Case Report: Rare site for intraoral meningioma [version 1; peer review: 1 approved with reservations]

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Abstract

Extracranial meningioma is very rare with few cases reported, especially in the oral cavity. Its diagnosis considered a challenge owing to the unusual site of occurrence. We report, to our knowledge, the first case of extra-cranial meningioma as a primary tumor in the palate with no detected intracranial extension. A 59-year-old female Egyptian patient presented with a 22-year history of a large painless swelling at the right side of the palate, which could not be seen on radiographs. An incisional biopsy was taken and, after assessment with a panel of immunohistochemical markers, the lesion was diagnosed as extracranial meningioma. The patient did not show up for surgical excision and follow-up was not performed because of loose of contact with the patient. Intraoral meningioma is a rare unsuspected tumor. Immunohistochemical markers are important when confirming this diagnosis.

Keywords

Intra-oral meningioma, Benign tumor, Ectopic meningioma, Palatal lesion

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Introduction

Meningioma is a benign neoplasm of meningothelial cells. Meningioma may develop as a direct extension of a primary intra-cranial meningioma or as a true primary extra-cranial meningioma.

Extra-cranial (ectopic) tumors are mostly seen in the head and neck region with no connection intra-cranially. The most common extra-cranial site is the orbits. Meningioma arising in the oral cavity is extremely rare. To the extent of our knowledge, 18 cases have currently been reported in the oral cavity and we are reporting the first case in the palate.

Case report

A 59-year-old female patient presented to the outpatient clinic in the Oral and Maxillofacial Surgery Department, Cairo University in January 2019 complaining of a large painless swelling in the palate (Figure 1). The patient reported that the swelling had been present in her oral cavity for 22 years. The patient’s medical and familial histories were unremarkable. Upon clinical examination the day of admission, a large palatal swelling (3 cm × 3 cm) was evident on the right side of the hard palate. The swelling was covered by normal mucosa and showed a slight bluish tinge. The lesion was not visible on radiographs.

An incisional biopsy of the lesion was performed. Hematoxylin and eosin stained sections revealed meningothelial cells arranged in lobules. The cells exhibited round to oval nuclei (Figure 2). Psammoma bodies were also present (Figure 3). No mitotic activity and no cellular atypia were found. Immunohistochemical staining for tumor-associated markers was performed to confirm a diagnosis of meningioma diagnosis and to exclude other mimic tumors as metastatic carcinomas, schwannoma, neurofibroma, paraganglioma and perineurioma. Cells were positively stained using primary antibodies for epithelial membrane antigen (EMA) and vimentin (Figure 4a, b), but were not stained when using primary antibodies for S100, pancytokeratin, p63, chromogranin and renal cell carcinoma glycoprotein (Figure 5a–e).

No therapy was administered to the patient during her admission. Unfortunately, the patient did not show up for surgical excision and follow-up.

Discussion

Primary extra-cranial meningioma is an unusual tumor, especially in the oral cavity. The first intraoral meningioma reported was by Brown et al. in 1976, which presented as a periapical radiolucency anterior maxillary region.

To the extent of our knowledge, 18 cases of primary meningioma in the oral cavity have been reported. Of these, 13 were in female patients, which is also true of the present case. However, the age range was large in the reported cases.
Figure 5. Meningioma tumor cells react negatively following immunohistochemical staining for (a) renal cell carcinoma glycoprotein, (b) S100, (c) chromoginin, (d) p63, (e) PanCK (magnification, ×100).

Table 1. Clinicopathological and radiographic data of the documented cases of extracranial meningioma.

<table>
<thead>
<tr>
<th>Study</th>
<th>Age, years</th>
<th>Gender</th>
<th>Site</th>
<th>Tumor size</th>
<th>Radiographic findings</th>
<th>Treatment</th>
<th>Follow-up</th>
</tr>
</thead>
<tbody>
<tr>
<td>Brown et al.</td>
<td>69</td>
<td>M</td>
<td>Maxilla</td>
<td>NA</td>
<td>ML RL</td>
<td>Not completed</td>
<td>8 years</td>
</tr>
<tr>
<td>Simpson and Sneddon</td>
<td>63</td>
<td>F</td>
<td>Maxillary alveolus</td>
<td>4.5 × 2.7 × 2.7 cm</td>
<td>Well-defined mixed RL RO</td>
<td>Surgical excision.</td>
<td>Under review</td>
</tr>
<tr>
<td>Landini and Kitano</td>
<td>48</td>
<td>F</td>
<td>Mandible</td>
<td>NA</td>
<td>Well-defined RL</td>
<td>Block resection</td>
<td>2 years</td>
</tr>
<tr>
<td>Reddi et al.</td>
<td>26</td>
<td>F</td>
<td>Maxilla</td>
<td>3 cm</td>
<td>Ill-defined RL</td>
<td>Surgical excision</td>
<td>2 years</td>
</tr>
<tr>
<td>Pfeifer et al.</td>
<td>77</td>
<td>F</td>
<td>Maxilla (temporal fossa)</td>
<td>NA</td>
<td>Dense soft tissue mass</td>
<td>Surgical resection</td>
<td>NS</td>
</tr>
<tr>
<td>Jones and Freedman</td>
<td>41</td>
<td>F</td>
<td>Mandible</td>
<td>4 × 2 cm</td>
<td>Well defined RL</td>
<td>Excisional biopsy</td>
<td>NS</td>
</tr>
<tr>
<td>Jones and Freedman</td>
<td>74</td>
<td>F</td>
<td>Mandible</td>
<td>4 × 3 cm</td>
<td>Well-defined RL</td>
<td>Excisional biopsy</td>
<td>NS</td>
</tr>
<tr>
<td>Kubota et al.</td>
<td>10</td>
<td>M</td>
<td>Mandible</td>
<td>NA</td>
<td>Well-defined RL</td>
<td>Enucleated</td>
<td>4 years</td>
</tr>
<tr>
<td>Mussak et al.</td>
<td>62</td>
<td>M</td>
<td>Mandible</td>
<td>7 × 3 cm</td>
<td>Well-defined RL</td>
<td>Segmental mandibulectomy</td>
<td>NS</td>
</tr>
<tr>
<td>Loll et al.</td>
<td>40</td>
<td>F</td>
<td>Mandible</td>
<td>NA</td>
<td>Well-defined RL</td>
<td>NS</td>
<td>NS</td>
</tr>
<tr>
<td>Mosque-de-Taylor et al.</td>
<td>53</td>
<td>F</td>
<td>Mandible</td>
<td>4 cm</td>
<td>III-defined mixed RO RL</td>
<td>Surgical excision</td>
<td>6 months</td>
</tr>
<tr>
<td>Rushing et al.</td>
<td>NA</td>
<td></td>
<td>Mandible</td>
<td>NA</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Simsek and Komerik</td>
<td>51</td>
<td>F</td>
<td>Maxilla</td>
<td>2 × 2 cm</td>
<td>III-defined mixed RL-RO</td>
<td>Surgical excision</td>
<td>5 years</td>
</tr>
<tr>
<td>Pinting et al.</td>
<td>59</td>
<td>M</td>
<td>Maxilla</td>
<td>NA</td>
<td>Well-defined RL</td>
<td>Surgical excision and radiotherapy</td>
<td>NS</td>
</tr>
<tr>
<td>Maeng et al.</td>
<td>66</td>
<td>F</td>
<td>Buccal mucosa</td>
<td>2 cm</td>
<td>Heterogenously enhanced mass</td>
<td>Surgical excision</td>
<td>Year and half</td>
</tr>
<tr>
<td>Nair et al.</td>
<td>60</td>
<td>F</td>
<td>Buccal mucosa</td>
<td>4 × 3 cm</td>
<td>Mass of heterogeneous density</td>
<td>Surgical resection</td>
<td>One year</td>
</tr>
<tr>
<td>Rege et al.</td>
<td>35</td>
<td>M</td>
<td>Mandible</td>
<td>NA</td>
<td>III-defined ML RL</td>
<td>Partial resection</td>
<td>5 years</td>
</tr>
<tr>
<td>Rommel et al.</td>
<td>20</td>
<td>F</td>
<td>Mandible</td>
<td>2 × 1.8 cm</td>
<td>Well defined RL</td>
<td>No surgical intervention.</td>
<td>One year</td>
</tr>
</tbody>
</table>

M, male; F, female; RL, radiolucent; RO, radiopaque; UL, unilocular; ML, multicellular; NA, not available; NS, not stated.

10 and 77 years old\textsuperscript{2,4–19}; in the present case, the patient was 59 years old. Regarding the reported cases of intraoral primary meningioma, 6 of the 18 were in the maxilla\textsuperscript{2,4–6,8,10,11}, 10 were in the mandible\textsuperscript{7,9–14,18} and 2 in the buccal mucosa\textsuperscript{16,17}. To our knowledge, we report the first case in the palate.

The histopathological criteria of extracranial meningiomas are similar to those of their intracranial counterparts. All documented cases shared the same characteristics: whorls of spindle cells or epithelioid cell proliferation and psammoma bodies. In our case, diagnosis was challenging because of the tumor’s
similarity with other tumor entities of peripheral nerve origin, as well as the uncommon location of the tumor. An immunohistochemical panel of tumor-associated markers were used to confirm the diagnosis and to avoid unnecessary aggressive treatment. Most of the 18 cases reported in the literature achieved their diagnosis using immunohistochemical markers. All reported cases that used immunohistochemistry techniques to diagnose meningioma\textsuperscript{4,8,10,12,13,15,16,18,19} observed that the tumor cells stained positive for monoclonal antibodies against EMA and vimentin, with no immunoreactivity for S-100 protein, which was similar to our findings.

Unfortunately, our patient did not show up for surgical excision and follow-up was not done because of lose of contact with the patient. However, most of the documented cases was treated successfully without recurrence by surgical excision. Some of studies, such as that by Rommel et al.\textsuperscript{10}, preferred only to follow-up with the patient rather than conduct surgical intervention.

However, others preferred to perform aggressive treatment, such as as segmental mandibulectomy or segmented resection\textsuperscript{7,11}

In conclusion, meningioma is a rare intraoral benign neoplasm. Immunohistochemical markers are an important tool to achieve a final diagnosis, especially for the differentiation from histological mimic entities of peripheral nerve origin, such as perineurioma and neurothekeoma. Vimentin and EMA are the two important markers to confirm extra-cranial meningioma diagnosis from other lesions.

**Data availability**

All data underlying the results are available as part of the article and no additional source data are required.

**Consent**

Written informed consent for publication of their clinical details and clinical images was obtained from the patient.

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**References**


Open Peer Review

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The case report is well written and presents an interesting case of ectopic meningioma. Some amendment is needed however.

1. Radiological findings should be included. The authors merely mention that the lesion was not seen by radiology. Please clarify the technique used and relevant findings.

2. The grade of meningioma was not given

3. The differential diagnosis of meningioma from mimics was not sufficiently addressed. Different entities in the differential diagnosis were mentioned at different parts of the article without discussing the differentiating points. And the performed stains would not help in differentiating meningioma from perineurioma, both are positive for EMA and vimentin.

4. Grammatical and spelling mistakes are noted here.

Is the background of the case’s history and progression described in sufficient detail?
Yes

Are enough details provided of any physical examination and diagnostic tests, treatment given and outcomes?
Partly

Is sufficient discussion included of the importance of the findings and their relevance to future understanding of disease processes, diagnosis or treatment?
Partly

Is the case presented with sufficient detail to be useful for other practitioners?
Partly
Competing Interests: No competing interests were disclosed.

Reviewer Expertise: Neuropathology.

I confirm that I have read this submission and believe that I have an appropriate level of expertise to confirm that it is of an acceptable scientific standard, however I have significant reservations, as outlined above.

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