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Ameloblastoma: Demographic, Clinical, and Histopathological Data, and Outcomes of Recurrence

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Abstract

Introduction: Ameloblastoma (AMB) is a benign aggressive odontogenic tumor of epithelial origin, characterized by unlimited growth capacity with a high rate of recurrence. These tumors commonly affect the mandible and require extensive surgery. The aims of this study are to characterize demographic, clinical data of AMB and to investigate criteria influencing recurrence and outcomes.

Materials and Methods: A retrospective study was conducted for patients treated for AMB during a 12-year period. Patients were included if they had a confirmed histologic diagnosis of AMB.

Results: There were 8 patients (5 females and 3 males). The mean age of the patients was 33.5 years. The horizontal branch of the mandible was the most common site. The most common clinical manifestation was swelling of affected region. Patients were treated by enucleation or extensive resection.

Conclusion: The demographic profiles of the studied tumor corroborates with data reported in the literature. This study confirmed that AMB to be more common in the mandible. Outcomes were not influenced by the predicting criteria associated with recurrence.

Keywords: Ameloblastoma; Recurrence; Predicting; Factors

Introduction

Ameloblastoma (AMB) is a rare benign odontogenic tumor of epithelial origin [1,2]. Otolaryngologists and maxillofacial surgeons are often powerless in the face of the local aggressiveness of the tumor and its high rate of recurrence [3]. To date, there are no established preventive measures for the patients who are between the ages of 30 and 60 years. The AMB etiology is unknown. Recently, some researches supported that the development of AMB should be related to dysregulation of molecular and genetic factors that promote oncogenic transformation of odontogenic epithelium to AMB. These dysregulations should be strongly associated with the expression of multiple genes associated with mitogen-activated protein kinase, sonic hedgehog, and WNT/beta-catenin signaling pathways [4]. The current treatment of AMB is based on a large surgical resection removing large healthy margin with regard to the high invasion potential of the tumor and the high risk of recurrence. In fact, the high recurrence rate of AMB is associated with molecular, and histopathological factors, the type of surgical approach, and the time of diagnosis. To date, there is a lack of published data on the demographics, clinical data, and treatment outcomes of AMB.

The aims of this study are to characterize demographic and clinical data of AMBs and to investigate outcomes of recurrence.

Materials and Methods

The otolaryngology and maxillofacial surgery databases from a single institution (CHU Saint-Pierre, Brussels) were searched for reported cases of AMB from 2001–2013. Patients were included if they had a confirmed histologic diagnosis of AMB performed by a senior pathologist according to the current WHO classification system. The following variables were retrieved: gender; age; AMB localization; clinical presentation; and recurrence characteristics.

Results

A total number of 8 patients were included (3 males, 5 females). The mean age of the patients was 33.5 years. AMB were located in the mandible (N=6), and in maxillary sinus (N=2). Among the mandibular AMBs, 3 were located at the horizontal branch of the mandible, 2 at the mandibular angle, and 1 in the intersymphyseal region. Between the two maxillary cases, one had a large bilateral extension into the maxillary sinus up to the nasal conchae and the orbital floor and the second AMB case invaded ethmoidal and frontal sinuses. In the majority of cases (N=7), the most common clinical manifestation was swelling of affected region. Multilocular radiolucency was observed in 3 cases, whereas 5 were unilocular. In 5 cases, the tumor was greater than 6cm.

In our series, segmental mandibulectomy was done in 4 cases (50%). The most common approach for reconstruction of acquired mandibular defects was the use of vascularized osseous flaps. The mean follow-up of our patients is 10 years. A conservative surgical approach was chosen in 4 cases. Nowadays, 2 recurrences were diagnosed. One patient treated by enucleation had a recurrence 28 months after the initial treatment and was successfully treated by hemi-mandibulectomy and the use of a free flap of the fibula for the reconstruction. The second patient was treated by radical surgical resection. The recurrence was treated by salvage surgery.

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The two patients are, at the end of the follow-up (respectively 8 and 10 years), free of disease. The two recurrences were not related to the anatomopathological factors. Nor the type of AMB the tumor's dimensions were associated with the recurrence rate.

Discussion

Ameloblastoma is a rare, locally aggressive odontogenic neoplasm, accounting for less than 1% of head and neck tumors [5]. The incidence of ameloblastomas varies from 0.7 to 2.41 / million / year. These tumors showed a male predilection. In our study, the male: female ratio was 1/1,33. However, others studies reported that most cases occurred in white women. Ameloblastoma is mostly diagnosed in the third and fourth decades, as in our series. The average age of the patients was 33.5 years (33.7 +/- 16.8 years reported in the literature). Moreover, different ethno geographic backgrounds influence the appearance of ameloblastomas [6]. Hendra et al found, in their meta-analysis, that the peak incidence in Africa and South America was in the third decade, while the peak incidence in Europe and North America was in the fifth and sixth decades [5].

Ameloblastoma is often asymptomatic, presenting as a slowly enlarging facial swelling (6/8 cases in our series) or an incidental finding (1/8 case in our series). Furthermore, in one case, the diagnosis was made after that the patient developed hypoesthesia of the lower dental nerve.

There are various types of this tumor and confusion still exists among the clinicians as to its correct classification according to the current 2005 WHO classification of odontogenic tumors. Multicystic ameloblastoma is the most frequent subtype while unicystic ameloblastoma can be considered a variant of the solid or multicystic subtype. Unicystic ameloblastoma is considered a less aggressive tumour with a variable recurrence rate. This subtype of tumor is more seen in young population. Furthermore, some authors advised preoperative biopsy to avoid radical surgery in these samples [7]. In our series, 5 patients presented an unicystic ameloblastomas. These patients had no recurrence.

The mandible was the chief anatomical location involved with the tumors, as described in the literature. Maxillary ameloblastoma without orbital involvement was diagnosed in one case. Maxillary ameloblastomas are rare and associated with an aggressive course because of the anatomic composition of the maxilla and adjacent structures. In case of ameloblastoma with orbital involvement, the recurrence rate is high (70%). The visual compromise, death and disease-related mortality were respectively 26%, 39% and 22% [8]. We confirmed the rarity of the disease in our 12-year period study.

In our series, at the end of the follow-up, our patients are still alive without recurrence. In our series, the surgical treatment was applied for all cases. Segmental mandibulectomy was done in four cases (50%). The most common approach for reconstruction of acquired mandibular defects was the use of vascularized osseous flaps. A conservative surgical approach was chosen in four cases. The morbidity rate was not different in both groups. In our retrospective study, two patients recurred but were salvaged by a second operation. The therapeutic decision was based on the tumor location and the extension of the associated bone defect.

But, till date, lot of controversies exist regarding the surgical treatment options [4,9]. High recurrence rates of 50-80% with conservative treatment in some subtypes warrants radical surgical resections resulting in high morbidity. However, some authors advocated a less invasive surgical approach. Recurrence rates are higher in these series, but the morbidity is less with tumoral control rates of 60% [10,11]. Others authors preferred a more aggressive surgical approach followed by a reconstruction with a vascularized osseous flap [12,13]. The main arguments to choice this treatment plan is the associated lower recurrence rates. However, actually in the literature, no guidelines to determine the treatment are well defined. Therefore, in their study [4], Troiano et al examined whether such a difference exists in the relapse rate between the conservative and radical approaches. Their results revealed a higher recurrence rate after a conservative approach compared to the surgical approach. These results were confirmed by Antonoglou et al. in their study [9].

However, the recurrence and progression of ameloblastoma are unpredictable [14].

Therefore, Yang et al. [15], in their study, developed a staging system to predict early recurrence and cancerization. Following this staging system, tumor larger than 6cm and invasion to soft tissue or adjacent anatomical structures are associated with early recurrence. In our study, after analyzing the staging system, recurrence was not guided by the same criteria. Moreover, the recurrence was not clearly associated with the type of resection. Nor the type or the extension of the tumor the tumor's dimensions were also related to recurrence. Interestingly, in their study, the surgical method did not influence the recurrence time when adjusted for confounding variables [16].

Conclusion

In our study, the demographic profiles of our patients treated for ameloblastoma were the same that the data reported in the literature. Outcomes were not influenced by the type of resection or by the type, the extension and the dimensions of the tumor.

We confirmed the potential of recurrence of these tumors, independently of predicting factors. Despite the lack of guidelines, a close follow-up for five years after initial surgery is recommended.

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