East African Scholars Journal of Medicine and Surgery
Abbreviated Key Title: EAS J Med Surg
ISSN: 2663-1857 (Print) & ISSN: 2663-7332 (Online)
Published By East African Scholars Publisher, Kenya

Volume-2 | Issue-1 | Jan-2020 | DOI: 10.36349/EASJMS.2020.v02i01.003

OPEN ACCESS

Review Report

Ameloblastic Carcinoma: A Rare Pathology But Not Exceptional

Charles Bengondo Messanga[1], Ernest Kenna[1], David Ntep Ntep Bienvenue*[1], Joseph Nkodo Mendimi[2], Stive Fokam Tamoh[1], William Bakari Ndjidda[1]

[1]Department of Odontostomatology and Maxillofacial Surgery, Yaounde University, Teaching and Hospital Center, Cameroon
[2]Yaounde Jamot Hospital, Cameroon

Article History
Received: 04.12.2019
Accepted: 11.12.2019
Published:

Journal homepage:
http://www.easpublisher.com/easms/

Quick Response Code

Abstract: Ameloblastic carcinoma is a tumor which combines histological characteristics of ameloblastoma and the cytological features of malignity. According to the evolution, it can be primary or secondary. The treatment is not yet well defined. Surgery which remains the main treatment modality should be more aggressive than in a usual ameloblastoma. This article reports a case of a secondary ameloblastic carcinoma and its treatment in our context. The patient was a woman, age 45 years, treated four times before by conservative surgery method on the mandible for ameloblastoma, each followed by recurrences. A panoramic radiography exam showed a destruction of the corpus of the mandible. From a biopsy, the histological analysis concluded to an ameloblastic carcinoma. The treatment was a radical surgery resection of the whole corpus of the mandible far from the ends of lesion. This corpus was immediately rebuild by two metallic plaques disposed one over the other, fixed on the two ramus. Post operative period was simple and five years after, there is no recurrence.

Keywords: ameloblastic carcinoma, radical surgery, reconstruction.

INTRODUCTION

Ameloblastic carcinoma is a relatively rare odontogenic malignancy, constituting approximately 1.6 to 2.2% of odontogenic tumors (Rizzitelli, A. *et al*., 2014). It differs from "malignant" ameloblastoma, which presents with metastases but remains benign on histopathological examination (Panda, S. *et al*., 2014; Sciubba, J.J. *et al*., 2005; Srikanth, M.D. *et al*., 2014; Uzawa, N. *et al*., 2015; Kumaran, P.S. *et al*., 2014).

Ameloblastic carcinoma is a tumor that associates with the microscopy the characteristics of ameloblastoma and cytological atypia such as cellular pleomorphism, focal necrosis, nuclear hyperchromatism, a high number of mitoses (Ramesh, M. *et al*., 2011; França, D.C.C. *et al*., 2012; Li, J. *et al*., 2014; Kar, I.B. *et al*., 2014). There are two variants: primary and secondary. The primary type develops de novo, on an odontogenic tumor with histological features of an ameloblastoma and cytological elements of malignancy. The secondary type results from the malignant transformation of a preexisting ameloblastoma, regardless of the presence or absence of metastases (Sciubba, J.J. *et al*., 2005; Kumaran, P.S. *et al*., 2014; Perumal, C.J. 2012).

Its incidence is difficult to assess because f its scarcity. About 100 cases have been reported in the literature (Rizzitelli, A. *et al*., 2014). As a result, the clinical features have not yet been well established, and the appropriate therapeutic modalities remain poorly codified (França, D.C.C. *et al*., 2012; Li, J. *et al*., 2014).

This article was intended to report a case of this rare pathology, presenting the diagnostic aspects and the therapeutic modality adopted, in our context.

OBSERVATION

In January 2011, a 45-year-old woman presented at the Department of Odontostomatology and Maxillofacial Surgery of the University Teaching Hospital Center of Yaoundé (Cameroon) for 23 years of recurrent mandibular tumefaction.

She had undergone four conservative surgical procedures, indicated for mandibular ameloblastoma, followed each time by recurrence. The last intervention would have been 11 years ago, followed by a recurrence three years later.

Following this, the surgeon told her that she did not have a long time to live and that it was no longer possible to operate on her. Resigned, the patient would have joined prayer groups waiting for her death without hope, until she was referred to our service by a former patient operated successfully 14 years ago.

***Corresponding Author:** Ntep Ntep David Bienvenue Email:

The general condition was maintained despite the desperation that was reflected on the patient's face.

The exo-oral examination noted a mandibular swelling ranging from the left gonion to the right gonion, of firm and painful consistency, forming part of the bone, renitated in places. She gave the patient the appearance of a prognath. Cervico-facial lymphadenopathy was not perceptible on palpation. A slight labial-chin paresthesia was evident. The scar of past interventions was visible, left side.

Oral examination revealed vestibular and sublingual filling with elevation of the floor of the mouth. The mucosa was erythematous and budding, with necrotic patches in places. Only teeth 38 and 48 were in place. These exo and endo-oral preoperative aspects are shown in *Figure 1*.

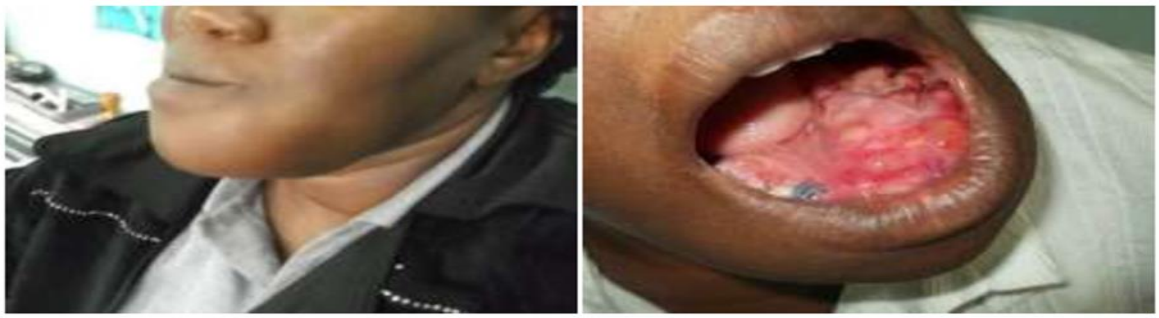

Figure 1: Preoperative view
a) Swelling of the lower part of the face.
b) Erythematous, budding and necrotic appearance of the mucosa in relation to the lesion.

The orthopantomogram performed, shown in *Figure 2* showed an extensive osteolytic lesion of the mandibular body. This osteolysis extended from 38 to 48. The edges of the lesion were clear and the basilar bone cortical was broken.

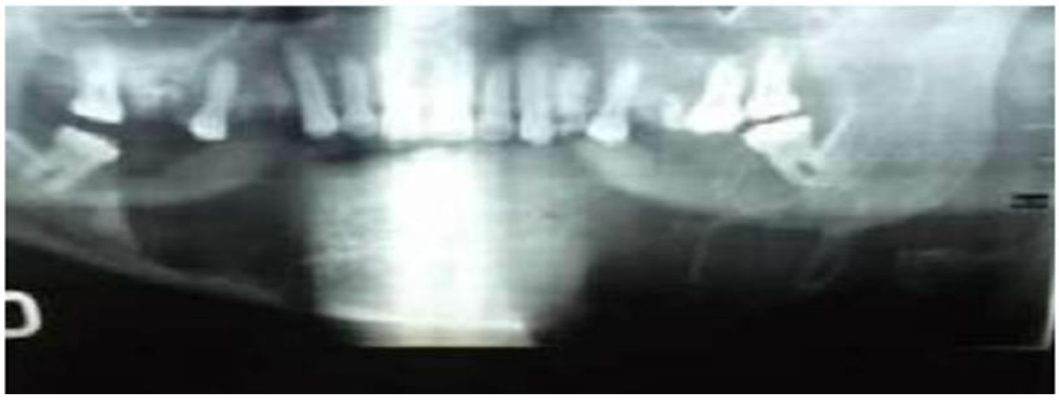

Figure 2: Orthopantomogram showing an osteolytic lesion of the mandibular body.

By anatomopathological analysis, two biopsies performed at two different sites of the lesion favored ameloblastic carcinoma.

At the end of January 2011, the patient underwent surgery under general anesthesia. It consisted of an interruptive resection in a healthy zone of the body of the mandible, on both sides of the lesion. The approach was cutaneous and endo-oral, using the scalpel blades and the electrosurgical knife. The mandibular resection was done behind the teeth 38 and 48.

We used for this an electric micromotor driving bone cutters, under permanent irrigation with saline. The edges of the bone thus resected were cauterized with electrocautery and trichloroacetic acid, to ensure that any ameloblastic cells were all destroyed.

Soft tissue losses in the floor of the mouth were significant. All the mucosa visibly affected was resected, including the fibers of the mylo-hyoid muscle. The digastric, genio-hyoid and genio-glosses muscles have been preserved. The operative specimen of this mandibular resection is shown in *Figure 3*.

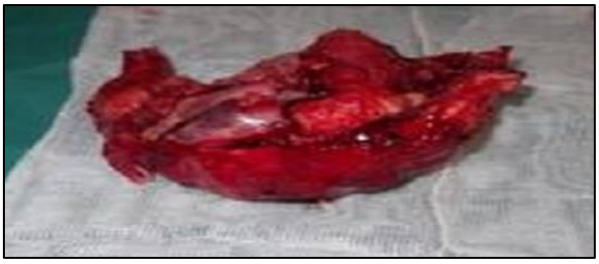

Figure 3: View of the operative piece.

Immediate reconstruction of bone loss by endoprostheses consisting of two rigid metal plates screwed onto the two ascending branches was performed to maintain the mental curve and the morphology of the lower floor of the face. These screwed metal plates fixed in stages on the stumps of mandibular rising branches are shown in Figure 4, through the skin approach.

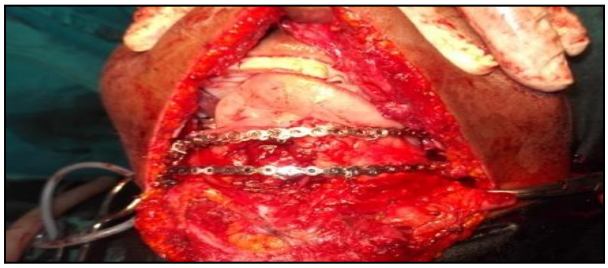

Figure 4: Intraoperative view of the reconstruction of bone loss by two screwed plates.

The anterior endings of the genio-hyoid, genio-gloss, and digastric muscles were sutured in Y through the holes in the lower metal plate. Mylohyoid muscle fibers were sutured through the holes in the upper plate. The mucous and cutaneous sutures were last performed. All of these sutures allowed relatively normal lingual repositioning and acceptable speech.

An antibiotic treatment based on amoxicillin and clavulanic acid, analgesics and anti-inflammatories were administered. The postoperative course was simple. After healing, the patient said she was very satisfied because she recognized herself.

A second pathological analysis of the tissues collected confirmed ameloblastic carcinoma. At the immediate periphery of the resection areas of the operative specimen the anatomopathological examination did not show signs of malignancy.

The patient has been seen every three months to date. After 65 months follow-up, no recurrence was observed. The appearance of the patient is shown in Figure 5. The metal plates used should easily support a dental prosthesis.

Figure 5: Patient after 65 months of follow-up.

DISCUSSION

Ameloblastic carcinoma is an odontogenic malignant tumor of epithelial origin. Its incidence is low, making it a rare and poorly documented pathology (Rizzitelli, A. *et al*., 2014; Sciubba, J.J. *et al*., 2005). It still represents a challenge both at the diagnostic and therapeutic level (Srikanth, M.D. *et al*., 2014). The demographic data concerning this pathology show a male predilection (Kumaran, P.S. *et al*., 2014; Ramesh, M. *et al*., 2011; França, D.C.C. *et al*., 2012), contrary to the case that we presented.

It can reach all age groups, but occurs preferentially during the fifth, sixth and seventh decades of life (Li, J. *et al*., 2014; Kar, I.B. *et al*., 2012; Jindal, C. *et al*., 2010). The mandible is more affected than the maxillary, with a predominance for the posterior regions (Kumaran, P.S. *et al*., 2014; Li, J. *et al*., 2014). These data are consistent with the case we report, including mandibular involvement and age over 40 years.

Ameloblastic carcinoma can develop de novo or result from the malignant differentiation of a pre-existing ameloblastoma (Uzawa, N. *et al*., 2015; Kumaran, P.S. *et al*., 2014 Kar, I.B. *et al*., 2014). This malignant differentiation seems to occur spontaneously, or after repeated surgeries on a recurrent ameloblastoma as was the case for this lady, or develop on those having undergone radiotherapy. However, lesions evolving for long durations as well as those having recurred several times have been reported in the literature, without suffering malignant degeneration. The mechanism of

this malignant differentiation remains unclear to this day (Li, J. *et al*., 2014; Kar, I.B. *et al*., 2012; Perumal, C.J. 2012).

The symptomatology found in the case described is consistent with that observed in malignant tumors of the jaws in general, including swelling of rapid evolution, pain, infiltration of the mucosa covering the lesion, giving a budding, ulcerative, hemorrhagic, with sometimes dysplasias (Srikanth, M.D. *et al*., 2014; Kumaran, P.S. *et al*., 2014; Li, J. *et al*., 2014; Jindal, C. *et al*., 2010). Paresthesia or anesthesia can be found, signifying infiltration or compression of the nerve in the vicinity of the lesion (Perumal, C.J. 2012), as in the patient presented in our description.

Rapid and progressive osteolysis results in thinning and sometimes even perforation of residual bone cortical (Srikanth, M.D. *et al*., 2014; Kumaran, P.S. *et al*., 2014; França, D.C.C. *et al*., 2012; Li, J. *et al*., 2014). This is objectivable to palpation by a renitent aspect of the lesion. The extent of the lesion varies, reaching high proportions such as that observed in this study where the entire mandibular corpus was reached.

The radiographic image obtained by the orthopantomogram is that of an ameloblastoma, notably a multilocular image partitioned into a soap bubble (Srikanth, M.D. *et al*., 2014; França, D.C.C. *et al*., 2012; Jindal, C. *et al*., 2010), as described in our observation. It presents extensive osteolysis, with rupture of the basal cortical. This image explains well the variable consistency found in the clinical examination.

Histology, the only examination to obtain the diagnosis of certainty, reveals cellular atypias characteristic of malignant lesions, all associated with elements in favor of an ameloblastoma. However, the secondary character of ameloblastic carcinoma is not related to histology, but to the presence of preexisting ameloblastoma, as observed in this patient.

Patients, driven to total despair by some practitioners, often find comfort in prayer groups that sometimes promise miraculous healings. The time spent at these places of prayer considerably delays the medical care that alone, has proven to date.

The care is not well codified. In our case, we performed surgery in the rules of carcinology as recommended by Uzawa *et al*., Ramesh *et al*., (2015; 2011), hoping to avoid recurrence. We did not associate lymph node dissection because lymphatic nodes were not perceptible. In addition, there was no adjuvant radiotherapy based on carbon ions as recommended by Jensen *et al*., (2011) lack of resources and technical platform.

Fastening the muscles by sutures through the holes of the metal plates allowed a good muscular spread, an almost anatomical repositioning of the muscles under mylo-hyoidens, thus resulting not only in an acceptable reconstruction of the lower stage of the face. but also to a good lingual repositioning and a satisfactory speech.

We would have used more complex methods like fibrous grafts. But the weakness of our technical platforms and especially the low level of the financial base of the patient did not allow us to go so far. For just over five years this patient has been followed, we have not noticed any signs of recurrence.

CONCLUSION

Amelobelastic carcinoma is a rare condition. For its otherwise difficult diagnosis, a good association between clinical, radiological and anatomo-pathological data is necessary. The treatment is still not standardized , but requires bone resection with safety margins and significant musculo-mucous losses. In our context, surgical treatment with screwed metal plate reconstruction has led to a satisfactory postoperative result.

REFERENCES

1. Rizzitelli, A., Smoll, N.R., Chae, M.P., Rozen, W.M., & Hunter-Smith, D.J. (2014). Incidence and Overall Survival of Malignant Ameloblastoma. *PLOS ONE 10(2),e0117789. doi: 10.1371/journal. Pone .0117789.*
2. Panda, S., Sahoo, S. R., Srivastav, G., Padhiary, S., Dhull, K. S., & Aggarwal, S. (2014). Pathogenesis and nomenclature of odontogenic carcinomas: Revisited. *Journal of oncology*, *2014*.
3. Sciubba, J.J., Eversole, L.R., & Slootweg. P.J. (2005). Odontogenic/ameloblastic carcinomas. In: Barnes, L., Eveson, J., Reichart, P.A., Sidransky, D., editors. World Health Organization Classification of Tumours: Pathology and Genetics Head and Neck Tumors. *Lyon: IARC Press, pp. 286– 295.*
4. Srikanth, M.D., Radhika, B., Metta. K., & Renuka, N.V. (2014). Ameloblastic carcinoma: Report of a rare case. *World J Clin Cases, (February); 2(2),48-51.*
5. Uzawa, N., Suzuki, M., Miura, C., Tomomatsu, N., Izumo, T., & Harada, K. (2015). Primary ameloblastic carcinoma of the maxilla: A case report and literature review. *Oncology Letters 9:459-67.*
6. Kumaran, P.S., Anuradha, V., Gokkulakrishnan, S., Thambiah, L., Jagadish, A.K., & Satheesh, G. (2014). Ameloblastic carcinoma: A case series. *J Pharm Bioallied Sci. Jul; 6(Suppl 1): 208–211.*
7. Ramesh, M., Sekar, B., Murali, S., Mathew, S., Chacko, J., & Paul, G. (2011). AMELOBLASTIC CARCINOMA-REVIEW AND HISTOPATHOLOGY OF 5 CASES. *Oral & Maxillofacial Pathology Journal*, *2*(2).

8. França, D.C.C., Moreira, J.M., Ávila De Aguiar, S.M.H.C., De Carvalhos, A.A., & Goiato, M.C. (2012). Ameloblastic carcinoma of the maxilla: *A case report. Oncology Letters, 4:1297-1300.*
9. Li, J., Du, H., Li, P., Zhang, J., Tian, W., & Tang, W. (2014). Ameloblastic carcinoma: An analysis of 12 cases with a review of the literature. *Oncology letters*, *8*(2), 914-920.
10. Kar, I.B., Subramanyam, R.V., Mishra, N., & Singh, A.K. (2014). Ameloblastic carcinoma: A clinicopathologic dilemma – Report of two cases with total review of literature from 1984 to 2012. *Ann Maxillofac Surg, (Jan-Jun); 4(1), 70–77.*
11. Perumal, C.J. (2012). Ameloblastic Carcinoma of the Maxilla with Extension into the Ethmoidal Air Cells and Close Proximity to the Anterior Skull Base: *A Rare Case Presentation. Craniomaxillofac Trauma Reconstruct, 5:169–74.*
12. Jindal, C., Palaskar, S., Kaur, H., & Shankari, M. (2010). Low-grade spindle-cell ameloblastic carcinoma: report of an unusual case with immunohistochemical findings and review of the literature. *Current Oncology*, *17*(5), 52.
13. Jensen, A. D., Ecker, S., Ellerbrock, M., Nikoghosyan, A., Debus, J., & Münter, M. W. (2011). Carbon ion therapy for ameloblastic carcinoma. *Radiation Oncology*, *6*(1)13.