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Mini Review

Surgical Treatment of Mandibular Ameloblastoma: Radical or Conservative Surgery? A Report of 23 Cases

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Abstract:
Ameloblastoma is a benign but locally invasive odontogenic tumor with a high recurrence potential, and the choice between conservative and radical surgery remains debated. We aimed to describe the epidemiological, clinical, radiological and therapeutic features of mandibular ameloblastoma and to compare recurrence rates according to the type of surgery. We conducted a retrospective study from January 2010 to December 2020 in a department of otorhinolaryngology and head and neck surgery (Hassan II University Hospital and Omar Drissi Hospital, Fez), including 23 patients operated on for mandibular ameloblastoma, and compared two groups formed according to the treatment received (conservative or radical). Ameloblastoma accounted for 34% (23/67) of the benign odontogenic tumors and cysts of the mandible operated on. Mean age was 35 years (range 10–75), the sex ratio was 2.28, and the mean time to consultation was 25.75 months; mandibular swelling was present in all patients. Eleven patients (48%) underwent conservative surgery (enucleation with thorough curettage) and 12 (52%) radical surgery, followed by immediate reconstruction in nine of them. With a mean follow-up of 30 months, a single infratemporal recurrence was observed, in the radical group, whereas none occurred in the conservative group, and no malignant transformation was observed. In our series, a conservative approach appears justified for limited lesions, provided that follow-up is regular and prolonged, whereas radical surgery remains necessary in cases of major bone fragility or of lesions that would be difficult to control in the event of recurrence, and must be accompanied by reconstruction and dental-prosthetic rehabilitation.

Keywords: Ameloblastoma; Mandible; Conservative treatment; Radical treatment; Recurrence; Mandibular reconstruction

Introduction:

Ameloblastoma is a benign odontogenic tumor with local invasive and evolutive potential. It develops from odontogenic epithelium within a fibrous stroma, without producing calcified tissues such as enamel or dentin. It accounts for approximately 1% of tumors of the jaws and 11% of odontogenic tumors [1]. Its etiopathogenesis remains poorly understood tooth extractions, caries, trauma, inflammation and nutritional deficiencies have been suggested [2]. Clinically, it may remain asymptomatic until a facial swelling is discovered; radiologically, it appears as a radiolucent osteolytic lesion, unilocular or multilocular, with a cyst-like appearance [3]. Ameloblastoma is characterized by a high recurrence potential, which requires a precise histological diagnosis and a surgical treatment whose modalities are still debated.

Conservative treatment (enucleation, curettage) preserves bone integrity [4] but appears to carry a high recurrence rate. Radical treatment, for its part, may be aesthetically and functionally disabling and may require reconstruction, notably with a free flap [5,6]. The choice between these two approaches is therefore a fundamental question in the management of these tumors.

The aim of this work was to describe the epidemiological, clinical, radiological and therapeutic features of 23 cases of mandibular ameloblastoma managed in our department, to compare recurrence rates after conservative and radical treatment, and to compare our results with those of the literature.

Materials and methods

Study design and population

This was a retrospective descriptive study conducted over 11 years (January 2010 – December 2020) in the Department of Otorhinolaryngology and Head and Neck Surgery of the Hassan II University Hospital and the Omar Drissi Hospital in Fez. All patients, regardless of age and sex, who underwent surgery for mandibular ameloblastoma were included. Patients managed for another condition, or whose medical records were unusable, were excluded. Written informed consent was obtained from all participants prior to inclusion, and the study adhered to institutional ethical guidelines.

Data collection

For each patient, the medical record was reviewed using a standardized data collection form. We recorded age, sex and personal and family history, followed by the time to consultation, functional signs, the characteristics of the swelling and the findings of the intraoral examination. Imaging data comprised the panoramic radiograph, facial computed tomography and any preoperative biopsy. We also collected the type of anesthesia, the surgical approach, the type of surgery, the reconstruction technique and the pathology result of the surgical specimen. Finally, postoperative complications, recurrence, malignant transformation and dental rehabilitation were noted during follow-up.

Definitions and analysis

Conservative surgery was defined as enucleation of the tumor followed by thorough curettage of the residual cavity. Radical surgery was defined as bone resection through healthy tissue, at a distance from the tumor, either without interruption of mandibular continuity (non-interruptive mandibulectomy or marginal resection) or with interruption (interruptive segmental or laterotermінal mandibulectomy) [7]. Data were entered and analyzed with Microsoft Excel 2019. A descriptive analysis of the population was performed first; two groups were then formed according to the type of surgery in order to compare their recurrence rates. This comparison is purely descriptive.

Results

Baseline characteristics

The baseline demographic, clinical and radiological characteristics of the 23 patients are presented in Table 1. Among 67 benign odontogenic tumors and cysts of the mandible operated on during the study period, 23 were ameloblastomas (34%), i.e. a frequency of 2.1 cases per year. The mean age at diagnosis was 35 years (range 10–75), with a male predominance (sex ratio 2.28), and six patients (26%) reported a previous tooth extraction. Patients consulted late, after a mean of 25.75 months (range 1 month to 7 years), for a mandibular swelling that was present in all cases, hard and regular in most, and larger than 5 cm in 14 patients. Pain and dental signs were uncommon (17.4% each) and no patient reported paresthesia. The lesions were located mainly in the body (43.5%) and the ramus (26%) of the mandible. Panoramic radiography showed a unilocular image in 61% of patients and a multilocular image in 26%, and computed tomography, performed in 91% of patients, showed osteolytic, sometimes expansile lesions, with cortical breach in some cases. A preoperative biopsy was obtained in three patients because of clinical and radiological features suspicious for malignancy and concluded to ameloblastoma; in the other patients, the diagnosis was established on pathological examination of the surgical specimen.

Table 1. Baseline demographic, clinical and radiological characteristics of the 23 patients.

Characteristic	Value
Demographics	
Age at diagnosis, years, mean (range)	35 (10–75)
Most affected age group (20–29 years), n (%)	6 (26)
Male sex, n (%)	16 (69.6)
Female sex, n (%)	7 (30.4)
Sex ratio (M/F)	2.28
Medical history	
Previous tooth extraction, n (%)	6 (26)
Previous surgery for a palatal tumor, n (%)	1 (4.3)
Family history of ameloblastoma, n (%)	1 (4.3)
Clinical presentation	
Time to consultation, months, mean (range)	25.75 (1–84)
Mandibular swelling, n (%)	23 (100)
Painful swelling, n (%)	4 (17.4)
Dental signs (pain, mobility or bleeding), n (%)	4 (17.4)
Tumor size, cm, range	1–14

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Characteristic	Value
Tumor size greater than 5 cm, n (%)	14 (60.9)
Tumor size smaller than 5 cm, n (%)	8 (34.8)
Tumor size not specified, n (%)	1 (4.3)
Cutaneous inflammatory signs, n (%)	3 (13.0)
Mucosal inflammation with ulceration, n (%)	3 (13.0)
Ectatic mucosa, n (%)	1 (4.3)
Purulent mucosal fistula, n (%)	1 (4.3)
Paresthesia, n (%)	0 (0)
Location of the lesion	
Body of the mandible, n (%)	10 (43.5)
Ramus, n (%)	6 (26)
Hemimandible, n (%)	2 (8.7)
Angle, n (%)	2 (8.7)
Symphysis, n (%)	2 (8.7)
Ramus and body, n (%)	1 (4.3)
Imaging and biopsy	
Panoramic radiograph performed, n (%)	20 (87)
Unilocular image, n (%)	14 (61)
Multilocular image, n (%)	6 (26)
Facial computed tomography performed, n (%)	21 (91)
Preoperative biopsy performed, n (%)	3 (13.0)

Treatment

Surgery was performed under general anesthesia in 22 patients and under local anesthesia in one. The approach was intraoral (lower vestibular in 69.5% of patients, lower intrasulcular in 8.7%) or external cutaneous of the Sebileau type in four patients (17.3%). Eleven patients (48%) underwent conservative surgery and 12 (52%) radical surgery (Table 2). Immediate reconstruction was performed in nine of the 12 patients of the radical group, i.e. all those who had a segmental mandibulectomy: iliac crest bone graft fixed with a plate in six, and plate alone, used as a space maintainer, in three patients who refused a free flap for a defect longer than 10 cm. Pathological examination of all specimens concluded to ameloblastoma, without specification of the histological subtype.

Table 2. Type of surgery performed (n = 23).

Type of surgery	No. of cases	%
Conservative surgery: enucleation with thorough curettage	11	48
Radical surgery: interruptive segmental mandibulectomy	9	39
Radical surgery: interruptive lateroterminal mandibulectomy	1	4.3
Radical surgery: non-interruptive mandibulectomy (marginal resection)	2	8.7
All radical surgery	**12**	**52**

Postoperative course and follow-up

The immediate postoperative course was uneventful in all patients. Three patients developed surgical site infection with suture dehiscence (two infections and one submental collection). One patient developed, on day 10, an infection with exposure of the iliac graft and of the plate, requiring removal of the graft, lavage and watertight closure of the oral mucosa under antibiotic therapy; secondary reconstruction is planned. One patient developed paresis of the marginal mandibular branch of the facial nerve, which resolved completely within one month under vitamin therapy and corticosteroids. None of the patients whose tumor involved the tooth-bearing portion received dental rehabilitation, and all complained of masticatory discomfort.

Mean follow-up was 30 months (range 6 months to 5 years); two patients were lost to follow-up after the pathology result. A single recurrence (4.3% of the series) was observed: an infratemporal lesion occurring three years after a segmental mandibulectomy that had left the condyle in place without repair. It was treated by lateroterminal mandibulectomy, whose sequela was facial asymmetry with lateral deviation and temporomandibular joint dysfunction. This recurrence therefore concerned the radical group (1/12), none having been observed in the conservative group (0/11). No malignant transformation or metastasis was detected.

Discussion

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This retrospective series of 23 mandibular ameloblastomas operated on over 11 years shows a predominantly conservative first-line management (48%), with no recurrence in the conservative group after a mean follow-up of 30 months, and a single recurrence in the radical group.

Epidemiological and clinical features

With an estimated annual incidence of 0.5 new cases per million inhabitants, ameloblastoma is a rare tumor, more frequent in Africa, China and India than in Western countries [8]. Our frequency of 2.1 cases per year is comparable to those reported in Tunisia (2 cases per year) [3] and in a Moroccan series (1.28 cases per year) [9]. The mean age of 35 years is consistent with the literature, which places the peak incidence between 30 and 60 years, with a mean age close to 36 years [8,10] (Table 3). A slight male predominance is reported in Africa, North America and Asia, and a non-significant female predominance in South America and Europe [11]. The marked male predominance in our series (sex ratio 2.28) contrasts with the sex ratio close to 1 of most comparable series, which may partly reflect the limited size of our sample.

Table 3. Comparison of our series with the literature.

Series	Country	n	Mean age (years)	Sex ratio M/F	Conservative surgery (%)
Kamal et al. [9] (2008)	Morocco (Rabat)	32	33	1	59.3
Bel Hadj Hassine et al. [3] (2017)	Tunisia	22	36	1.43	32
Ba et al. [12] (2016)	Mali	51	31.1	0.94	56.9
Laborde et al. [13] (2017)	France	27	46.3	1.45	61.2
Ruslin et al. [14] (2018)	Indonesia	56	39.7	0.6	62.5
Haq et al. [4] (2016)	United Kingdom	31	–	–	87
Our series	**Morocco (Fez)**	**23**	**35**	**2.28**	**48**

Diagnosis was late in our series (mean time to consultation 25.75 months), as in a previous series from our institution (25 months) [15] and in a Tunisian series (35 months) [3]. This delay is explained by the paucity of clinical signs and the slow growth of the tumor, but also, in developing countries, by difficulties of access to care in a modest socioeconomic context; in developed countries, ameloblastoma is more often discovered during a routine dental check-up [16]. The large lesions (greater than 5 cm in 14 of our patients) are in line with observations from African series [12]. Mandibular swelling, constant in our series, is the cardinal symptom; pain is rare (17.4%), and the absence of paresthesia contrasts with the inferior alveolar nerve hypoesthesia observed in a third of the patients of the Tunisian series [3].

Imaging and diagnosis

The radiological features of ameloblastoma are characteristic but not pathognomonic [8]. Panoramic radiography, the first-line examination, showed a unilocular image in 61% of cases and a multilocular one in 26%. Computed tomography, performed in 91% of our patients, defines the volume, local extension, cortical breach and vascular and nerve relationships; it is more informative than plain radiography for morphological assessment and treatment planning [17]. A definite diagnosis rests on pathological examination of the surgical specimen: in our series, a preoperative biopsy was performed in only three patients. In the absence of a preoperative histological classification, the therapeutic approach is guided by the radiological appearance [13].

Conservative or radical surgery?

Enucleation alone is associated with a recurrence rate of 60 to 90%, reduced to about 55% when combined with curettage [8,17]. This approach is considered feasible only for unicystic ameloblastomas with intraluminal growth [8]. Some authors supplement enucleation with cryotherapy of the bone walls [18] or application of Carnoy's solution [19] in order to destroy residual epithelium. In our series, a conservative approach was used in 48% of patients, a proportion slightly lower than in recent series (56.9 to 62.5%, Table 3) but much higher than in a previous series from our institution (10%) [15], reflecting an evolution of our practice toward more conservative surgery. Ruhin-Poncet et al. consider enucleation to be the treatment of choice (85% of their patients), owing to the small size of the lesions related to early consultation [20].

The therapeutic choice cannot be standardized and must rely on clinical, radiological and histological arguments and, above all, on tumor extension [9]. In our series, the indication for radical treatment depended neither on the histological type nor on radiological characteristics, but essentially on the fragility of the inferior border and of the posterior border of the ramus, which carried a significant risk of intraoperative fracture; our approach is therefore conservative in the first instance. The drawbacks of radical surgery are the resulting facial asymmetry and functional impairment, and the need for reconstruction.

Recurrence and follow-up

In the literature, the recurrence rate is markedly higher after conservative than after radical treatment (Table 4) [9,13,21]. In our series, no recurrence was observed in the conservative group (0/11) and only one in the radical group (1/12): an infratemporal lesion appearing three years after a segmental mandibulectomy that had left the condyle in place without repair, which could correspond to the progression of a residual tumor rather than a true recurrence.

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Table 4. Recurrence rate according to the type of surgery: comparison with the literature.

Series	n	Recurrence after conservative surgery	Recurrence after radical surgery
Nakamura et al. [21] (2002)	78	33.3%	7.1%
Kamal et al. [9] (2008)	32	9.4%	0%
Laborde et al. [13] (2017)	27	37%	3.7%
Our series	**23**	**0% (0/11)**	**8.3% (1/12)***

** Calculated on the 12 patients of the radical group; relative to the 23 patients of the whole series, the recurrence rate is 4.3%.*

This favorable result in the conservative group must nevertheless be interpreted with caution: the sample is small (11 patients); mean follow-up is only 30 months whereas more than 50% of recurrences occur within five years and up to 10 to 15 years after surgery, which justifies follow-up of at least 10 years [22]; finally, the two groups are not comparable a priori, since the least extensive lesions were preferentially treated conservatively. Moreover, recurrence is not synonymous with surgical failure or poor prognosis as long as the new lesion remains similar to or smaller than the initial one [7].

The risk of recurrence also depends on the histological type: after conservative treatment, solid or multicystic forms recur more often (40%) than the desmoplastic form (15%) [23], and the Ki67 proliferation index and stromal CD10 expression have been proposed as markers of local aggressiveness and recurrence [24]. As the histological subtype was not specified in our pathology reports, these factors could not be analyzed. No malignant transformation or metastasis was observed; metastases of ameloblastoma are exceptional and occur most often in the lung (80% of cases) [25].

Reconstruction and rehabilitation

Interruptive mandibular defects after excision of a benign tumor are the best indication for immediate one-stage reconstruction, whose aim is to restore the morphology of the lower third of the face and to allow dental rehabilitation as well as mastication, speech and swallowing [26]. All our patients who underwent segmental resection were reconstructed during the same operation: six with a non-vascularized iliac graft fixed with a plate, and three with a plate alone after refusing a free flap. Plates alone expose to a high risk of fracture, exposure, screw extrusion and infection, and only the addition of a bone graft allows a durable functional reconstruction [27]; indeed, we experienced exposure of the iliac graft and of the plate on day 10. Vascularized free flaps (fibula, iliac crest) are widely described for these reconstructions [6,28]; none was performed in our series. Finally, the absence of dental rehabilitation in our patients, all of whom had masticatory discomfort, underlines the importance of close collaboration between the maxillofacial surgeon and the dentist.

Limitations

The limitations of this work are its retrospective and single-department design, the small sample size, the limited mean follow-up (30 months, two patients lost to follow-up), the lack of specification of the histological subtype, the absence of comparative statistical analysis, and the fact that treatment allocation depended on lesion extension and bone fragility, which exposes the study to selection bias.

Conclusion

Mandibular ameloblastoma is a locally invasive benign tumor with slow evolution and a long-silent clinical picture, dominated at an advanced stage by swelling. Radiological assessment guides the diagnosis, which is confirmed by pathological examination of the surgical specimen. Given its benign nature and slow growth, conservative treatment may be considered, taking quality of life into account: it appears justified for limited lesions, provided that follow-up is regular and prolonged. Radical surgery remains unavoidable in cases of fragility of the inferior border or of the posterior border of the ramus with a risk of intraoperative fracture, or when the radio-clinical characteristics of the lesion would not allow it to be controlled in case of recurrence. It must be accompanied by reconstruction and appropriate dental-prosthetic rehabilitation, through close collaboration between the maxillofacial surgeon and the dentist.

Conflicts of interest: The authors declare that they have no conflicts of interest relevant to the content of this article.

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