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Recurrence of Ameloblastoma after Mandibular Resection and Fibula Free Flap Reconstruction: Case Discussion

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Abstract **Case Report**

Ameloblastomas are odontogenic tumors that develop from epithelial remnants. They are slow-growing, locally aggressive and have a high recurrence rate, 10 to 20 or even 30 years after initial resection. Recurrence of ameloblastoma may be due to inadequate treatment or to the aggressive nature of the tumor cells, which readily destroy surrounding tissue. We report the case of a 53-year-old patient who had undergone repeated curettage of a mandibular cyst in the setting of ameloblastoma. In 2012, a segmental mandibulectomy with placement of a screw-plate space maintainer had been performed on an outpatient basis. Three years later, he was referred to our department for debricolage of the mandibular plate. He underwent removal of the osteosynthesis material and reconstruction with a fibular free flap. 7 years later, the patient presented with a recurrence of the ameloblastoma, which depended on the remaining parasymphyseal bone and the fibular free flap. The patient underwent a segmental mandibular osteotomy, removing the free fibular graft and the right condyle. The bone deficit was filled by a stent made of surgical cement, mounted on a mini reconstruction plate. Ameloblastoma is a benign tumor. Segmental resection with 2cm margins remains the standard treatment for the multilocular and follicular types. Recurrence on a free fibula flap is uncertain and rare.

Keywords: Ameloblastoma; recurrence; free fibula flap.

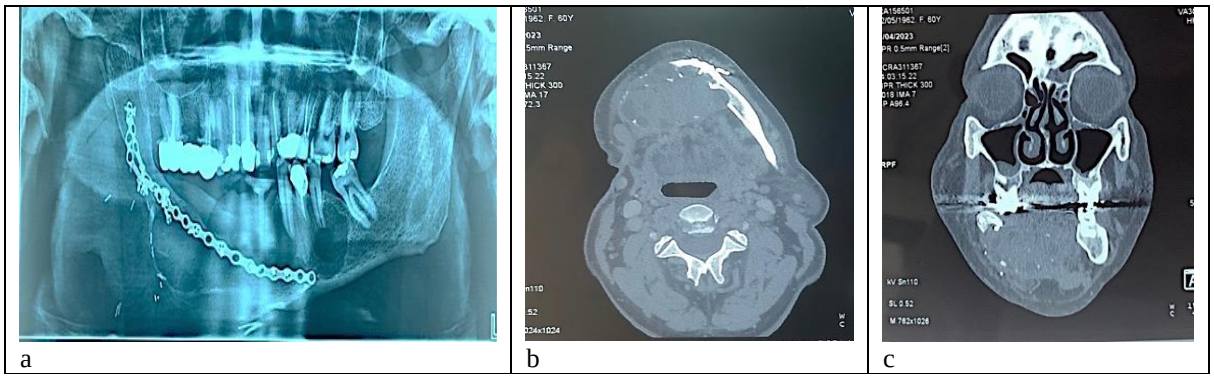
INTRODUCTION

Ameloblastomas are odontogenic tumors that develop from epithelial remnants. They are slow-moving, locally aggressive and have a high recurrence rate, 10 to 20 years or even 30 years after the initial resection [1]. Ameloblastoma represents 1% of all oral tumours, preferably located at the mandible in 80% [2]. Although ameloblastoma is a benign tumor, it can cause functional and morphological damage such as impaired masticatory function, facial and mandible deformation. The recurrence of ameloblastoma may be due to inadequate treatment or aggressive tumour cells that easily destroy surrounding tissues. Local carcinological excision, at healthy margins, is necessary to ensure a curative effect in the case of multicystic and follicular type ameloblastoma [3]. The bone deficit should be reconstructed by a bone graft or vascularized free bone transfer. Although these bone inputs do not contain odontogenic tissue, some cases of recurrence in the bone graft have been reported in the literature, especially in the fibular bone [4]. We discuss a case of massive recurrence of ameloblastoma on fibula bone transfer, 7 years after segmental resection and mandibular reconstruction.

CLINICAL CASE REPORT

Patient aged 53 who had a curettage of a mandibular cyst between 1999-2011, with 8 relapses in an ameloblastoma. In 2012 a segmental mandibulectomy with the placement of a space maintainer by a screwed plate had been performed externally. Three years later, he is taken into our department for mandibular plaque debriefing. He benefited from an ablation of the osteosynthesis material, reconstruction by a free flap of fibula. The follow-up to the operation was simple, with regular clinical and radiological checks every 6 months. The patient was lost to view during the covid-19 pandemic. In 2023, it presents with a low jugular swelling, opposite the area of the fibular graft and the right mandibular angle. The endooral examination finds a mass between tooth 32 and the right ramus, hard and painless fixed relative to the bone and mucous plane, and filling the lower right vestibule and the homolateral buccal floor. The examination finds free cervical ganglion areas. A radiographic examination of an

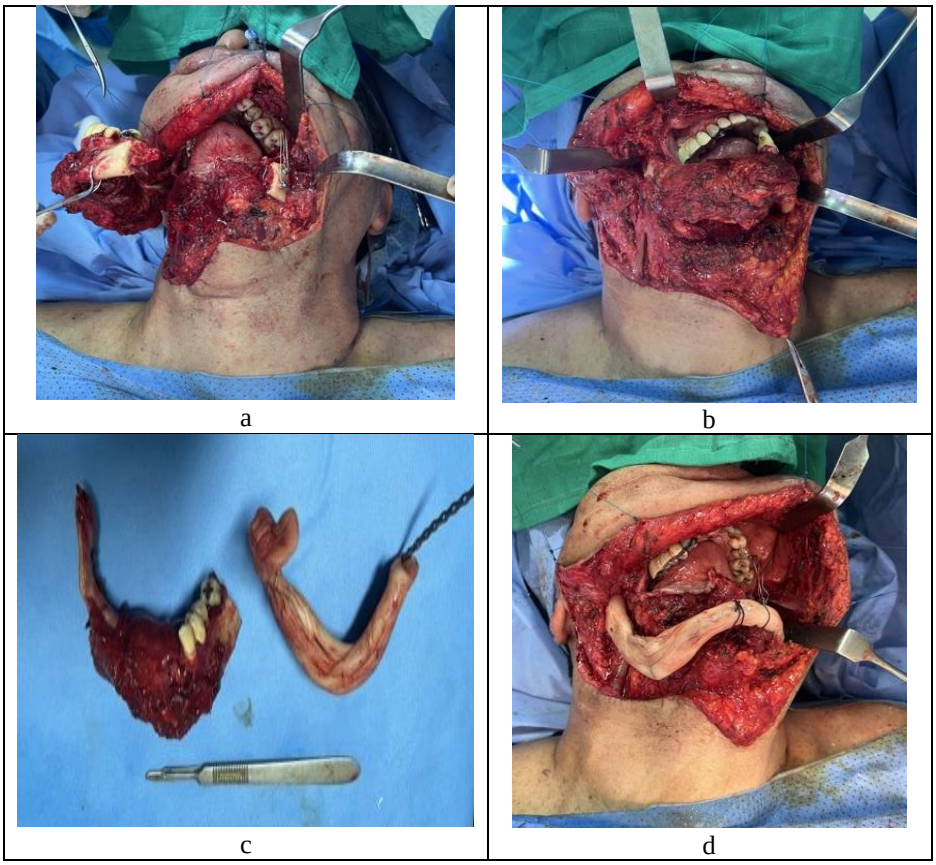
orthopanthogram (Fig 1a) shows a multilocular radioclear image from 37 to right ramus, blowing the external cortical of the fibular graft and rhizalysis of teeth 34 and 35. The computed tomography obvi-sit ed a mandibular mass measuring 60 x 40 mm in the axial plane, (Fig 1b) blowing the bone corticals, the soft gingival parts were repressed and not infiltrated, signaling a recurrence of ameloblastoma on the left parasymphyseal bone and the free fibular flap.



a b c

Figure 1: (a) multilocular radioclear image from 37 to right ramus blowing the external cortical of the fibular graft and rhizalysis of 34;35. axial cut (b); coronal cut (c): mandibular mass measuring 60 x 40 mm in the axial plane blowing the bony corticals, the soft gingival parts are repressed and not infiltrated, and partial filling of the medial and paramedical buccal floor (recurrence of symphyseal ameloblastoma and bilateral para-symphyseal has right predominance)

The patient was treated with a segmental mandibular osteotomy involving the 37 molar, the free graft of the fibula and the right condyle (Fig 2a and 2b). The bone deficit was filled by an endoprosthesis made of surgical cement, mounted on a mini reconstruction plate profile 1 mm, All fixed on the left rising branch by 5 screws 9 mm, and the anterior heads of the digastric and hyo-glosses have been fixed on the prosthesis (Fig 2d). The anatomopathological study of the excision piece shows a recurrence of a follicular type ameloblastoma.



a b c d

e

Figure 2: (a) and (b) segmental mandibular osteotomy involving molar 37, free graft of the fibula as well as the right condyle (c) endoprosthesis made by surgical cement, mounted on a mini reconstruction plate profile 1 mm (d) endoprosthesis fixed on the rising branch by screws of 9 mm and with the anterior heads of the digastric muscles and hyo-glosses (e) panoramic radio control

DISCUSSION

Ameloblastoma is a benign odontogenic tumor, defined by the World Health Organization (WHO) as a tumor formed by an odontogenic epithelium with a mature fibrous stroma without odontogenic ectomesenchyma. Although benign and slow-moving, ameloblastoma is characterized by local aggressiveness and a high recurrence rate, even with complete excision with healthy resection margins [5, 6], more than 50% of patients relapse in the first 5 years post-operative [7]. Moreover, recurrence on free bone flap is rare. In case of large, multilocular or follicular tumor invading the underlying soft parts, it is recommended to make a wide segmental resection with bone margins of 2 cm, Curettage of the proximal and distal bone marrow space and excision of underlying soft tissue.

A recent literature review was conducted in 2022 by H. Zhang *et al*., and updated in 2023 regarding patients with relapsed ameloblastoma on bone reconstruction (Table 1) [8-11, 4]. She reported 3 cases of relapses on fibula bone flap for an examination that had included 23 patients with a sex-ratio male/female of 1/1, for a recurrence delay of 2 to 36 years (with an average of 14.7 years). 13 (56.5%) cases of relapses on iliac bone graft with an average delay of 14.3 years; 5 (21.7%) cases on costal bone graft with an average delay of 22.6 years; 3 (13%) cases on free bone transfer of fibula with an average delay of 10.6 years and 2 (8.6%) For other bone transplants. It should be noted that there are currently only 4 cases of relapse of ameloblastoma on free limb of the fibula published in the literature, including our case and the time limits for relapse of the work of Basat [12] and Sharma [13] which are respectively 7 and 8 years support our result which is also 8 years.

The recurrence of ameloblastoma can be explained by three hypotheses: the recurrence may come from the bone banks, the underlying soft tissues, or by contamination of the surgical field with tumor cells [14].

Some authors report the existence of a mechanism that could promote tumor recurrence during bone repair. Cytokines that contribute to tumor neovascularization may also contribute to tumor invasion and metastasis. The bone graft contains viable bone cells, and has osteogenesis capabilities, and its capillaries can also anastomose directly from the graft area to establish blood circulation. Proliferating capillaries may develop in the graft and complete the vascularization process [15].

This may explain the fact that the average recurrence time in case of a free fibular flap is shorter compared to the iliac graft and the costal graft, because the vascularization of the fibula is more abundant leading to a rapid supply of nutrients for the tumor cells.

Table 1: Reported cases of relapsing ameloblastoma in bone grafts

Auteurs	Sexe	Type de greffe	Délais moyen de récidive (années)
Graffe [10]	F	Os iliaque	13
Carvalho [11]	F	Os iliaque	20
Dolan [16]	M	Os costal	13
Marinelli [17]	M	Os iliaque	12
Stea [8]	F	Os iliaque	7
Zachariades [18]	M	Costal borne	36
	M	homogeneous bone	2
	M	Pelvic bone	4
	F	autogenic borne	7
Vasan [19]	F	Pelvic bone	28
Bianchi [20]	F	Pelvic bone	27
Martins [21]	F	Pelvic bone	16

Su [22]	F	Pelvic bone	16
	M	Pelvic bone	3
	M	Pelvic bone	4
Choi [23]	M	Pelvic bone	20
Essaadi [24]	M	Costal bone	33
Jian [25]	M	Pelvic bone	16
	F	Costal bone	14
Basat [26]	F	Febular bone	7
Sharma [27]	F	Febular bone	8
Hamdy [28]	F	Costal bone	17
Zhang [4]	M	Febular bone	17
Notre cas	M	Febular bone	8

According to published studies, there are predictors of tumor recurrence in ameloblastoma: mandibular localization, multicystic histological type, and follicular [27]. All these predictive elements were present in our patient. In addition to these factors predictive of relapses, a recent study showed that there was a correlation between the expression of the cell proliferation index (Ki67) and CD10 by stromal cells at immunoassayhistochemical, and the aggressive local and recurrent profile of ameloblastoma. So, the expression of these markers are a factor in poor prognosis of ameloblastoma. Unfortunately our patient did not benefit from an immunohistochemical examination.

CONCLUSION

Ameloblastoma is a benign odontogenic tumor that remains aggressive locally, with a relatively high recurrence rate. Segmental resection with 2cm margins remains the reference treatment for multilocular and follicular type. Recurrence on free flap of fibula is uncertain and rare, long-term follow-up remains necessary.

Conflict of Interest: Authors do not report any conflict of interest

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