Ectopic meningioma of the mandible in a 20-year-old woman: a case report and literature review

Niklas Rommel*†, Oliver Bissinger†, Andrea Rau, and Thomas Muecke

Department of Oral and Maxillofacial Surgery, Technische Universität München, Klinikum rechts der Isar, Munich, Germany

*Correspondence address. Department of Oral and Maxillofacial Surgery, Technische Universität München, Klinikum rechts der Isar, Ismaninger Str. 22, D-81675 Munich, Germany. Tel: +49-4140-9744; Fax: +49-89-4140-2934; E-mail: niklasrommel@tum.de

Abstract

Ectopic meningiomas are a very rare tumor entity. We present a case of a meningioma arising in the mandible of a young woman and initially supposed to be a radicular cyst. Histopathological and immunohistochemical evaluation showed typical cell characteristics of a meningioma. Only six cases of ectopic meningiomas in the mandible have been described in the literature until now, mainly in women at an advanced age and with surgical removal of all tumors. For the first time, no surgical excision has been performed in this case and follow-up control after 12 months showed no significant progression or increasing clinical complaints. Hence, surgical removal seems non-urgent. In conclusion, unclear lesions of the jaws, even if they seem to be clear following diagnostics, should be evaluated by incisional biopsy and histopathological evaluation.

INTRODUCTION

Meningiomas are one of the most common neoplasms arising from cellular elements of the meninges, in particular, from arachnoid villi structures. Commonly, meningiomas are attached to the dura and grow in the cranial cavity or intraspinal region. However, ectopic locations of meningiomas cannot be excluded. In this report, we present a rare case of this special tumor entity in the mandibular bone of a young woman.

CASE REPORT

A 20-year-old woman was referred to our Department with the suspicion of a radicular cyst resulting from a periapical infection of tooth 36 in the mandibular bone. The patient reported slight local pain in the left mandible over a long period. On radiographic examination, panoramic X-ray and computed tomography (CT) imaging showed a 2 × 1.8-cm radiolucent lesion of the left mandible. The lesion involved both apices of the first molar and the mesial apex of the second molar and extended to the caudal border of the mandibular bone.
the left posterior mandible. The lesion involved both apical roots of the first molar and the mesial apical root of the second molar. It showed an expansive character and penetrated the medial corticals of the mandibular bone (Figs 1 and 2).

After completion of a discussion of the results, the patient was treated under general anesthesia by intraoral subtotal incisional biopsy by using Piezosurgery (Mectron, Cologne, Germany). Microscopic examination revealed a mesenchymal tumor with fragments of lamellar bone including resorption signs. The tumor had a high cell density including fascicular, storiform and spindle-shaped patterns. The cells were ordered into cell cords, but also into whorls. Additionally, collagenous and hard matter areas were present evoking psammom bodies, and the cells clearly formed reticulin fibers. No mitotic activity and no cell atypia were found (Fig. 3A). Using avidin-biotin complex immunostaining, we found a strong positive reaction with antibodies for vimentin, epithelial membrane antigen (EMA) and somatostatin and also a positive reaction for desmoplakin in a smaller amount of tumor cells (<20%) (Fig. 3B–D). The final histopathological results of the biopsy revealed a mesenchymal tumor classified as an ectopic meningioma WHO grade I.

**DISCUSSION**

Meningiomas are one of the most common tumor entities in the central nervous system, are generally benign and have their origin in the arachnoid villloid structures of the meningocytes. However, in rare cases, ectopic forms of this tumor entity can appear extracranially and extraosseously in the head and neck region. With respect to the jaws, we have found only eight cases including two meningiomas of the maxilla [1, 2] and six meningiomas of the mandible [3–7] in the current literature. We have found the seventh case of an extracranial meningioma of the mandible in a young woman who presented with a cystoid-like lesion in the left mandibular bone and no specific clinical symptoms. Because of the absence of typical radiographic features, no clear diagnosis was possible either with a panoramic X-ray or 3D imaging. However, the role of CT is seen significantly to assess the relationship between the tumor and the bony surfaces and to exclude potential malignancy [8].

Several hypotheses have been proposed for the occurrence of extracranial meningiomas, including extradural enclosing of arachnoid cell nests during embryogenesis, ectopic migration, and the development of arachnoid cells in combination with the peripheral nerves, or metaplasia of the mature peripheral nerve sheath cells or progenitor cells [8]. However, ectopic meningiomas have also been postulated to be mesenchymal tumors that arise from multipotential mesenchymal cells, particularly if no associations to the cranial nerves are apparent [9]. In the head and neck region, this tumor entity is often associated with cranial nerves and, therefore, is considered to be derived from ectopic arachnoid tissue present around these nerves [3]. Therefore, the origin of the tumor arising in our patient seems to be the perineural cells of the mandibular nerve. However, ectopic arachnoid cells within the mandibular bone as a source of tumor growth cannot be excluded.

The histopathologic and immunohistochemical features of ectopic meningiomas are seen similar to those of their more frequent intracranial counterparts [4, 8]. The general histologic diagnosis of meningiomas describes spindle-shaped cells that...
Ectopic meningioma of the mandible in a 20-year-old woman

REFERENCES


Figure 5: Likewise to panoramic X-ray, follow-up CT after 12 months showing no significant progression of the lesion.

The conclusion of this case is that unclear lesions of the jaws, even if they seem to be clear following diagnostics, should be evaluated by incisional biopsy and histopathological evaluation.

CONFLICT OF INTEREST STATEMENT
None declared.

ACKNOWLEDGEMENTS

All authors have read and approved all versions of the manuscript and its content. No ethical approval has been required. The authors would like to thank Theresa Jones for the final language correction of the manuscript.

FUNDING
None.