SAGE Open Medical Case Reports
Volume 11: 1–4

DOI: 10.1177/2050313X231180374
journals.sagepub.com/home/sco
Sage

A juvenile pleomorphic adenoma of the palate

Bouthaina Hammami[1,2], Imen Achour[1,2], Ghada Yousfi[1,2], Omar Walha[1,2], Malek Mnejja[1,2], Mohamed Amin Chaabouni[1,2] and Ilhem Charfeddine[1,2]

Abstract

Salivary gland tumors in children are rare; involvement of accessory salivary glands is exceptional. We report a case of pleomorphic adenoma of the palate in a child (an 8-year-old girl) addressed by her dentist for discovering a swelling in the palate. Clinical examination revealed a firm, non-tender, nodular swelling in the left hard palate, measuring 1.5 cm × 1.5 cm, situated adjacent to the upper left second molar. Physical examination did not show signs of inflammation or surface ulceration. Oral cavity computed tomography scan did not show bone lysis. The tumor was removed with negative margins. No recurrence was noted. We aim to describe the clinical, radiological features, as well as the management of this rare localization of pleomorphic adenoma.

Keywords

Pleomorphic adenoma, palate, child, minor salivary glands

Date received: 23 April 2022; accepted: 19 May 2023

Introduction

Benign salivary tumors are rare during childhood; only 3%–5% of all salivary gland neoplasms occur in children and adolescents.[1] Pleomorphic adenomas (PAs) are the most common benign neoplasms in salivary glands. Between 70% and 85% of PAs occur in the parotid gland, while 5% occur in the minor salivary glands.[2] The palate is the most commonly affected site among minor salivary glands, but other intraoral sites were reported such as the upper and lower lips, buccal mucosa, gingiva, and tongue.[3,4] We report a case of PA of the palate that occurred in a 12-year-old child in order to discuss the clinical features, treatment, and prognosis of this unusual localization.

Case report

A 12-year-old girl, without a remarkable medical or surgical history, was referred to our department by her dentist for the discovery of a palatal swelling that had been evolving for 2 months. The patient did not report other complaints.

Clinical examination revealed a firm, non-tender, nodular swelling in the left hard palate, measuring 1.5 cm × 1.5 cm, situated adjacent to the upper left second molar. Physical examination did not show signs of inflammation or surface ulceration (Figure 1). Dental examination was normal. There were no enlarged cervical lymph nodes.

A computed tomography (CT) scan of the head and face was performed with axial and coronal slides and contrast injection; it showed a regular, well-limited tumor in the left soft palate, measuring 1.6 cm, exerting a mass effect on the hard palate without perforation of the under lying bone (Figure 2).

The patient underwent resection of the mass under general anesthesia. A palatal incision was raised from the upper left second premolar to the upper left second molar. During the operation, the capsule of the tumor was thin and discontinuous. The lump was excised alongside with the capsule

[1]Department of Otolaryngology – Head and Neck Surgery, Habib Bourguiba Hospital, Sfax, Tunisia
[2]University of Sfax, Sfax, Tunisia

Corresponding Author:
Ghada Yousfi, Department of Otolaryngology – Head and Neck Surgery, Habib Bourguiba Hospital, El Ain Street Km 0.5, Service ORL, 3029 Sfax, Tunisia.
Email: ghada.yousfi.orl@gmail.com

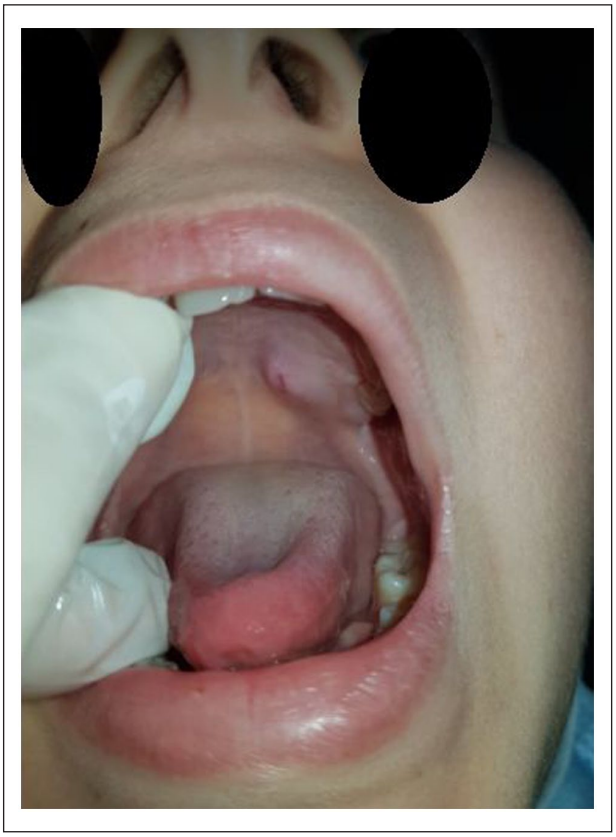

Figure 1. A swelling at the left hard palate.

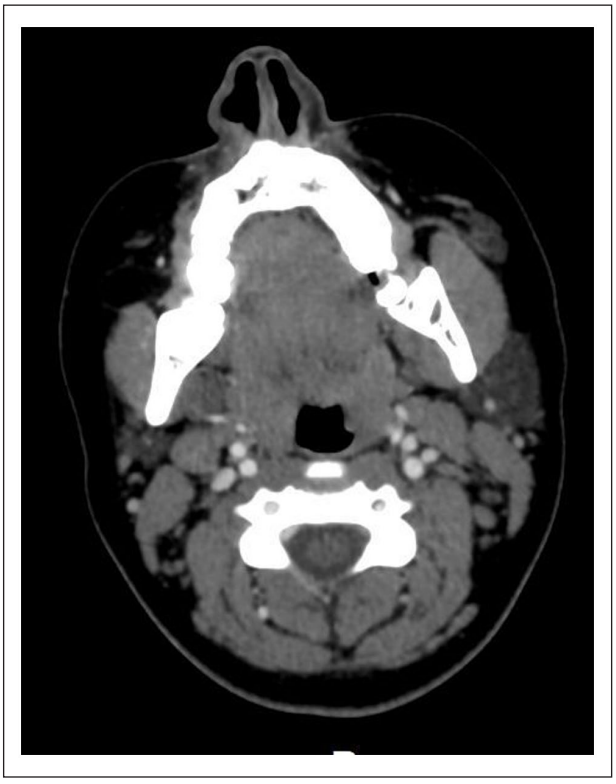

Figure 2. Facial CT scan, axial contrast-enhanced slide showing the absence of bone lysis.

and the overlying mucosa. The defect was allowed to granulate (Figure 3).

The histopathologic examination of the specimen revealed a glandular epithelium and myoepithelial cells with a mesenchymal-like background with a thin intact capsule. The diagnosis of PA without signs of malignancy was retained.

The patient was controlled 2 weeks postoperatively; we noted a good wound healing (Figure 4).

After a regular follow-up period of 1 year, we did not note any sign of recurrence.

Discussion

Juvenile PA of the palate is an unusual disorder. According to Ritwik and Brannon,[1] 28.6% of PA arising from minor salivary glands in children occur at the first decade of life, with a female gender predilection of 2.8:1. The hard and/or soft palate is affected in 69.1% of cases.[1]

PA usually presents as a painless, slow-growing tumor.[5] The duration of symptoms evolution is variable; according to Honghai, it ranges from 20 days to 4 years.[6]

In most cases, palatal PA presents as an asymptomatic, firm bulge in the palatal mucosa. A few cases with ulceration and bleeding, usually resulting from trauma during mastication, have been reported. It should be noted that PA tends to be small and fixed in the palate as well as in the other minor salivary glands compared to the parotid gland where the lesion rather tends to be larger and mobile.[2,7,8] In this localization, tumors are mainly seen at the junction of the hard and soft palates.[5] In our case, the tumor was asymptomatic, accidentally discovered by a dentist and localized in the hard palate. For the size of the tumor, a study of 74 palatal PAs in adults and children showed an average size of 1.9 cm, and the tumors size ranged from 1 to 4 cm.[9]

Magnetic resonance imaging (MRI) and CT are recommended in salivary glands tumors; CT is more commonly used in the diagnosis of minor salivary gland PA. For palatal PA, coronal view scans are recommended. They are used to determine the size of palate lesions as well as to detect any bony involvement. But this technique exposes the child to ionizing radiation; its role in the characterization of soft tissue is also limited.[7,8]

Alsufyani[8] reported a case of an incidental finding of palatal PA in the cone beam CT, due to pressure resorption of the hard palate, without break in the floor of the nasal cavity. MRI provides excellent soft tissue characterization and does not expose patients to any radiation, but it cannot evaluate the bone involvement in the case of a palatal localization.[7]

Ultrasound has limited indication in palate lesion. For other minor salivary glands lesions, it is only used for buccal and parapharyngeal space masses.[7]

Fine-needle aspiration can provide up to an 80% accuracy in the diagnostic of salivary glands tumor, but young patients may not tolerate this procedure without sedation, making it a less favorable diagnostic tool than no invasive imaging techniques.[3,6]

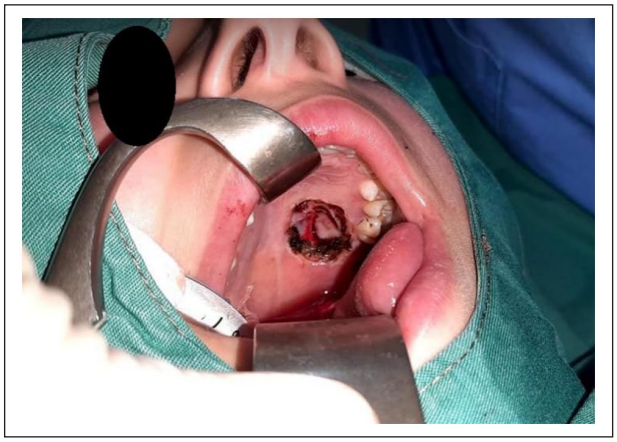

Figure 3. A total resection off the mass with the overlying mucosa.

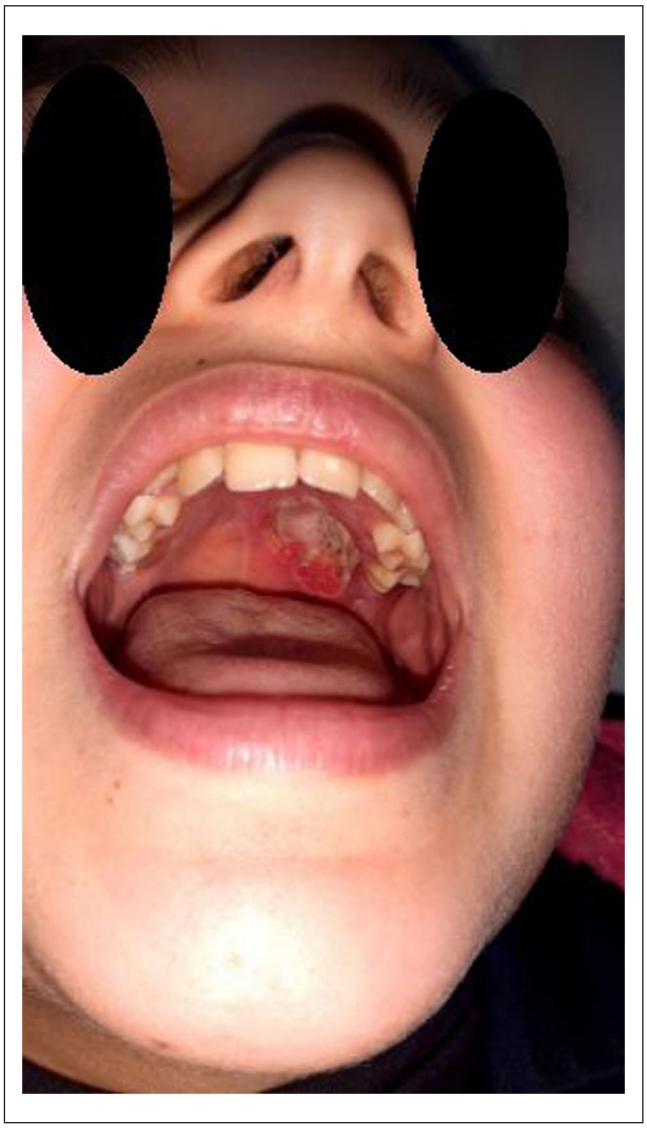

Figure 4. A good wound healing with granulating mucosa.

Surgical treatment of juvenile palatal PA is the same as in adults. It involves a wide local tumoral excision including the capsule and involved surrounding bone removal to avoid recurrence. According to Moon,[10] in the cases of an ulcerated overlying mucosa or a capsular infiltration, or in the case of the presence of a thin capsule at biopsy prior to surgery, the overlying mucosa should be excised in order to prevent recurrence. The defect can be repaired with a local flap or allowed to granulate.[10] In our case, the capsule was thin and discontinuous. Consequently, mucosal resection was performed with safety margin.

When substance loss is significant, to reduce postoperative hematoma and ensure that the palatal flap is closely approximated to the palate, fabricated palatal splints can be used.[3] Recurrence and carcinoma arising from PA are the main risk of PAs.[11] According to a literature review published by Daniels et al.,[3] the recurrence rate following PA excision is 12.5%. In one case, recurrence is observed after 5 years.[3] The risk of recurrence and malignant transformation are higher in children: that may be explained by the smaller anatomy, the tendency toward conservative surgery, and the longer life expectancy in pediatric population.[5] In the systematic review of Alsufyani et al,[5] the rate of carcinoma in recurrent PAs ranged from 1.5% to 23%.[5] Moon[10] reported one case of recurrence of carcinoma ex adenoma pleomorphic; per operatively, a tumoral capsular rupture was noted.[10] Therefore, a complete resection with safe margins and prolonged as well as regular appropriate clinical monitoring are required, to improve the prognostic of these tumors.

Conclusion

PA of minor salivary glands is rare in pediatric population; palate is the most common localization. Imaging, especially CT scan is important for planning surgical treatment. Complete resection with negative margins is the best option to avoid recurrence. Nevertheless, patients and doctors should remain vigilant for any recurrence that may occur in the follow-up period which must be extended to 5 years at least.

Author contributions

Collecting of the data: B.H. and G.Y. Writing: G.Y. and O.W. Reviewing: I.A., M.M., M.A.C., and I.C.

Declaration of conflicting interests

The author(s) declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

Funding

The author(s) received no financial support for the research, authorship, and/or publication of this article.

Ethical approval

Our institution does not require ethical approval for reporting individual cases or case series.

Informed consent

Written informed consent was obtained from a legally authorized representative(s) for anonymized patient information to be published in this article.

ORCID iDs

Imen Achour https://orcid.org/0000-0002-9511-2392
Ghada Yousfi https://orcid.org/0000-0001-6536-2289
Mohamed Amin Chaabouni https://orcid.org/0000-0003-0880-1032

References

1. Ritwik P and Brannon RB. A clinical analysis of nine new pediatric and adolescent cases of benign minor salivary gland neoplasms and a review of the literature. *J Med Case Rep* 2012; 6: 287.
2. Shaaban H, Bruce J and Davenport PJ. Recurrent pleomorphic adenoma of the palate in a child. *Br J Plast Surg* 2001; 54(3): 245–247.
3. Daniels JSM, Ali I, Al Bakri IM, et al. Pleomorphic adenoma of the palate in children and adolescents: a report of 2 cases and review of the literature. *J Oral Maxillofac Surg* 2007; 65(3): 541–549.
4. Dhanuthai K, Sappayatosok K and Kongin K. Pleomorphic adenoma of the palate in a child: a case report. *Med Oral Patol Oral Cir Bucal* 2009; 14(2): E73–E75.
5. Alsufyani NA, Altowaijri AA, Alshehri BM, et al. Systematic review of clinical and radiographic signs of pediatric pleomorphic adenoma of minor salivary glands. *J Contemp Dent Pract* 2021; 22(9): 1063–1068.
6. Fu H, Wang J, Wang L, et al. Pleomorphic adenoma of the salivary glands in children and adolescents. *J Pediatr Surg* 2012; 47(4): 715–719.
7. Dombrowski ND, Wolter NE, Irace AL, et al. Pleomorphic adenoma of the head and neck in children: presentation and management. *Laryngoscope* 2019; 129(11): 2603–2609.
8. Alsufyani N. Incidental cone beam CT finding of juvenile pleomorphic adenoma. *Case Rep Dent* 2020; 2020: e8862657.
9. Wu YC, Wang YP, Cheng SJ, et al. Clinicopathological study of 74 palatal pleomorphic adenomas. *J Formos Med Assoc* 2016; 115(1): 25–30.
10. Moon SY. Surgical management of the palatal pleomorphic adenoma. *J Craniofac Surg* 2019; 30(6): e580–e582.
11. Pramod Krishna B. Pleomorphic adenoma of minor salivary gland in a 14 year old child. *J Maxillofac Oral Surg* 2013; 12(2): 228–231.