

(CASE REPORT)



Residual sinonasal ameloblastoma

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Abstract

Ameloblastoma is the most common odontogenic tumour of the mandible. This disease can occur in the maxilla, but it is very rare. This tumour originates from the epithelium involved in the process of tooth formation, but the trigger for neoplastic transformation in the epithelium still is not known with certainty. Ameloblastoma is defined as a locally aggressive benign tumour with a high probability of recurrence. These tumours have several variations in histopathological appearance, but the types most often seen are the follicular and plexiform types. In most cases, ameloblastoma is locally invasive, usually asymptomatic and slow growing.

It has been reported that a 45-year-old female patient with residual sinonasal ameloblastoma had a lateral rhinotomy performed using the Weber-Ferguson approach with good surgical results. Sinonasal ameloblastoma is a rare case with a high recurrence rate after surgery. Ameloblastoma can be diagnosed based on anamnesis, physical examination, and supporting examinations consisting of a CT scan and histopathology. Surgery is the standard treatment for ameloblastoma. A wide excision should be performed with 2 cm of normal bone removed from around the tumour margin to prevent the re-occurrence.

Keywords: Residual Sinonasal Ameloblastoma; Wide excision; Lateral rhinotomy; Midfacial Degloving; Odontogenic

1. Introduction

Ameloblastoma is the most common odontogenic tumour of the mandible. This disease can occur in the maxilla, yet it is very rare. This tumour originates from the epithelium involved in the process of tooth formation, but the trigger for neoplastic transformation in the epithelium is still unknown. Ameloblastoma is defined as a locally aggressive benign tumour with a high probability of recurrence. These tumours have several variations in histopathological appearance, but the types most often seen are the follicular and plexiform types. In most cases, ameloblastoma is locally invasive, usually asymptomatic and slow growing [1,2,3]

Ameloblastoma accounts for 1% of all cases of oral tumours and 9-11% of odontogenic tumours. Approximately 80% of ameloblastoma locations occur in the mandible, especially the molar and ramus areas, and the remaining occur in the maxilla. Cases are often found in adults aged 30-40 years, with an even ratio of men and women. The incidence of ameloblastoma is estimated at 0.6 cases per 1 million population. As many as 8.7 – 15% of ameloblastoma cases also found in children [2,4]

Based on WHO and the International Agency for Research on Cancer, it is included in the category of benign tumours that contain odontogenic epithelium and stromal fibrosis. Ameloblastoma is further classified into multicystic, extraosseous/peripheral, and unicystic. Multicystic ameloblastoma was found in 83% of cases [2,5]

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The primary treatment for ameloblastoma cases is surgery. The choice of surgical technique is still controversial. In general, the determination of the method depends on several factors, such as the type of tumour and clinical manifestations [2,4,6]

The incidence of recurrent ameloblastoma increases in cases treated with conservative surgery. Multicystic ameloblastoma has a higher recurrence rate than unicystic ameloblastoma. It is thought to be due to the more aggressive nature of multicystic ameloblastoma [7,8]

2. Case Report

A 45-year-old female patient came to the ENT-HN outpatient clinic at Prof. dr. I.G.N.G Ngoerah Hospital Denpasar and complained that her right nose was blocked three months ago. Complaints are felt to be getting worse and accompanied by a feeling of fullness in the right cheek and intermittent headaches. There are no complaints of discharge from the nose or shortness of breath. The patient said she had the same complaint before and had a right midfacial degloving surgery performed in December 2021. The histopathological examination during the previous operation concluded a follicular type ameloblastoma. On general physical examination, the patient's general condition was good with compos mentis awareness, blood pressure 110/70 mmHg, pulse 84 x/minute, respiration 20 x/minute, axillary temperature 36.7°C. On examination of the nose, there was a mass in the right nasal cavity, while the left nasal cavity looked broad. Examination of mucosa, septum and left nasal concha appeared normal, whereas the right nose was difficult to evaluate. On examination of the ear and throat, there were no abnormalities.

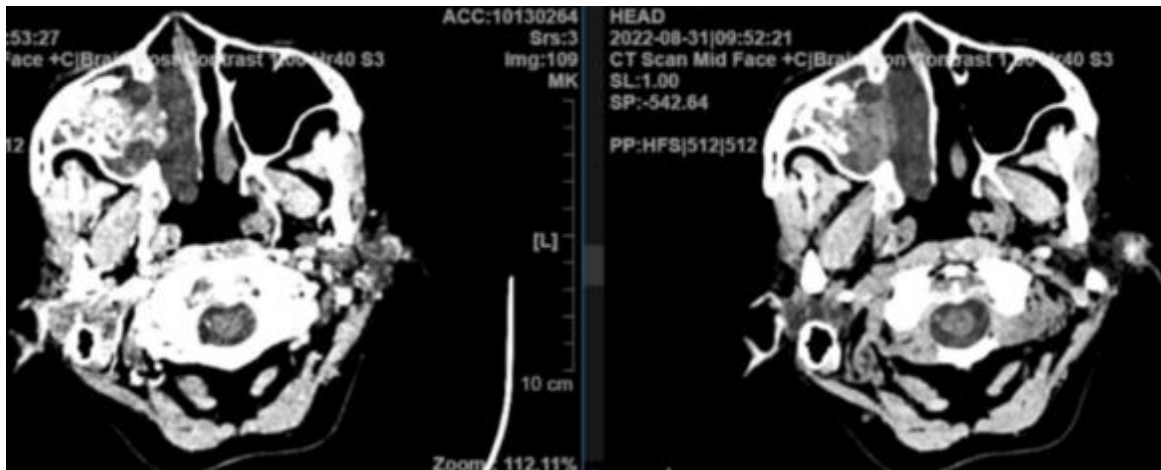


Figure 1 Nasal and paranasal sinuses CT scan with contrast

A CT scan of the nasal and paranasal sinuses, axial and coronal sections with 2 mm slices with contrast, showed an expansive lytic lesion with a septal component and geographic type of calcification, a narrow transitional zone, in which a soap bubble appearance appeared on the right maxillary bone without periosteal reaction or involvement of the surrounding soft tissue with an extension of the lesion to the right maxillary sinus, destroying the medial wall of the right maxillary sinus and extending to the right nasal cavity which is suspected of destroying the medial nasal concha and right inferior, supports the illustration of ameloblastoma. Based on the anamnesis, physical examination and supporting examinations, the patient was diagnosed with residual right sinonasal ameloblastoma. The patient was then prepared for right lateral rhinotomy surgery with the Weber Ferguson approach.

In preparation for the surgery, we performed complete blood tests, chest x-rays and consultation with the anesthesiologist to assess the feasibility of the operation. Laboratory examination results were Hb 12.97 gr/dL, leukocytes 8.67/mm³, haematocrit 38.11%, platelets 303.400 mm³, PT 13.6", APTT 30". Laboratory results are within normal limits. The chest x-ray results also showed no abnormalities. The anaesthesiologist agreed to surgery with physical status ASA 1.

On September 25th 2022, a lateral rhinotomy was performed using the right Weber-Ferguson approach. A bone-density tumour mass was seen in the right maxillary bone extending to the ethmoid sinuses, and the right nasal cavity measuring 6x4x2 cm. The tumour mass was sent to the anatomical pathology department for histopathological examination.



Figure 2 Surgery result

Postoperative therapy consisted of ceftriaxone 1 gram every 12 hours intravenously, tranexamic acid 500 mg every 8 hours intravenously, analgesics according to the anesthesiologist and nasal tampons was maintained to control bleeding.

On the second postoperative day, we removed the nasal tampon. There was no active bleeding and complaints of nasal congestion were reduced. The patient was discharged with cefixime 200 mg every 12 hours intraorally and paracetamol 500 mg every 8 hours intraorally. On November 1st, 2022, the patient went to the ENT-HN outpatient clinic for the first time. Complaints of nasal congestion have decreased with minimal pain in the surgical wound. On anterior rhinoscopy, a broad nasal cavity was observed. The therapy was still the same as previously but added nasal washing with NaCl 0.9% solution.

Two weeks to one month after surgery, the patient is routinely monitored at the outpatient clinic. The patient has no complaints of nasal congestion, while the surgical wound has been well-maintained. Postoperative anatomic pathological examination results showed ameloblastoma histomorphology.

3. Discussion

Ameloblastoma is an odontogenic tumour. Based on various literature and studies that have been performed, most cases of ameloblastoma located in the mandible, and only 20% of cases are found in the maxilla. In this patient, we found ameloblastoma in the maxilla, which extended to the paranasal sinus and nasal cavity which is very rare. According to Dhanuthai et al., ameloblastoma located in the maxilla would potentially expand to surrounding structures, including the paranasal sinuses and nasal cavities [1]

The patient came with complaints of nasal congestion and a feeling of fullness in the right cheek. It can occur due to the tumour location in the nasal cavity and maxillary sinuses. Tumours in the nasal cavity can cause complaints of nasal congestion due to poor air passage. In addition, involvement of the maxillary sinus also results in impaired mucus drainage, which exacerbates patient complaints and also causes the full-feeling in the cheeks. Masthan et al. said the symptoms of patients with ameloblastoma are usually asymptomatic. However, in ameloblastoma with a large size, facial deformity, nasal congestion, and pain can be found [5]. Effiom et al. stated that the incidence of ameloblastoma was not influenced by gender, where the ratio between men and women was 1: 1 and most often occurred at the age of 30-40 years. In this case, when ameloblastoma was first diagnosed, the patient was 44 years old [2].

The diagnosis of ameloblastoma can be enforced by supporting examinations such as CT scans and histopathological examinations. On the CT scan of this patient, a soap bubble appearance was obtained on the right maxillary bone without periosteal reaction or involvement of the surrounding soft tissue, an extension of the lesion to the right maxillary sinus was observed which destroyed the medial bone of the right maxillary sinus extending to the right nasal cavity with suspicion of destroying the medial and right inferior nasal suggesting the ameloblastoma. According to Kreppel et al., a soap bubble appearance is characteristic of multicystic ameloblastoma [9]. The histopathological illustration of patients with ameloblastoma according to Brown et al. shows a fibrous stroma with islands or masses of proliferating epithelium [10]. Histological variants include follicular, plexiform and acanthomatous types. On histopathological examination, this patient previously showed a follicular kind of ameloblastoma. A previous history of ameloblastoma was one of the considerations for diagnosing residual ameloblastoma in this patient.

According to Choi et al., the recurrence of ameloblastoma cases is quite high, with a 50-70% chance after surgery [11]. The radical procedure is the standard for the management of ameloblastoma. One goal is to reduce the risk of recurrence. Nonetheless, the risk of recurrence still exists. This case is one of the patients who experienced recurrences after midfacial degloving in 2021. According to Addel et al., recurrences are predicted to occur five years after the first case [12]. However, in this case, post-operative recurrence occurred only one year postoperatively. One of the factors that caused the recurrence to occur more quickly was the location of this patient's ameloblastoma. Removal of tumour tissue in maxillary ameloblastoma is relatively more difficult than ameloblastoma that occurs in the mandible because it often involves other surrounding structures. The rest of the tumour tissue can develop again into a residual mass. Laborde et al. stated that ameloblastoma is not radiosensitive and surgery is the treatment of choice [13]. In this case, a lateral rhinotomy was performed using the Weber-Ferguson approach because there had been a recurrence at the same location of the lesion, namely the right maxilla, one year after the first surgery. The Weber-Ferguson technique is more radical when compared to the midfacial degloving procedure because the incision is wider but also causes fewer aesthetic scars on the face. Nonetheless, the Weber-Ferguson procedure was performed because the previous midfacial degloving technique still left tumour tissue. It is expected that this technique will widen the visual field during surgery so that the tumour tissue will be completely removed.

4. Conclusion

We reported a 45-year-old female patient with residual sinonasal ameloblastoma that had a lateral rhinotomy performed using the Weber-Ferguson approach with good surgical results. Sinonasal ameloblastoma is a rare case with a high recurrence rate after surgery. Ameloblastoma can be diagnosed based on anamnesis, physical examination, and supporting examinations consisting of a CT scan and histopathology. Surgery is the standard treatment for ameloblastoma. A wide excision must be performed by removing 2 cm of normal bone from the tumour margin to prevent a recurrence.

Compliance with ethical standards

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Disclosure of conflict of interest

The author reports no conflicts of interest in this work.

Statement of informed consent

Informed consent was obtained from participant included in the study.

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