Case report

Rapidly progressing palatal pleomorphic adenoma in an adolescent

Sami P. Moubayed, Fahad AlSaab, Sam J. Daniel*

Department of Otolaryngology, Head and Neck Surgery, Montreal Children’s Hospital, McGill University, Montreal, QC, Canada

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. 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.

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.

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

* Corresponding author at: Otolaryngology, Head and Neck Surgery (B-240), McGill University Health Care Centre, Montreal Children’s Hospital, 2300 rue Tupper, Montreal, QC H3H IP3, Canada. Tel.: +1 514 776 6789; fax: +1 514 412 4342.
E-mail address: sam.daniel@muhc.mcgill.ca (S.J. Daniel).
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].

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>
<thead>
<tr>
<th>Author</th>
<th>Age (years)</th>
<th>Gender</th>
<th>Size (cm)</th>
<th>Follow-up (years)</th>
<th>Recurrence</th>
</tr>
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<tr>
<td>Byars et al. [5]</td>
<td>7</td>
<td>F</td>
<td>28</td>
<td>5 years</td>
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<td>2</td>
<td>5</td>
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</tr>
<tr>
<td>Fonseca et al. [8]</td>
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<td>F</td>
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<tr>
<td>de Courten et al. [7]</td>
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<td>2.3 × 2</td>
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<tr>
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<td>2 years</td>
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M: male; F: female.
mucosa regenerated. A small oro-antral fistula closed sponta-
neously.

Pleomorphic adenoma is encapsulated, and incomplete exci-
sion 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.

References

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