CASE REPORT

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Aggressiveness tumor: a case report of recurrent ameloblastoma in the mandible

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ABSTRACT

Background: Ameloblastomas are rare odontogenic neoplasms of the mandible and maxilla. They have high recurrence rates if improperly treated. Due to their aggressive nature and high recurrence rate, treatment remains a matter of debate. Complete excision of the lesion with the least morbidity would be the therapeutic challenge.
Case Report: A 67 years old woman complained of swelling on her left jaw four years ago, the jaw was small swelling initially, and then it has grown to the size of a tennis ball. She had a history of similar swelling on the same site 12 years back for which she was operated on (enucleation). On physical examination, there is a mass in the left mandibular bone above the surgical wound, hard, painless and motionless, with a size of 18 cm x 11 cm x 11 cm. Plain x-ray examination showed multiple cystic lesions in the left mandible. In this case, segmental mandibular resection was performed, followed by reconstruction using a K-wire.
Discussion: Many treatment options range from conservative treatment of curettage, enucleation to radical surgical approaches of wide margin excision. Radical treatment approaches have the advantage of lowering the recurrence rates but at the same time pose extremely difficult challenges of reconstruction of the surgical defects.
Conclusion: Ameloblastoma has high recurrence rate if it is not treated properly. At least 1 cm of healthy bone should be removed during surgical procedure beyond panoramic radiograph visible margins. In our patient, though radical surgery eliminated a large possibility of recurrence.

Keywords: Ameloblastoma, aggressive, recurrence.
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INTRODUCTION

Ameloblastoma is a neoplasm derived from benign odontogenic cells but has locally invasive nature. This disease often involves the mandibular area (80%) and maxilla. Conservative therapy has a high recurrence rate. Cusack first described this neoplasm in 1827. Ameloblastoma comes from the ancient French "*amel*" which means enamel, and the Greek word "*blastos*" which means germ or shoot. Over time, this condition has various names, including "*adamantine epithelioma*," "*adamantinoma*," and later "*ameloblastoma*".[1]

Ameloblastoma shows varying geographic prevalence. In China and Africa, ameloblastoma is the number one odontogenic benign tumor most often found and number two in America and Canada (*Odontoma* is the largest). African-Americans are five times more likely to develop ameloblastoma than Caucasians. The global incidence rate is estimated to be around 0.5 / 1,000,000 population per year. Most cases are found in patients aged 30-60 years.[1] Ameloblastoma can be classified as solid, cystic, multicystic, grows relatively slowly and has the appearance of a benign tumor, but invasive and has high recurrence rate.[2–4]

Management of ameloblastoma varies from curettage to radical resection of bone, with or without reconstruction.[2–5] This case report aims to provide an overview of ameloblastoma and the management of this rare disease.

CASE REPORT

A 67-years-old woman was referred from the Haji General Hospital Surabaya, and she came to the Head and Neck Surgery Outpatient Clinic at Dr. Soetomo Hospital. She complained of a lump in her left jaw.

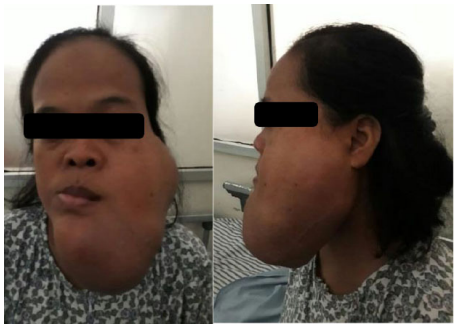

Figure 1. The clinical condition of the patient. Front view (left) side view (right).

The lump appeared four years ago, initially a small lump the size of the patient's hand thumb. Over 3.5 years, the lump grew slowly to a size of about 8 cm in diameter. However, within six months, the lump grew faster and doubled in size. The lump is painless, the patient can still eat soft

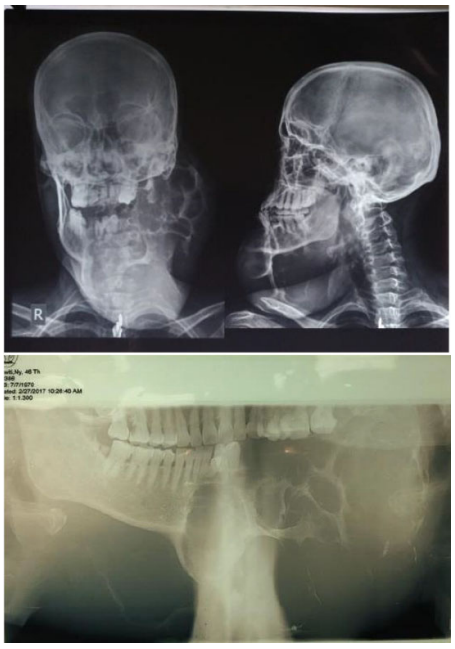

Figure 2. The X-ray head AP/Lateral (left) x-ray photograph panoramic (right)

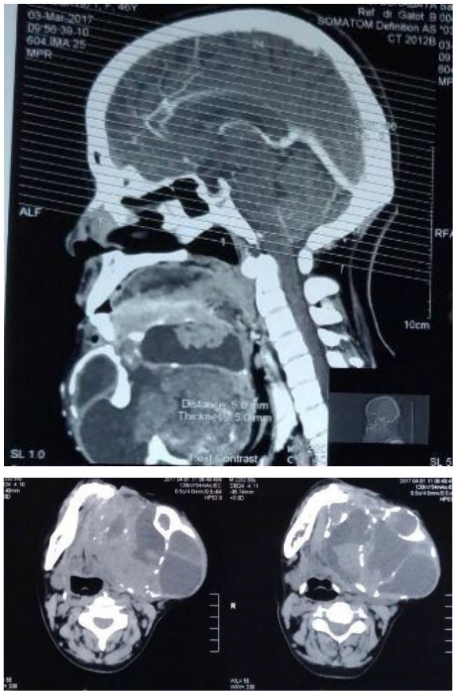

Figure 3. CT scan of mandible

food, and there is no shortness of breath. In the last four months, the patient also complained of blood mixed in the saliva that came out of the mouth every day.

From the history, the patient has experienced complaints of lumps in the same place before. The patient's left jaw lump has been experienced since 12 years ago, a lump the size of rambutan fruit. One year later, the surgery for removing the lump was carried out at the Serang Banten Hospital. Apart from removing the lump, the area around the lump was scraped, and no resection was done on the jawbone. Then the patient underwent outpatient control for two years at the ENT Outpatient Clinic of Serang Banten Hospital. Eight years later, the lump grew back in the same place, and it was felt to be expanding and getting bigger. The patient had time to go to Serang Hospital for control and was advised to undergo surgery, but the patient refused due to economic reasons. The patient had denied a family history of tumors.

From the physical examination, it was found that the patient's general condition was insufficient, vital signs and other general status were within normal limits, on the left mandibular region of the visible mass in the scar area.

On palpation, the mass is partly solid and mostly dense, spongy, uneven surface, humped, with a size of 18x11x11 cm, there is no tenderness, fixed to the base, the skin above the mass can be moved, the margins are not clear. In the intra-oral region, the inspection revealed a hyperemic mass, humped, and no ulcer. The mass appeared to fill the left intra-oral side and pushed the tongue to the opposite side. There were caries on the 1st and 2nd premolar teeth. On palpation, the mass was solid, painless, fixed to the base (see figure 1.)

There were no enlarged lymph nodes in the neck on physical examination of the head and neck region. Examination of the chest region is within normal limits.

On laboratory examination, it was obtained Hb 10.1 mg/dl with 17.370 leukocytes and 490.000 platelets. Other blood tests were within normal limits. The patient's hemoglobin level was at lower normal limits. No preoperative transfusion was performed in this patient, but two bags of PRC blood were prepared for the operation.

X-ray examination of the head from the AP/Lateral view from the previous hospital illustrates soft tissue tumors with left mandible bone destruction. A panoramic radiograph showed multiple cystic lesions in the left mandible (Figure 2). On the CT scan of the head with contrast, a soft tissue mass was obtained with a cystic part with a mass size of 10.39x8.77x9.98 cm accompanied by destruction of the mandible on the left from the column, angle to the mandibular body and no intracranial mass infiltration. The CT scan shows a primary bone tumor (Figure 3). The preoperative chest x-ray showed no abnormalities in the heart and signs of metastases in the lung and projected bone. There was no sign of metastases in the liver, lymph nodes of the aorta, and other projected organs on upper-lower abdominal ultrasound examination.

In this patient, the FNAB was examined. A performed puncture on the left mandible obtained a reddish serous fluid of approximately 12 ml. From the microscopic image, it was found that the smear contained the distribution of inflammatory cells, histiocytes, mononuclear, and PMN with a large mucoid matrix background. In the results of these examinations, it was found that the cystic tumor suggested ameloblastoma.

The operation was performed on April 3rd, 2017. The patient was placed on the operating table in a supine position under general anesthesia.

Intraoral disinfection was chlorhexidine and done with liquid extra-oral disinfection performed by administering 75% alcohol solution. Then an incision was made in the left submandibular area surrounding the previous postoperative scar. A multilobular tumor mass measured 17 x 16 x 9 cms, starting from the left condyles to the two right incisors. The mass of the tumor was sharply released from the surrounding tissue. Then performed a mandibular resection of the second premolar with a margin of 1 cm from the tumor. The mandible to the left mandibular condyles is removed. Reconstruction was carried out using a K-wire (Figure 4-8).

The operation took 6 hours with bleeding of ± 1,000 ml. Postoperative anemia was corrected, and three bags of PRC transfusion were given. There are no other complications. The drain was removed on the 6th postoperative day. The patient was then discharged on day seven post-surgery and continued with postoperative evaluation from Head and

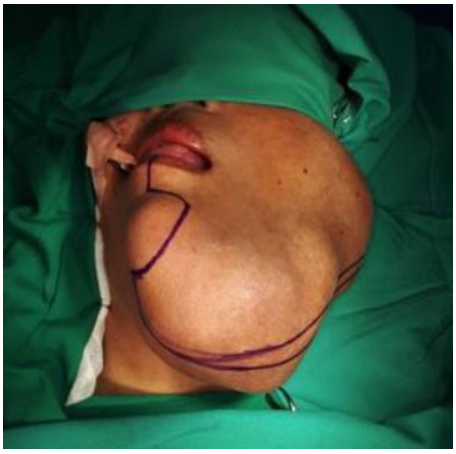

Figure 4. Incision design

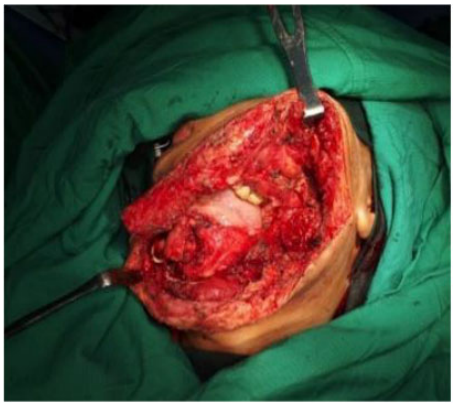

Figure 5. A multilobular tumor on the left mandible

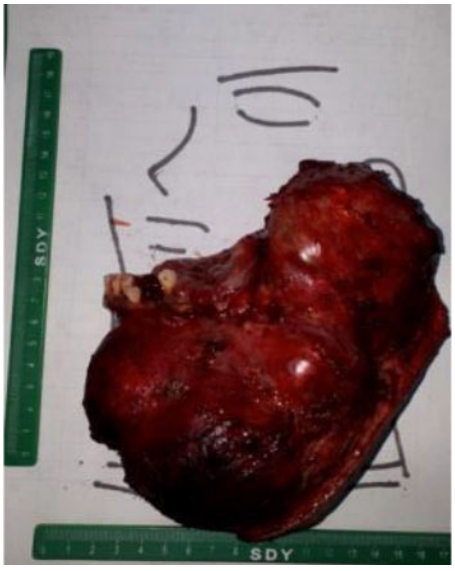

Figure 6. The mass of the tumor and teeth in the left mandible after hemimandibulectomy was performed

Neck Surgery Outpatient Clinic at Dr. Soetomo General Hospital (Figure 9).

Pathology examination results showed that macroscopically a tissue weighing 850 grams, size 21x11x10 cm, an impression of a mass with 18 x10 x 9 cm as seen in Figure 5. The distance between the mass and the end of the proximal bone resection is 2 cm. The distance between the mass and the distal bone resection tip is 1 cm. In the preparation, also obtained seven teeth. The slices appear multicystic with a diameter of 1.5 - 4 cm, filled with a thick liquid partially brownish color. The impression of the mass has destroyed the bone.

The results of the microscopic analysis showed pieces of benign neoplasm tissue, consisting of proliferation of odontogenic epithelial cells, with a round nucleus, monotone, fine chromatin, arranged palisading with stroma stellate reticulum on the lumen forming a follicular pattern. The distance between the tumor and the posterior resection edge is 2 cm, and the distance between the tumor and the anterior resection edge is 1 cm. The conclusion from the results of the PA analysis is follicular type ameloblastoma.

DISCUSSION

Ameloblastoma is a benign ectodermal tumor originating from odontogenic tissue and is the most common odontogenic epithelial neoplasm. This condition is 1% of all tumors or cysts in the jaw. About 75-80% of ameloblastoma occurs in the mandible, especially in the bicuspid and molar areas and at the mandibular angle. The rest, or about 20-25%, occurs in the maxillary area.[1–3] Tumors usually occur at the age of no difference between women and men. Ameloblastoma has been reported to metastases to the lungs, brain and bone, but metastasis is extremely rare. Local aggressiveness or localized infiltration is common, and recurrences frequently occur after inadequate surgical measures.[6]

Clinically, ameloblastoma can be classified into four groups: unicystic, solid or multicystic, peripheral, and malignant. Cystic union ameloblastoma usually presents as a "cystic" lesion with the intraluminal or intramural proliferation of the cystic layer. Radiographically, this will appear as a slow-growing circular radiolucent image. Multiple cystic ameloblastomata can infiltrate adjacent tissues and can regrow and even metastasize. Its prevalence is in the age group slightly older than that of unicystic ameloblastoma. Radiographically, it generally looks unilocular or multilocular. Peripheral ameloblastoma predominantly appears on the alveolar mucosa. That is a soft tissue version of ameloblastoma but can also involve the underlying bone.

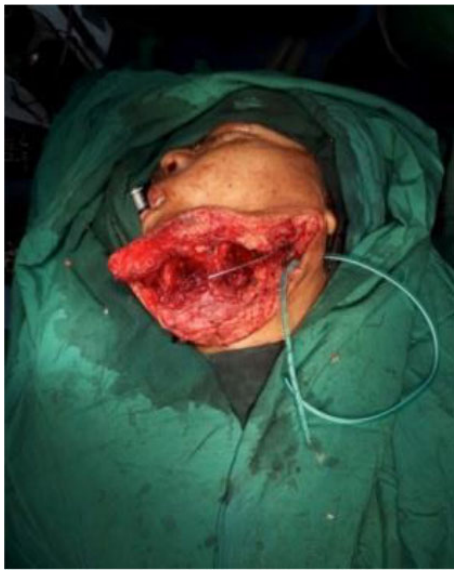

Figure 7. K-wire reconstruction and drain placement

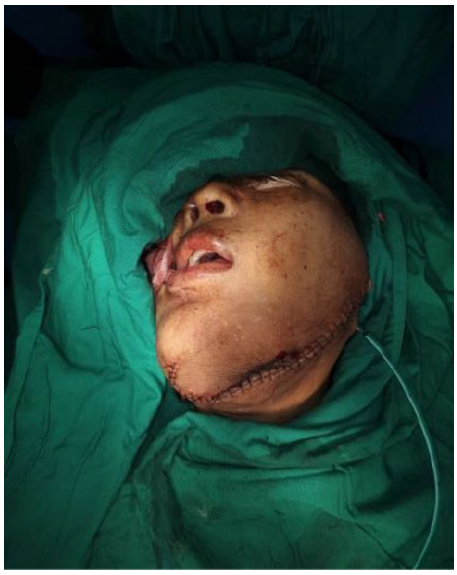

Figure 8. Wound closure

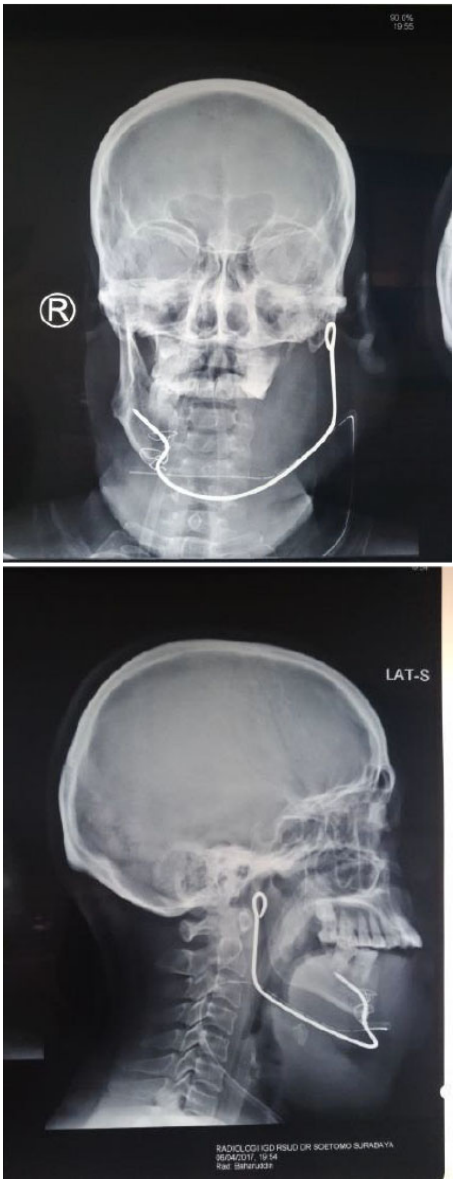

Figure 9. X-ray of the AP/Lateral head for postoperative evaluation

Malignant ameloblastoma is a rare type. It is defined as ameloblastoma that has spread but still retains the classic microscopic appearance.[5]

Determination of tumor size preoperatively is very important to obtain an adequate level of resection. Determination of this limit can be obtained using x-rays, CT scans, MRI and angiography. The use of 3 dimensional the 4th decade to the fifth decade of life and diagnostic imaging can better depict tumor sections by combining the sliced tissue images coherently. Preoperative embolization helps reduce the rate of blood loss during surgery and assists other experts in determining tumor margins.[6]

Various treatment algorithms for ameloblastoma have been reported, But, a universal approach has not been made and remains controversial. The treatment algorithm chosen depends on size, anatomical location, histological variant, and anatomical involvement. (ameloblastoma management algorithm) Treatment of ameloblastoma can vary from curettage to wide excision of the bone, with or without reconstruction. Radiological examination is very important as a postoperative evaluation because more than 50% of recurrences occur in the first five years postoperatively.[2]

In this case, this patient had a history of tumor and suspected ameloblastoma in the same area 12 years ago. Then the patient underwent a conservative operation in the form of enucleation and curettage at Serang Hospital, Banten. Then the patient complained about the appearance of a lump in the area after the surgery eight years later, which was felt to be getting wider and bigger.

In the latter case, surgery was performed with segmental resection of the mandible with a tumor-free margin of 1 cm. Imaging examinations determine tumor boundaries in panoramic x-ray examinations and CT scans.

From the panoramic x-ray examination, it was found that there was a multilobular tumor in the left mandible that extended to the right incisor.[2]

Meanwhile, the CT scan showed a soft tissue mass image with a cystic part with a mass size of 10.39 x 8.77 x 9.98 cm accompanied by destruction of the left side of the mandible starting from the column, angle to the mandibular body, and there was no picture of intravenous mass infiltration cranial. The operation was a left hemimandibulectomy, which removed the tumor tissue with the mandibular bone and attached teeth. The incision margin is carried out on the right second premolar to remove the left condyles. That is done according to the standard, namely, meeting the minimum limit of 1 cm tumor-free edge.[7]

Research by Dandriyal et al. (2011) shows that conservative therapy has a high recurrence rate, reaching 60% compared to 10% in therapy with radical resection. In that study, enucleation with curettage was done in 10 cases, out of which 6 showed recurrence, whereas one case in the surgical group showed recurrence.[5]

CONCLUSION

Ameloblastoma is a benign odontogenic neoplasm of the maxilla and mandible with a high recurrence rate if they are not treated properly. At least 1 cm of healthy bone should be removed during surgical procedure beyond radiographically visible margins. In our patient, though, radical surgery eliminated a large possibility of recurrence. A temporary mandibular reconstruction with K-Wire is a cheap and aesthetically acceptable alternative before definitive reconstruction with autogenous bone grafts or prosthetics.

DISCLOSURE

Conflict of Interest

The author declared that there is no conflict of interest regarding the publication report.

Ethical Consideration

The patient has been signed the informed consent and agrees to publish their data as a case report article.

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None.

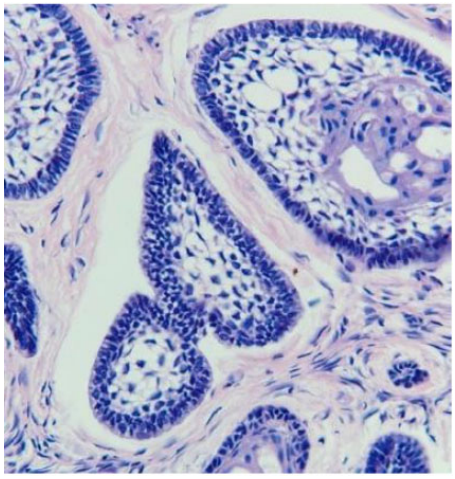

Figure 10. Follicular type ameloblastoma with odontogenic epithelial cells arranged palisading with the stroma reticulum forming a stellate pattern

Author Contribution

All authors equally contributed to the study from initial assessment, operative procedure until reporting the outcome.

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