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Original Article

Maxillary ameloblastoma: results of the treatment in 11 patients

Rasmané Béogo[1,*], Tarcissus Konsem[2], Mathieu Millogo[2], Hèra Michel Kohoun[1], Toua Antoine Coulibaly[1], Ibraïma Traoré[1]

[1] Department of Stomatology and Maxillofacial Surgery, CHU Souro Sanou, Burkina Faso
[2] Department of Stomatology and Maxillofacial Surgery, CHU Yalgado Ouédraogo, Ouagadougou, Burkina Faso

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Abstract – Introduction: Surgery of maxilla ameloblastoma is mutilating and the tumor propensity for recurrence high. **Patients and Methods:** The oral and visual functions, facial morphology and tumor recurrence in 11 patients after maxillary ameloblastoma surgery are retrospectively reviewed. **Results:** Facial morphology was satisfactory in 7 patients who had all tumor removal by a type 1 or 2A maxillectomy and surgical wound closure. Out of these patients, 4 who had dental rehabilitation by conventional prosthesis presented satisfactory mastication. Four patients subjected to the tumor removal by a type 3 maxillectomy had all facial asymmetry. Out of these, 1 patient who did not have the orbital floor defect repair presented diplopia and enophtalmos, and 2 patients subjected to the palate defect repair by a prosthetic obturator or oral mucosa had elocution impairment. The tumor recurrence occurred in 2 patients after tumor enucleation and in 1 patient after radical surgery out of 8 patients who had a postoperative follow-up. **Discussion:** In ameloblastoma surgery, achieving both the tumor recurrence prevention and a satisfactory facial reconstruction is challenging. **Conclusion:** Avoiding the tumor recurrence should be the major goal when patient post-surgical follow-up cannot be guaranteed.

Introduction

Ameloblastoma is the most frequent benign but locally aggressive odontogenic tumor of epithelial origin, representing about 14% of the jaw tumors and cysts [1–3]. In Africa, it afflicts predominantly patients between the third and the fourth decades of life [4,5]. Histologically, the tumor is classified in a diversity of subtypes of which solid/multicystic/conventional and unicystic ameloblastomas are the most frequent [6,7]. Surgical resection with wide margins of healthy tissue is the mainstay of the current treatment with proven efficacy [8]. Such an approach may result in functional and esthetical impairments as usually mutilating whereas the midface reconstruction is challenging. Additionally, postoperative recurrences of ameloblastoma are common with some of the recurrent tumors lethal [9]. Likely due to the rarity of maxilla ameloblastoma whose highest reported frequency is 25.7% of all ameloblastomas [10], papers dealing with the treatment of this tumor are scarce in the literature. This article aimed to report the oral functions, visual function, facial morphology and tumor recurrence in 11 patients after maxillary ameloblastoma surgery.

Patients and methods

The medical records of 11 patients who underwent surgical treatment for maxilla histologically confirmed ameloblastoma from 1994 to 2012 in two referral hospitals in Burkina Faso (CHU Souro Sanou and CHU Yalgado Ouedraogo) are retrospectively reviewed. On clinical and radiological grounds, the tumor excision was performed under general anesthesia *via* buccogingival sulcus incision with or without paralateronasal cutaneous incision, according to the lesion extent. Bone reconstruction consisted in immediate non-vascularized bone grafting. The graft was harvested from the iliac crest or the calvaria and fixed with screws and plates. In teeth loss after partial maxillectomy, patients were proposed a conventional dental prosthesis. Four patients had this rehabilitation. Histological diagnosis of ameloblastoma was established post operatively, on the surgical specimen, in accordance with the practice in management of clinically benign tumors or cystic lesions in Burkina Faso. Patients' postsurgical follow-up was done as long as possible and consisted in clinical examination

* Correspondence: rbeogo@yahoo.fr

and computed tomography scan. It was possible in 8 patients, for a period ranging from 2 to 12 years.

The data collected included the tumor localization and extent, the surgical approach (radical resection *versus* conservative surgery), the surgical defect extent, the post-operative oral functions, visual function and facial morphology, the tumor eventual recurrence and the time of its onset. Anterior maxilla referred to localization between the two maxillary canines. Posterior maxilla referred to localization behind the canine. Radical resection consisted in the tumor removal with healthy margins confirmed at the histological examination. Conservative surgery referred to the tumor enucleation with or without the margins curettage.

The surgical defect extent was graded according to the Cordeiro and Santamaria maxillary defects classification system [11]: type 1 maxillectomy defect was defined as that consecutive to loss of one or two maxilla walls, excluding the palate; type 2A maxillectomy defect was that which resulted from resection of the lower five walls and less than 50% of the palate with preservation of the orbital floor; type 2B maxillectomy defect referred to that resulting from loss of the lower five walls and more than 50% of the palate with preservation of the orbital floor; type 3 maxillectomy defect referred to that which resulted from loss of all the six maxillary walls; type 4 maxillectomy defect was defined as that consecutive to resection of the orbital contents and the maxilla walls, excluding the palate.

Oral functions referred to mastication after dental rehabilitation and elocution. They were broadly scored as satisfactory (ability of mastication, normal elocution) or unsatisfactory (difficulty of mastication, disturbance of elocution). Visual function was evaluated by presence of diplopia after type 3 maxillectomy. Facial morphology was evaluated by presence of enopthtalmos after type 3 maxillectomy and facial asymmetry irrespective of the maxillectomy type.

Results

Facial morphology was satisfactory in 7 patients who had all the tumor removal by a type 1 or 2A maxillectomy and surgical wound closure (Tab. 1). Out of these patients, 4 who had dental rehabilitation by a conventional prosthesis presented satisfactory mastication. Four patients subjected to the tumor removal by a type 3 maxillectomy had all facial asymmetry (Fig. 1). Out of these, 1 patient who did not have the orbital floor defect repair presented diplopia and enophtalmos, 2 patients subjected to palate defect repair by a prosthetic obturator or oral mucosa had elocution impairment. The tumor recurrence was noted in 2 patients after tumor enucleation and in 1 patient after radical surgery out of 8 patients who had a postoperative follow-up.

Discussion

As in the treatment of any benign tumor, one of the goals of maxillary ameloblastoma surgery is the cure by performing the tumor complete removal. To achieve this, authors mostly recommend the tumor excision 1–2 cm beyond its radiological limits as presence of the tumor cells is reported 0.8–12 mm beyond these limits [12,13]. Such an approach implies sacrifice of a variable extent of osseous tissue involving the alveolar ridge, the hard palate, the maxillary sinus walls and even sometimes the orbital floor [10,12,14] but sparing usually the tumor overlying skin and mucosa. In type 1 and even type 2A maxillectomy defects, closure of the surgical wound using the tumor overlying soft tissue may enable to achieve satisfactory oral functions and facial morphology. Type 3 maxillectomy defects commend tissue transfer or prosthesis for imperative reconstruction of the orbital floor, the hard palate, the maxillary arch and the cheek as key functional or morphological structures of the midface. The best results in face substance loss repair are achieved currently thanks to vascularized composite bone-containing free flaps of which fibula, scapula, iliac crest, and radial forearm flaps [15,16]. Alternatives of these means whose technology is hardly available in underserved setting are non-vascularized bone grafts and prostheses. Despite the myriad of maxillary reconstruction options, results of extensive maxillectomy repair may be fairly good or even vexing. This is particularly true where non-vascularized bone grafting and prostheses use are the only available options of facial reconstruction. Propensity of ameloblastoma to recur even after a proper surgery is another frustration in this neoplasm treatment. Rates of recurrence higher after conservative surgery and reaching up to 90–100% support the limited role of this approach in the current management of ameloblastoma [9,17,18]. Carlson *et al*. declare not rational to treat conservatively a so aggressive lesion with intension to cure [8]. For these authors, the so-called tumor recurrence after such approach is in reality a progression of the lesion [8]. Some authors however advocate conservative surgery in pediatric patients arguing for the risk of facial growth compromise in radical surgery [19–21]. Patient's wish is another indication of ameloblastoma conservative surgery. In this way, Sachs *et al*. report that some patients would consent for conservative and iterative surgery rather than a radical mutilating excision and defect repair [22]. In some patients of this study, decision of ameloblastoma enucleation comes from misdiagnosis given lack of histological examination of the neoplasm prior to its removal. That treatment should be followed by a complementary excision of the tumor operative site margins as Chapelle *et al*. recommend after enucleation of unicystic ameloblastoma with mural invasion [17]. Choice may be challenging between radical surgery resulting potentially in alteration of quality of life and conservative surgical approach with unavoidable recurrence of ameloblastoma. In this dilemma, our opinion is that in a setting of poorly compliance of patients for post-operative follow-up such as Africa, recurrence should be regarded as the major consideration. With respect to that consideration, Effiom *et al*. recommend aggressive radical surgery whenever possible [7]. Whatever the surgical modality, ameloblastoma has a potentiality for recurrence commending a strict post-surgical follow up of

Table 1. Tumor extent, surgery modalities, results and patient outcomes.

Patient	Tumor localization and extent	Surgery approach	Defect extent	Defect repair method	Defect repair result	Outcome
1	Ant and Post Max	Radical surgery	Type 2 Maxillect	Wound closure	Satisfatory morphology Satisfatory élocution and oral continence	Lost of follow-up less than 1 year
2	Ant Max	Enucleation	Type 1 Maxillect	Wound closure and dental prosthesis	Satisfatory morphology Satisfatory masticatin and elocution	Lost of follow-up less than 1 year
3	PS: Post Max RS: Post Max	PS: Enucleation RS: Radical surgery	Type 1 Maxillect Type 2 Maxillect	PS: Wound closure RS: Wound closure and dental prosthesis	PS: Satisfatory morphology RS: Satisfatory morphology Satisfatory mastication and elocution	PS: Recurrence 1 year after RS: Free of récurrence at 12 years
4	Post Max	Radical surgery	Type 1 Maxillect	Wound closure	Satisfatory morphology Satisfatory élocution and oral continence	Lost of follow-up less than 1 year
5	PS: Ant and Post Max RS: Post Max and temporal fossa	PS: Radical surgery RS: Radical surgery	Type 2 Maxillect Type 3 Maxillect	PS: Wound closure RS: Orbital floor bone grafting and prosthetic obturator	PS: Satisfatory morphology RS: Facial asymetry Elocution impairment Satisfatory visual function	PS: Recurrence 10 years after RS: Free of récurrence at 3 years
6	PS: Ant Max RS: Ant Max	PS: Enucleation RS: Radical surgery	Type 1 Maxillect Type 2 Maxillect	PS: Wound closure RS: Wound closure and dental prosthesis	PS: Satisfatory morphology RS: Satisfatory morphology Satisfatory mastication and elocution	PS: Recurrence 10 years after RS: Free of recurrence at 4 years
7	Ant Max, Post Max, temporal fossa and orbit	Radical surgery	Type 3 Maxillect	Orbital floor bone grafting and prosthetic obturator	Facial asymetry Satisfatory élocution Satisfatory visual function	Free of recurrence at 5 years

Table 1. (continued).

Patient	Tumor localization and extent	Surgery approach	Defect extent	Defect repair method	Defect repair result	Outcome
8	Post Max	Enucleation	Type 2 Maxillect	Wound closure and dental prosthesis	Satisfatory morphology Satisfatory mastication and élocution	Free of recurrence at 2 years
9	Ant Max, Post Max, and orbit	Radical surgery	Type 3 Maxillect	Orbital floor bone grafting and palatal mucosa closure	Facial asymetry Satisfatory élocution and visual function	
10	Ant and Post Max	Radical surgery	Type 3 Maxillect	Wound closure	Facial asymetry Elocution impairment Diplopia	Free of recurrence at 3 years
11	Ant and Post Max	Radical surgery	Type 2 Maxillect	Wound closure	Satisfatory morphology Satisfatory elocution	Free of recurrence at 3 years

PS: Primary surgery, RS: Recurrence surgery, Ant: Anterior, Post: Posterior, Max: Maxilla, Maxillect: Maxillectomy

Fig. 1. Facial asymmetry in a patient with orbital floor bone grafting and prosthetic obturator after a type 3 maxillectomy.

the ameloblastoma patient. As a recurrent tumor course may be for a long time clinically asymptomatic, there is an evidence that modern imaging including computed tomography and magnetic resonance plays currently a key-role in earlier diagnostic of the recurrences. Feinberg and Steinberg recommend tomography 2–3 weeks, 6 months then every year, postoperatively [14] while Chapelle *et al*. recommend it every year during the 5 first years postoperatively then every two-year during 25 years [17] supporting lack of standardization in the timing of the follow-ups.

Conclusion

In setting of patient's poor post-surgical follow-up, wide excision with negative margins combined to a suited facial reconstruction should be the treatment of maxillary ameloblastoma whenever possible.

Conflicts of interest: The authors declare that they have no conflicts of interest in relation to this article.

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