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Case report

Rapidly progressing palatal pleomorphic adenoma in an adolescent

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ABSTRACT

Minor salivary gland neoplasms are rare in children. Only 5% of pleomorphic adenomas of the minor salivary glands have been reported in patients under 20 years of age. We report a case of a 13-year-old female who presented with a rapidly growing hard palatal mass, confirmed to be a myoepithelial cell-rich pleomorphic adenoma with cytological atypia. Pleomorphic adenomas should be included in the differential diagnosis of rapidly growing palatal masses in children and adolescents. Surgical excision is curative, and long-term follow-up is indicated as recurrence is frequent.

1. Introduction

Minor salivary gland neoplasms are rare in children [1]. The most common minor salivary gland tumor is pleomorphic adenoma of the hard palate [2]. In a recent review by Daniels, only 5% of pleomorphic adenomas of the minor salivary glands have been reported in patients aged 20 years or younger [3]. We report below a case of a rapidly progressing pleomorphic adenoma of the palate in an adolescent along with a literature review of this condition.

2. Case report

A 13-year-old female of Asian descent was referred to us for a rapidly growing hard palatal mass over a 2-week period. The patient had an unremarkable past medical history. She had no family history of cancer and denied weight loss, fever, or trauma to the palate.

Examination revealed a submucosal smooth nontender firm mass on the right posterior palate. An infused CT scan revealed a nonenhancing isodense 2.0 cm × 2.0 cm × 2.3 cm soft tissue mass at the right posterior palate, with mild scalloping of the hard palate (Fig. 1). There was no neck adenopathy. MRI showed an enhancing mass with a small area of calcification, and slight indentation of the maxilla without disruption of the palate (Fig. 3). A biopsy revealed myoepithelial cell-rich pleomorphic adenoma with cytological atypia.

A right hemipalatectomy was performed with excision of the deep bony margin. Multiple sequential frozen sections of the margins were sent to the pathologist until confirmed negative, an additional resection margin was also taken.

The histopathology report noted myoepithelial cell-rich pleomorphic adenoma with cytological atypia and four margins negative for neoplasia (Fig. 2).

3. Discussion

A review of the current English literature yielded only 16 cases of palatal pleomorphic adenoma involving patients under the age of 18 years [3–14] (Table 1). There were 9 females and 7 males aged 5–17 years, and 75% of patients were under the age of 14 years. In the Japanese literature, a review by Yamamoto et al. [15] reported 8 cases of juvenile palatal pleomorphic adenoma in patients aged 18 years and younger. These cases included 7 females and 1 male.

Palatal pleomorphic adenomas typically present as painless, slow-growing tumors. However few authors have described rapidly growing palatal pleomorphic adenomas such as in our current patient [7,12,14]. Lopez-Cedrun et al. [12] reported a case in a 16-year-old male which was only noted 2 weeks before presentation. de Courten et al. [7] reported a case in a 10-year-old female of 5–6 weeks duration. Shaaban et al. [14] reported a case in

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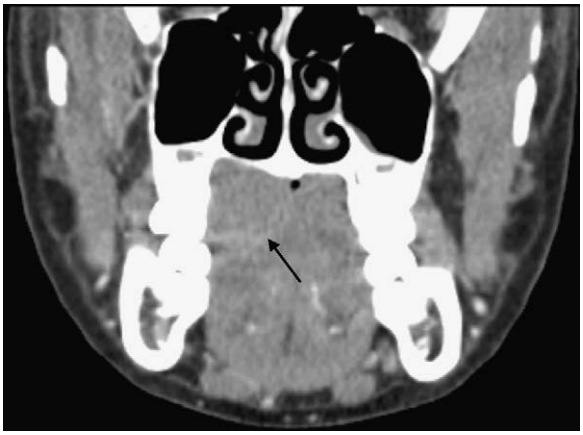

Fig. 1. Facial infused CT scan.

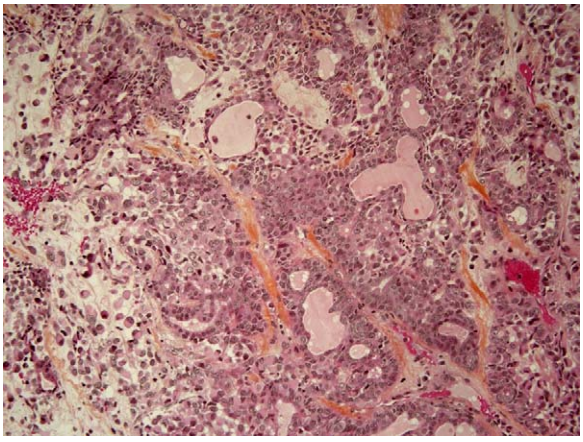

Fig. 2. Myoepithelial cell-rich pleomorphic adenoma with cytological atypia.

a 9-year-old male patient with a 4-day history of painless swelling over the palate.

Plain X-ray and hematologic investigations play no part in the diagnosis of salivary gland tumors of the palate [16]. CT is superior to MRI in evaluating erosion and perforation of the bony palate, or involvement of the nasal cavity or maxillary sinus [13,16]. MRI provides better definition of the vertical and inferior tumor extension [13], and more accurately indicates the degree of encapsulation [16]. MRI is also advantageous because of the absence of exposure to radiation and intravenous contrast medium [3].

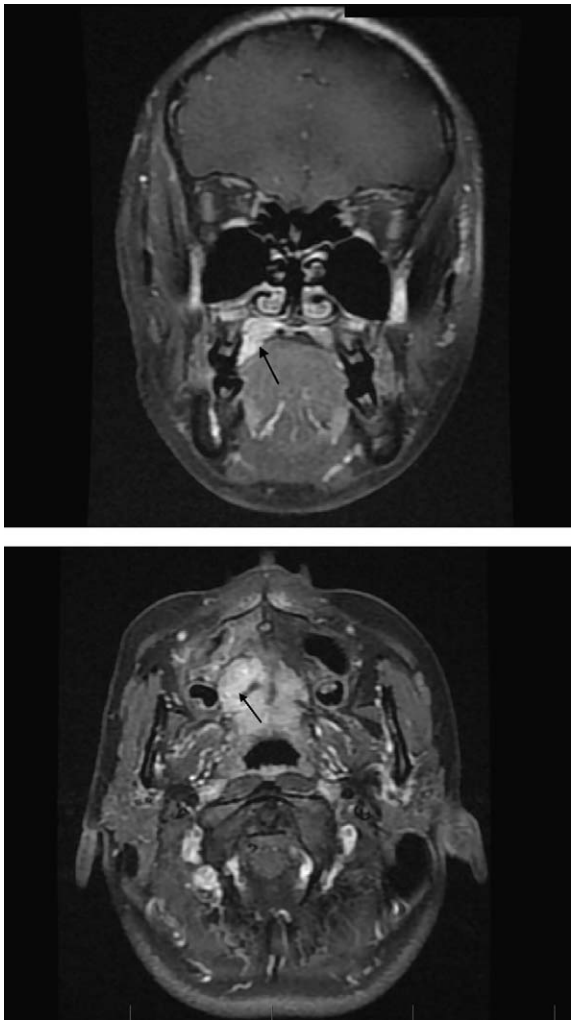

Fig. 3. Facial infused MRI.

A histological diagnosis is essential to plan the definitive management [16]. Treatment consists of wide local excision with clear margins involving the periosteum and associated mucosa, followed by curettage of the underlying bone with a curette or bur under copious sterile normal saline irrigation [3]. The overlying mucosa can sometimes be repaired using a local flap [14]. In our case, the patient did not require reconstruction as the palatal

Table 1
Previous reports of pleomorphic adenomas of the palate in children and adolescents.

Author	Age (years)	Gender	Size (cm)	Follow-up (years)	Recurrence
Byars et al. [5]	7	F		28	5 years
Byars et al. [5]	9	F		7	None
Crawford and Guernsey [6]	8	F	2.5	1	None
Galich [9]	12	F		0.5	None
Lack and Upton [11]	10	M	2	5	None
Fonseca et al. [8]	8	F			
Fonseca et al. [8]	16	F		1	None
Austin and Crockett [4]	10	M	2×3	1	None
Noghreyan et al. [13]	8	F	2.5×3	1.5	None
Lopez-Cedrun et al. [12]	16	M	5×5	3.5	None
de Courten et al. [7]	10	F	2.3×2	9	None
Shaaban et al. [14]	9	M	2×2	3	2 years
Jorge et al. [10]	11	M	3	9	None
Jorge et al. [10]	17	F	3	23	None
Daniels et al. [3]	5	M	2×1.5	3.8	None
Daniels et al. [3]	16	M	2×1	4	None
Our case (2009)	13	F	$2 \times 2 \times 3$	1	None

M: male; F: female.

mucosa regenerated. A small oro-antral fistula closed spontaneously.

Pleomorphic adenoma is encapsulated, and incomplete excision will leave residual tumor cells behind and result in recurrence, because of its high rate of implantability [3]. Recurrence after surgical treatment has only been reported in 2 of the 16 published cases of pleomorphic adenoma in children and adolescents, one of which recurred 5 years after excision [5], and another recurred 2 years after initial excision [14]. In the second case [14], the pleomorphic adenoma had been re-excised 2 weeks after an initial excisional biopsy with positive margins. However, another case of palatal pleomorphic adenoma having been re-excised 3 weeks after initial positive margins did not recur during the 5 years of follow-up [11]. Our patient has not experienced any recurrence after one year of follow-up.

4. Conclusion

We have reported a case of rapidly growing juvenile palatal pleomorphic adenoma. Surgical resection is curative. Long-term follow-up is warranted because of the increased risk of recurrence even several years after initial excision.

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