Adenomatoid Ameloblastoma in the Mandible and Maxilla: Report of a Case

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Abstract Ameloblastoma is an infrequent odontogenic neoplasm located in the mandible and/or infratemporal fossa extending into the maxilla. The authors report a patient who underwent a total of five operative interventions for recurrent lesions involving mandible and infratemporal fossa at other institutions before admission to our hospital. Radiographic imaging in our case demonstrated a solid/multicystic recurrent lesion of the right infratemporal fossa extending into the maxilla. Gross total excision of the tumour was done and the postoperative course was uneventful. In any case with a tumour located in this region, it is important to be aware of this condition because this lesion is unusual but serious pathology as illustrated in this report.

Keywords Ameloblastoma · Odontogenic tumour · Mandible · Maxilla · Subtemporal region

Introduction

Ameloblastoma, a locally aggressive tumour of odontogenic origin, constitutes only 1% of all oral tumours [1–6]. Clinically, it is painless and slow growing lesion, but behaves as invasive recurring tumours in spite of its benign histological appearance [1, 2]. There are three different types of adenomatoid ameloblastoma: solid/multicystic, unicystic, and peripheral [7, 8]. They are the most frequent odontogenic tumours of the mandible and maxilla. In this report, we present a rare case of solid and aggressive recurrent adenomatoid ameloblastoma in the infratemporal fossa extending into the maxilla.

Case History

A 55-year-old man presented with complaints of painful swelling and headache recurrent for 5–6 years. In his past medical history, there was an operation for a cystic lesion in his right lower jaw at another medical center in 1983. Then, he underwent a total of five surgical interventions again for his recurrent tumours located mandible involving the infratemporal fossa and the right zygomatic arc in various institutions. However, the patient began to suffer from severe headache which is aggravated during night and he was admitted to our hospital with these complaints.

Neurological examination revealed a sequela peripheral facial paralysis on the right and hypoesthesia in the second and third branches of the right trigeminal nerve. Computed tomography (CT) showed a low-density mass involving the infratemporal fossa (Fig. 1) and zygomatic process of temporal bone (Fig. 2) extending into the right maxillary sinus (Fig. 3). Pre- and post-contrast magnetic resonance imaging (MRI) revealed a recurrent tumour mass with well-defined border at the inferior temporal region and the posterior aspect of right maxilla (Fig. 4).

Under general anesthesia, the patient underwent a surgical intervention consisting of gross total removal of the tumour involving the surrounding muscle tissue via modified infratemporal fossa approach. The histopathological finding indicated solid/multicystic adenomatoid ameloblastoma (Fig. 5). Histologically, there was a follicular type ameloblastoma with large islands of odontogenic epithelial proliferation bordered by palisaded ameloblastic cells.

The postoperative course was uneventful. The patient was discharged free of pain on the second postoperative day. Control CT taken at early postoperative period confirmed the removal of the tumour (Fig. 6).

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Fig. 1 Axial CT section demonstrating a low-density mass involving the right inferior temporal region (*arrows*)

Fig. 3 Axial CT bone window setting disclosing the destruction of the lateral wall of maxillary sinus and partial involvement of the right maxillary sinus (*arrow*)

Fig. 2 Axial CT bone window setting showing the involvement of the right zygomatic process of temporal bone (*arrow*), but no extension into the pterygopalatine fossa

Discussion

Ameloblastomas are the most frequent odontogenic tumours of the mandible and maxilla and in the majority of the cases with ameloblastoma, the mandible is affected more frequently than the maxilla [8, 9]. As presented in this case report, proximity of ameloblastoma to vital structures such as the orbit, pterygomastoid fossa, and base of skull,

makes its treatment more difficult [3]. Moreover, it has been reported that the method of treatment and the type and the size of the tumour is important for the risk of recurrence [1]. In this paper, we reported a case of recurrent multicystic ameloblastoma of the mandible and infratemporal fossa extending into the maxilla.

From a surgical standpoint of view, it has been reported that the recurrence rate following a simple curettage is higher than radical resection, 75 and 20%, respectively [2, 6, 9]. In our case, the surgical resection had been difficult because of complexity of surgical anatomy of the maxilla and recurrence had been found in the surgical sites in these tumours.

Histopathologically, these tumours can be divided into at least six patterns: follicular, granular cell, basal cell, acanthomatous, desmoplastic, and plexiform types [10]. The cellular types are more aggressive than the less cellular, as presented in this case report [10]. Radiologically, solid ameloblastomas appear as multilocular radiolucent lesion without well-defined borders and expansion or resorption of the cortical bone is common [1].

Conclusion

In conclusion, ameloblastoma involving the mandible and/or infratemporal fossa extending into maxilla is a rare tumour but it should be included in the list of the osseous lesions of the maxillo-facial region. This case demonstrates the importance of follow-up because its high recurrence

Fig. 4 Axial pre- (left) and post- (center) contrast T1- and T2- (right) weighted MRIs showing a recurrent tumour mass with well-defined border at the posterior aspect of the inferior temporal region and the posterior aspect of right maxilla (arrows)

Fig. 5 Histological examination of the specimen indicated ameloblastoma with follicular pattern composed of solid islands of stellate reticulum cells surrounded by columnar and palisaded ameloblastic cells (HE $\times 100$)

Fig. 6 Control CT confirming the removal of the tumour at early postoperative period

potential. CT and/or MRI is essential to achieve the correct diagnosis in these patients.

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