glands are the palates, followed by the lips and the cheeks. Pleomorphic adenoma of the cheek is rare in children and only few cases have been reported so far.

Key Words: Cheek, children, pleomorphic adenoma

INTRODUCTION

Salivary gland tumors (SGT) are uncommon and account for 1% of all head and neck neoplasms. SGT are more common in adults.[1] Minor salivary gland tumors (MSGTs) are unusual, accounting for only 15-20% of all SGTs.[2] In a study by Vaidya *et al*., of 104 minor SGT, there was no gender predilection and the median age of presentation was 45 years, with a range of 11-74 years.[2] Only 0.32-5% of all salivary gland tumors occur in children aged less than 16 years.[1,3] About 90% of pleomorphic adenomas (PAs) occur in the parotid gland and 10% in the minor salivary glands.[4,5] SGT are rare in children and affect the major salivary glands. MSGTs are rare in children. They are responsible for only 5-10% of all salivary tumors under 20 years of age.[4-6] The most common sites of PAs of the minor salivary glands are the palate, followed by the lips and the cheeks.[3,6,7] Intraoral PA arising from the mucosa of the cheek is very rare, and most of them have been reported in adults.[5,8-10] We report a case of PA from the buccal mucosa in a 9-year-old female child.

Access this article online

Website: http//:drj.mui.ac.ir

CASE REPORT

A 9-year-old girl presented to the pediatric outpatient department of the JSS Hospital, Mysore, with a swelling in the right cheek of 4 months duration. There was no history of trauma. This was a painless swelling and did not bleed. She did not have fever. On oral examination, there was a firm, globular swelling measuring about 2 cm × 2 cm in the posterior part of the buccal mucosa opposite the 2nd right maxillary molar tooth. The swelling was non-tender and mobile and had a smooth surface. Her oral hygiene was good and there was no dental caries. The cervical lymph nodes were not enlarged. The tumor was just underneath the mucosa and away from the parotid gland. The tumor was removed totally along with a margin of normal tissue with wide excision by the intraoral route under general anesthesia. Macroscopically, it was a grey white globular tissue mass measuring about 2 cm × 2 cm [Figure 1]. Microscopic analysis showed a tumor with ductal cells arranged in sheets, glandular and acinar pattern in a background of abundant fibromyxoid areas — features consistent with the features of PA [Figure 2]. The patient was followed-up for 1 year without any problems.

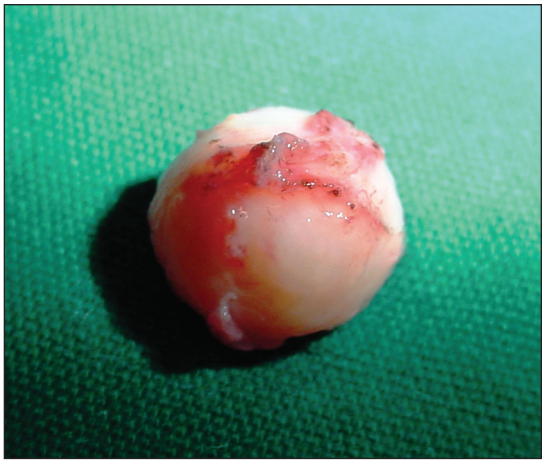

Figure 1: Photograph showing the resected specimen of the pleomorphic adenoma

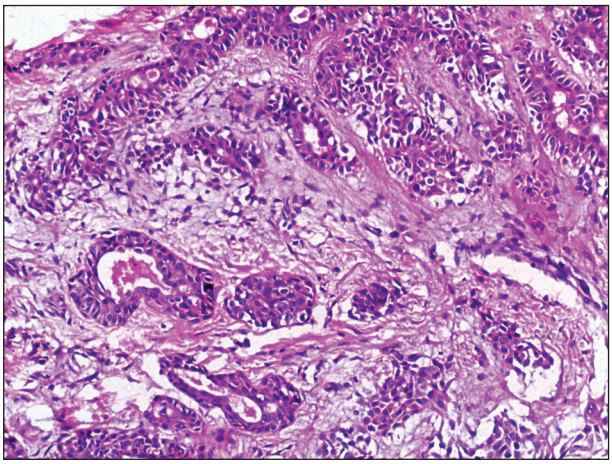

Figure 2: Histopathology of the resected specimen showing tumor with ductal cells arranged in sheets, glandular and acinar patterns in a background of abundant fibromyxoid areas

DISCUSSION

PA is the most common salivary gland tumor accounting for 40-70% of all major and minor SGTs and occurs mainly in the parotid gland.[6] They are commonly seen in 4th-6th decades of life, with a female predominance.[5,11] The minor salivary glands are widely dispersed in the upper respiratory tract, including the lip, palate, pharynx, larynx and nasopharynx. Maximum densities of glands are in the palate. In a study by Vaidya *et al.*, of the 104 MSGTs, 87 were malignant while 17 were benign tumors. All these 17 benign MSGTs were PAs.[2] Intraoral SGT are rare in children and are responsible for only 5-10% of all SGTs.[4,6] The palate is the most common site of MSGTs, and is followed by the lips and cheek.[3,5,11] In a study of pediatric oral lesions from Thailand, a large number of lesions belonged to the cystic category (35%), followed by the inflammatory (34%) and tumor/tumor-like categories (25%). PA was seen only in two cases out of the 1251 oral biopsies performed on their patients.[12] Bentz *et al.* analyzed 324 salivary gland masses in children and observed that 13.3% were SGTs and 21% were from the minor salivary glands, with the palate being the most common site.[1] Krolls *et al.* reviewed 430 cases of salivary gland lesions in children and found 11 PA of the minor salivary glands.[13] But, site, gender and ages were not mentioned.[13] However, there are only few case reports of PA of the minor salivary glands in children.[5-9] Jorge *et al*. reported five cases of PA of the minor salivary glands in children, and all of them were aged between 11 and 18 years. Of the five cases, two were affected on the lips and two on the palate and the tongue was involved in one case.[6] To the best of our knowledge, only few cases of PA from the buccal mucosa in children have been reported.[5,7-9] The tumors are named PA because of the epithelial and connective tissue components in varying degrees. PA from the minor salivary glands lack a capsule. PA is histologically characterized by a variety of tissues consisting of epithelial cells arranged in a cord-like cell pattern with a plasmacytoid appearance. Myoepithelial cells are responsible for the production of abundant extracellular matrix with chondroid, collagenous, mucoid and osseous stroma. Both epithelial and mesenchymal elements arise from the same cell clone, which may be myoepithelial or ductal reserve cell.[5] Among the intraoral SGTs, PA is the most frequently encountered lesion and is predominantly seen in females. The common presentation of intraoral SGT in children is a submucosal lump. PA usually presents as a mobile, slow-growing, painless mass; at times, it may grow rapidly, especially in the palate.[4] Painless intraoral swelling is the most common symptom.[7] A sudden increase in size may be indicative of infection, hemorrhage or malignant transformation. However, few cases of ulceration and bleeding have been reported.[1] Usually, the size of the PA ranges from 0.8 cm to 5 cm. Most of the lesions are asymptomatic; therefore, there will be a large interval between the appearance of the lesion and the diagnosis, ranging from a few days to 15 years.[6,8] Most of the PA from the minor salivary glands in children was noted between the ages of 10 and 18 years.[6,8,11] PA should be considered in the differential diagnosis of mass of the cheek in youngsters.[7] Differential diagnosis of

intraoral, solid, asymptomatic swelling include SGT, neurofibroma, rhabdomyosarcoma, lipoma, dermoid cyst, foreign body reaction, mucocele, buccal space abscess, fibroma, adenoid cystic carcinoma and mucoepidermoid carcinoma.[4-8] In our case, the lack of inflammatory signs, ulceration, pain and invasion ruled out infection and malignancy. The surgical treatment of PA is complete wide excision with good safety margins to prevent recurrence.[6] Jorge *et al*. treated all five juvenile PA surgically and, after a long follow-up period, there was no recurrence. He opined that PA behavior in children is similar to that of adults, with a low recurrence rate after complete surgical resection.[6]

CONCLUSION

To conclude, this case report alerts the clinicians about the unusual causes of intraoral swellings. As the swelling is usually asymptomatic, it may be discovered on routine oral examination. PA of the cheek is rare and should be considered in the differential diagnosis of intraoral swellings of the buccal mucosa.

REFERENCES

1. Bentz BG, Hughes CA, Lüdemann JP, Maddalozzo J. Masses of the salivary gland region in children. Arch Otolaryngol Head Neck Surg 2000;126:1435-9.
2. Vaidya AD, Pantvaidya GH, Metgudmath R, Kane SV, D'Cruz AK. Minor salivary gland tumors of the oral cavity: A case series with review of literature. J Cancer Res Ther 2012;8:111-5.
3. Dhanuthai K, Sappayatosok K, Kongin K. Pleomorphic adenoma of the palate in a child: A case report. Med Oral Patol Oral Cir Bucal 2009;14:73-5.
4. Lotufo MA, Júnior CAL, de Mattos JP, França CM. Pleomorphic adenoma of the upper lip in a child. J Oral Sci 2008;50: 225-8.
5. Dalati T, Hussein MR. Juvenile pleomorphic adenoma of the cheek: A case report and review of literature. Diagn Pathol 2009;4:32-8.
6. Jorge J, Pires R, Alves FA, Perez DE, Kowalski LP, Lopes MA, *et al*. Juvenile intraoral pleomorphic adenoma: Report of five cases and review of the literature. Int J Oral Maxillofac Surg 2002;31:273-5.
7. Gupta MK, Bailoor DN, Mhaske S, Raghuvanshi V, Nahar S, Raju Ragavendra T. Juvenile Pleomorphic Adenoma of the Cheek — A rare case with review of literature. IOSR J Dent Med Sci 2013;6:45-8.
8. Yamamoto H, Fukumoto M, Yamaguchi F, Sakata K, Oikawa T. PleomorphicAdenoma of the buccal gland in a child. Int J Oral Maxillofac Surg 1986;15:474-7.
9. Cohen MA. Pleomorphic adenoma of the cheek. Int J Oral Maxillofac Surg 1986;15:777-9.
10. Ananthaneni A, Undavalli SB Juvenile cellular pleomorphic adenoma. BMJ Case Rep 2013. pii: bcr2012007641. doi: 10.1136/bcr-2012-007641.
11. Savithri V, Sudha S, Shameena PM, Ipe VV. Juvenile pleomorphic adenoma. J Oral Maxillofac Pathol 2004;8:94-5.
12. Dhanuthai K, Banrai M, Limpanaputtajak S. A retrospective study of paediatric oral lesions from Thailand. Int J Paediatr Dent 2007;17:248-53.
13. Krolls SO, Trodahl JN, Boyers RC. Salivary gland lesions in children: A survey of 430 cases. Cancer 1972;30:459-69.

How to cite this article: Jagadishkumar K, Anilkumar MG, Krishna Kumar HC, Maggad R. Pleomorphic adenoma of the cheek in a child: A case report. Dent Res J 2014;11:522-4.

Source of Support: Nil. **Conflict of Interest:** None declared.