

Case Report: Residive Ameloblastoma of Mandible

Kania Difa Parama¹, Sunarto Reksoprawiro^{2*}

¹General surgeon Resident - Department of Surgery Faculty of Medicine, University of Airlangga, Dr. Soetomo General Hospital, Surabaya, Indonesia

²Division Head and Neck Surgery - Department of Surgery, Dr. Soetomo General Hospital, Surabaya, Indonesia

E-mail: difakania@gmail.com; sunartokl@yahoo.com

*Corresponding author details: Sunarto Reksoprawiro; sunartokl@yahoo.com

ABSTRACT

Ameloblastoma is a slow growing benign odontogenic tumor, most commonly occurs in the mandible, locally invasive but relatively no complaints and can grow larger and cause damage to the jaw and facial if it is not treated. Proper treatment of ameloblastoma is still controversial, since ameloblastoma recurrence rate is very high, so that clinicians perform a radical treatment with mandibulectomy. This report discussed about management of ameloblastoma of mandible with hemimandibulectomy previously and reconstruction with recon plate. A 60-years-old male complained about having a lump again in his lower jaw, with history of hemimandibulectomy Sinistra and reconstructions with recon plate 2015 with a diagnose ameloblastoma mandible. The patient was hospitalized one day prior surgery. After the hemimandibulectomy, reconstruction performed by placing a K-wire to gain contour of the mandible. The patient was followed up his post operation condition. Hemimandibulectomy and reconstruction with K-wire is the ideal.

Keywords: hemimandibulectomy; recon K-wire; ameloblastoma

INTRODUCTION

Ameloblastoma or adamantinoma, is a benign odontogenic tumor derived from the epithelium, locally invasive, growing slowly and persistently. This tumor is relatively rare, approximately 1% of all oral tumors.¹ This tumor is derived from residu of cells of the enamel organs (dental lamina), the epithelial wall of odontogenic cysts, especially dentigerous cysts and basal cells of the oral mucosal epithelium, the rest of Hertwig sheath present in the periodontal ligaments in the growing tooth or from the enamel organ.

This tumor occurs more in the mandible, i.e. 80%, 75% of this 80% occurs in the area of ramus molar, can occur in all age groups but is most often diagnosed in the third and fourth decades. The typical feature is that of the jaw region associated with the tooth and in the X-ray seen as a multiple cyst lesion.

Ameloblastoma is included in borderline odontogenic tumor because although the histologic features of this tumor show benign signs, clinically the tumor is destructive, has a high recurrence and is reported to have metastatic ability that is said to be a malignant or borderline malignancy tumor.

The Management of this disease can be either conservative or radical method depending on the type, size of the tumor, bone destruction and patient age. Radical management, especially hemimandibulectomy, often causes large facial defects in addition to loss of mandibular function alone. In his study, Young-Ju Jee investigated the emotional and psychological indicators according to vaping usage among

11313 middle and high school students using data from the 2015 Korea Youth Risk Behavior Web-based Survey (KYRBS) [5].

Results showed that students who had used e-cigarette planned to commit suicide within the last 12 months, which was statistically significant ($\chi^2=10.652$, $p=.001$), while students who had used e-cigarettes attempted to commit suicide within the previous year 12 months, which was also statistically significant ($\chi^2=9.638$, $p=.002$) [5].

Interestingly these findings are novel and highlight the potential for the link between vaping and suicidal behavior in adolescents. These persistent findings in teenagers, as well as a slew of case reports tying vaping to suicide, are cause for concern. While the biological mechanisms to explain the link between vaping and suicidality remain unknown, these results suggest exciting directions for future research. In summary, more prospective studies with longitudinal design are needed as this might help mitigate the negative impact of adolescent suicide among E-cigarette users.

CASE REPORT

A 60-year-old man came with another bump in the chin that grew larger since 2011. The patient had a surgical history in 2015 at RS Bedah Surabaya with a diagnosis on a left-side ameloblastoma, performed left hemi mandibulectomy with mini plate installation, but a year after installation the patient reports there's a small bolt that came up while eating from the mouth. (Figure 1)

The general condition of the patient is sufficient, with vital signs within normal limits. On intraoral examination the mass of bumps in the mandible from left corpus to right corpus with 10cm in diameter is unclear, normal skin, mucosal coated surface, color like tongue mucosa, solid consistency, fused with mandible bone. No bloody parts were found. No enlarged neck lymph nodes were found.



FIGURE 1: Clinical patient



FIGURE 2: Radiologic (previous preoperative mandibular panoramic photo 2015)



FIGURE 3: Radiologic (panoramic photo (A))



FIGURE 4: Anteroposterior (B) mandibles when the tumor is residif



FIGURE 5: Head CT Scan

The clinical diagnosis of this patient is a recurrent ameloblastoma of mandible. Then a wide excision of the right hemimandibulectomy, including the reconstruction plate taking in the tumor attached to the left subcondylus was performed in a temporary reconstruction using the kinshnerwire 1.7 to maintain the contours of the mandible. At the time of surgery, a mandibular tumor measuring 7x8x6cm in the left right corpus to a broken plate near the left mandibular subcondyle, the tumor mass is sharply detected with a tissue boundary of 1cm from the tumor, and the fracture plate is released. Performed on the right mandibular resection is measured from 1 cm from the edge of the tumor on the left to right mandibular angle with 1cm margin from the edge of the tumor. Conducted mandibular temporary reconstruction with K-wire and pairs of redon drain, sewing suture layer surgery, mucosa sewn waterproof jacks using polifilament 02 thread, subcutaneous sewn knot using monofilament 03, and skin node using silk thread 05 (figure 6.7, 8.) Intravenous anthraxin 2g intravenous antibiotic prophylaxis administered 1 hour before incision and continued postoperative administration every 12 hours for 1 day. In patients with nasogastric tube for feeding intake of 5-7 days and recommended to keep oral hygiene with antiseptic mouthpiece at least 3 times daily.



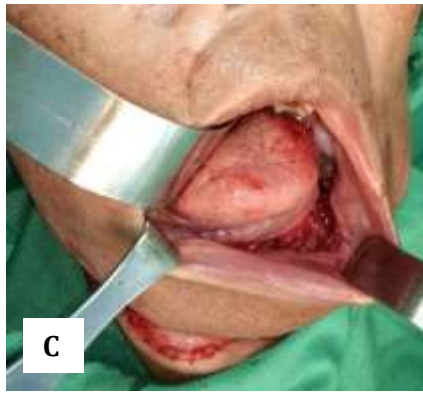


FIGURE 6: Durante surgical removal of tumor mass

Post-surgery patient gets infusion RL 1500 cc for 1 day, then get food and beverage through nasogastric tube as much 1500 calorie and liquid volume 2000 cc/day for 7 days. Patients are given mouthwash at least 3 times a day. On the 5th day post-surgery patient drinking exercise peroral, and on day 7 post-surgery patients are allowed to eat liquid peroral. On the 8th day the patient is allowed to go home.

The results of anatomical pathology examination macroscopically showed a piece of mandible tissue with a total mass weight of 250gr, size 10x10x5cm, long bone ± 2cm smooth surface. On the surface, the papillary solid mass appears, has white-grayish color, fragile solid, and destructive (figure 9). Microscopically, the mandibular tissue with tumor growth consists of the proliferation of odontogenic cells forming the inter canthated ribbons, the cells arranged palisading, the center of which appears in the form of stellate reticulum. The conclusion is to do plexiform ameloblastoma surgery and follicular pattern.

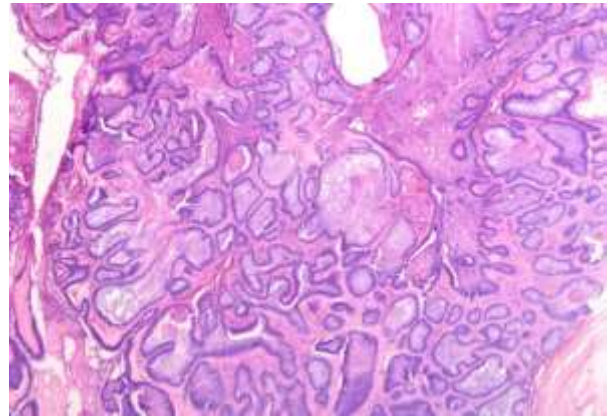


FIGURE 8: Anatomic pathology of plexiform and follicular ameloblastoma.



FIGURE 7: Macroscopic tumor



FIGURE 9: Clinically post surgery

DISCUSSION

Ameloblastoma is the most common odontogenic epithelial tumor. Presentation of ameloblastoma is 1% of all tumors and jaw cyst^{1,8-10}. According to White et al, mentioned in his book *Oral Radiology Principles and Interpretation* that the percentage of ameloblastomais 11%

of all odontogenic tumors.⁵ Meanwhile, according to Caldwell et al mentioned it as a locally malignant tumor with persistent growth. Growth of this tumor is slow and is a benign tumor but locally invasive. In the 1992 WHO also classified ameloblastoma as a benign but locally invasive epithelial odontogenic tumor.^{5,8-10}

Clinical manifestations of ameloblastoma usually grow in all directions, invading soft tissue and destroying bone either by direct pressure or by triggering bone resorption by osteoclasts.⁵ Ameloblastoma does not cause pain, slow growth with a large mass. Other clinical manifestations have felt pain or numbness in the affected area.

The radiological features of ameloblastoma are radiolucent lesions with unilocular or multi locular features with expansion in bone cortex.⁶

There are several kinds of surgery to treat ameloblastoma of the mandible, namely marginal resection, segmental resection, hemimandibulectomy resection. In these patients was performed resection of the mandible in 2014. Radical surgery is preferred by clinicians rather than conservative care because of the high recurrence rate of this tumor. Treatment with radical surgery may reduce the recurrence rate. According to Muller, the recurrence rate by the method is about 15% compared to conservative surgery having a recurrence rate greater than 50%⁹. In our patient was done mandible segmental resection in 2014, but 3 years later recurrence, it is possible resection of mandible which is limited to less radical tumor (distance to tumor <1cm).

The definitive course of action in this patient is re-resection with a 1cm limit from the tumor from the left subcondyle to the right mandible subcondyle. This action causes a large defect in the face, for that done a temporary reconstruction using Kirschner wire. The reconstruction of the mandibular bone depends on the amount of soft tissue available. It is associated with bone resistance, blood supply and systemic factors of the patient.

Patients with effects on the jaw after surgical therapy, can be replaced part of the missing. Miscellaneous techniques can be used for mandibular reconstruction, including using reconstruction plate for temporary reconstruction if within 6 months there is no recurrence; it is recommended for permanent reconstruction using bone stitch.

The goals and principles of mandibular reconstruction are restoring continuity, minimizing malocclusion and mandibular deviation, simultaneous correction of skeletal defects with good soft tissue coverage, minimizing operative and postoperative morbidity. Mandibular reconstruction is viewed from functions and cosmetics, which affect face shape, speech function, chewing and swallowing.⁶

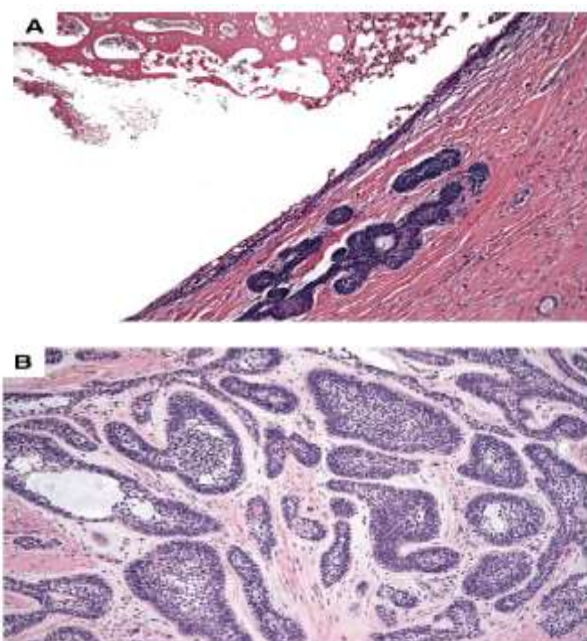
In the early 1980s, it was believed that ameloblastoma was radioresistant. Although some studies have reported adjuvant radiation on a positive (gross and microscopic) margin, in recurrent ameloblastoma, and unresectable ones, the results are poor.⁹ Gardner et al assumed that radiation can reduce the size of ameloblastoma, especially soft tissue composition, and recommend radiotherapy use in poor lesions, especially those located in the posterior maxilla. These lesions respond well to radiation, and a dose of 50-60 Gy in 5 to 6 weeks has resulted in significant regressions, even in large tumors.¹¹⁻¹² Adjuvant radiotherapy may be considered in patients with postresection of mandibular ameloblastoma with positive margins and patients refusing re-surgery.

Ameloblastoma has the possibility of recurrence after surgery. This is due to the nature of the lesion being locally invasive and inadequate operation.

Histopathologically, ameloblastoma resembles the odontogenic / enamel epithelium and ectomesenchymal normal. Odontogenesis consists of chronographic and reciprocal interactions between ectomesenchymal cells, derived from neural crystals, and oral epithelial lining. The ameloblastic epithelium has been hypothesized to arise from (1) cells of the remaining sixelorgan, but also of (2) sheets of Hertwig cells or epithelial epithelium of Malassez, (3) epithelial boundaries of odontogenic cysts, especially dentigerous cysts, (4) basal cells of the oral mucosa, (5) heterotopic epithelia from other parts of the body, such as pituitary.⁹

According to the 2005 WHO classification there are four subtype of ameloblastoma (1) Solid/multisystem is the most common type, comprising 91% of ameloblastoma, (2) unicystic 6%, (3) 2% extraosseous ameloblastoma, and (4) 1% desmoplastic type. Based on the most aggressive clinical / pathologic seen solid / multicystic solids, associated with a recurrence rate up to 90% with conservative surgery such as enucleation and curettage. The unicystic type is the most benign and further classified into the intraluminal and intramural subtypes. The intraluminal unicystic subtype does not show invasion of the connective tissue around, has a lower recurrence rate of two subtypes, and is probably the only histology that can be biased with conservative surgical treatment. In contrast to solid, unicystic and desmoplastic-centered ameloblastomas, encapsulated by bone, and called "central ameloblastoma," peripheral ameloblastomas are present in the extra-osseous and do not involve the underlying bone.⁹

Cellular atypic and mitotic activity are rarely present in any histologic subtype in the ameloblastoma, and any increased parameters increase suspicion of malignant processes such as ameloblastic carcinoma or odontogenic sarcoma. In addition, the microscopic pattern of ameloblastoma is follicular, plexiform, acanthomatous, spindle, basal cell-like, desmoplastic, and granular cells (Figure 10). Polyhilar are the same or different. Subtype of malignant ameloblastoma are not included in the WHO classification. However, these tumors may arise de novo or may be progressive from benign forms.⁹



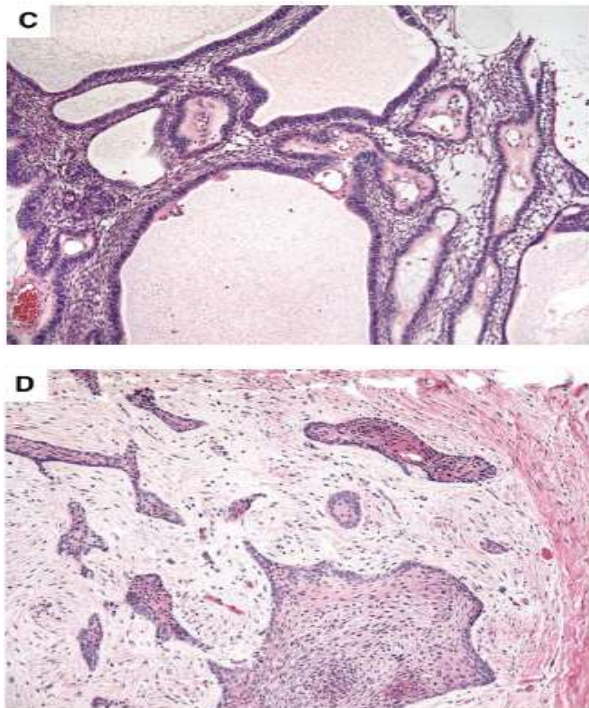


FIGURE 10: Histology of ameloblastoma (A) unicyclic, (B) follicular, (C) flexiform, (D) desmoplast

CONCLUSIONS

Surgery is a standard procedure for ameloblastoma. There are two surgical, conservative and radical options. Conservative surgery is performed bone cavity enucleation / curettage. The advantage of nuclease is an outpatient procedure that can be done by many different service providers (Oral Surgeon and ENT), because it does not require reconstruction. Some studies show a modest 60-90% enucleation recurrence rate, and this treatment modality is currently believed to play no role in multicystic ameloblastoma management.

The "radical" surgical option is the latest standard for ameloblastoma and performed bloc resection with 1-2 cm bone margin and immediate bone reconstruction to assist speech and swallowing function. Bone margin is defined as the distance from the predicted disease-free and safely oncologically safe radiographic margins for osteotomy. Recommended bone margins are 1-1.5 cm for unicystic and 1.5-2 cm for dense / multicystic histologic types. Ameloblastic carcinoma requires 2-3 cm bone margin. Elective neck dissection is not recommended especially in tumors derived from the maxilla.⁹

The prognosis for ameloblastoma depends on the patient's age, tumor size, disease level, tumor location, and histologic type. The recurrence rate is determined by the adequacy of surgical margins and elongation of maxillary ameloblastoma into vital structures (skull base, orbit, paranasal sinus). Maxillary ameloblastoma is more aggressive in terms of rates of disease and recurrence, with a relative hypothesis of the relative thinness of the maxillary cortical bone is a weak barrier for local-regional tumor spread. In addition, recurrence and surgery may lead to an increased risk of surgical complications.⁹

Recurrence after conservative treatment is due to persistent microscopic disease, which grows slowly in the previously evacuated cavity and takes decades to reappear. The "radical" surgical resection showed much better results.⁹

REFERENCES

- [1] Ackerman GL, Altini M, Shear M. The unicystic ameloblastoma: a clinic pathological study of 57 cases. *J Oral Pathol* 1988; 17: 541.
- [2] Kawamura M. Dredging method a new approach for the treatment of ameloblastoma. *Asian J Oral Maxillofac Surg* 1991.
- [3] Cumming CW. Editor. *Otolaryngology- head and neck surgery*. 2ndEd. St. Louis: Mosby; 1993.p.1430-5.
- [4] Neville BW. *Oral and maxillofacial pathology*. Philadelphia: WB Saunders Co, 2002.p.511-37
- [5] Kahairi A, Ahmad RL, Islah Wan L, et al. Management of large mandibular Ameloblastoma
- [6] Bailey BJ, Holt GR. *Surgery of the mandible*. New York: Thieme Medical Publisher; 1987. p.149-205
- [7] Purohit P, Vashishtha A, Sharma S, Trehan M. Management of Plexiform Ameloblastoma : A Case Report. 2014;8(May):122-4.
- [8] Suma M S,Sundaresh K J, Shruthy R, and Mallikarjuna R. Ameloblastoma: an aggressive lesion of the mandible. *BMJ Case Reports* 2008.
- [9] McClary AC, West RB at all, *Ameloblastoma: A clinical review and trends in management*. Berlin Heidelberg: Springer-Verlag Berlin Heidelberg 2015
- [10] Curi MM,LauriaL,PictoDS,Oral surg.Oralmed.Oral patolendod.1997.84.339-44.Amargo hospital.Brazil.
- [11] Garnder DG. Radiotherapy in the management of recurrent ameoblastoma. A long term follows up of a case. *NY State Dent J* 1990; 56:36-37
- [12] Fregnani ER, da Cruz Peres DE, de Almeida OP, Kowalski LP, Soares FA, de Abreu Alves F: Clinico pathological study and treatment outcomes of 121 cases of ameloblastomas. *Int J Oral Maxillofac Surg*;2010;39, 145-149
- [13] Curi MM, Dib LL, Pinto DS: Management of solid ameloblastoma of the jaws with liquid nitrogen spray cryosurgery. *Oral Surg Oral Med Oral Path*; 1997; 84, 339e344
- [14] Costa FWG, Soares ECS, Batista SHB: Criocirurgia no tratamento de lesõesbenignas dos maxilares: revisão de literatura e análise de 103 casospreviamentereportados. *Rev Sul-Bras Odontol*;2010: 7: 208-215,