Soft Tissue Recurrence of Ameloblastoma after Mandibular Resection

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Abstract

Ameloblastoma, a local invasive odontogenic benign tumor, has a high rate of recurrence in the long term. The present research is a retrospective study of patients with mandible ameloblastoma after receiving mandibulectomy. The aim of this research is to evaluate the recurrence of ameloblastoma after mandibulectomy within 1–5 years.

Twenty-eight patients who had been treated with mandibulectomy with 1–1.5 cm safety margins to the healthy tissue in 2010–2015 were recalled for physical examination and panoramic X-ray. Surgery data, including patient age, gender, size of ameloblastoma, type of ameloblastoma, type of mandible resection, and type of reconstruction, were obtained from surgery reports and medical records. Suspected recurrence was diagnosed through incisional biopsy and confirmed by histological check-up.

Among the 28 patients included in this research, only one case of acanthomatous ameloblastoma showed recurrence in the soft tissue. No recurrence involving bones was observed. Mandibulectomy of ameloblastoma with 1–1.5 cm extension to the healthy bone is adequate to manage ameloblastoma. However, surgeons must pay attention to the safety margin during soft-tissue excision, especially in the case of aggressive acanthomatous ameloblastoma.


Keywords: Acanthomatous ameloblastoma, mandible resection, reconstruction, safety margin, soft tissue recurrence.

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Introduction

Ameloblastoma is an odontogenic epithelial tumor originating from enamel organs, the dental lamina, dentigerous cysts in the epithelium, or basal cells in the oral mucosa. Ameloblastoma constitutes 1% of all oral tumors and cysts and often occurs in patients aged 30–40 years. World Health Organization (WHO) defines this type of lesion as a locally invasive polymorphic neoplasm. Ameloblastoma lesions are histologically benign but behave like invasive tumors with slow growth. The tumors are generally asymptomatic, except when they reach a size large enough to cause expansion and perforation in the surrounding soft tissues. Ameloblastoma generally occurs in the mandible (80%) but can also be found in the canine and molar regions of the maxilla (20%).¹,²

The optimal treatment for ameloblastoma remains highly debated because of the local aggressive nature of the tumor and its high rate of recurrence after action. The challenge for therapeutic intervention in this case is to achieve complete excision of the lesion with the lowest possible morbidity. A surgeon must be able to determine the tumor location, size, and type and assess the age of the patient to achieve good treatment results. Various methods have been developed to treat ameloblastoma. Minimally invasive actions that can be performed include curettage, enucleation, or marsupialization. Radical actions include marginal mandibulectomy, segmental mandibulectomy, and hemimandibulectomy with reconstruction. Among the histopathological types of ameloblastoma, the solid/multicystic type is the most aggressive type with the highest recurrence rate after excision while the unicystic type has the lowest recurrence rate. Minimally invasive actions also show a high level of recurrence. Therefore, such actions must be combined with the application of

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Carnoy’s, cryotherapy, or diathermy solutions to reduce the recurrence rate of the tumors. Peripheral ameloblastoma occurring in the gingiva and alveolar mucosa usually responds well to minimally invasive measures. This study aims to measure the level of recurrence of ameloblastoma after mandibulectomy.

Materials and methods

The subjects of this retrospective study were 28 patients who had been treated for ameloblastoma in 2010–2015. All patients provided informed consent prior to participating in this study. Ethical approval was obtained from the Department of Ethics and Advocacy Unit of the Faculty of Dentistry, Universitas Gadjah Mada. All patients were positively diagnosed with mandibular ameloblastoma by biopsy examination, and all tumors were excised by marginal mandibulectomy, segmental mandibulectomy, or hemimandibulectomy. Tumor excision was carried out by extending 1–1.5 cm to the healthy tissue from the border of the tumor wall.

Data were obtained from medical records, physical examinations, and panoramic X-rays and included age, gender, size, and type of ameloblastoma, type of mandibular resection, type of reconstruction after mandibulectomy, recurrence, and onset. If recurrence was suspected, the patient was subjected to incisional biopsy for diagnosis and confirmed by histology examination.

Results

The patients consisted of 12 males and 16 females aged 15–68 years (mean, 41.1 years). Fourteen lesions were medium sized (50%), seven (25%) were small, and seven (25%) were large. The types of ameloblastoma found were multicystic (17; 60.7%), acanthomatous (2; 7.1%), mixed (2; 7.1%), (follicular 2; 7.1%), unicystic (2; 7.1%), peripheral (1; 3.5%), and plexiform (1; 3.5%). These cases were treated by marginal mandibulectomy (6; 21.4%), segmental mandibulectomy (11; 39.3%), and hemimandibulectomy (11; 39.3%), as shown in Table 1.
Figure 3. Histopathology of the recurrence case. The tumor tissue consists of mucosoid stellate cells surrounded by palisade and basaloid cells infiltrating connective tissues.

Only one recurrence case in the soft tissue was detected. Interestingly, no recurrence involving bones was observed. The recurrence case occurred 3 years after radical hemimandibulectomy. Clinically, the tumor showed soft buccal swelling; it was moveable, painless, and did not bleed easily (Figure 1). Panoramic radiography showed no bone involvement (Figure 2). An incisional biopsy in recurrence lesions results in acanthomatous ameloblastoma. This is the same as the type of ameloblastoma that occurs before (Figure 3).

The histopathology of the recurrence case showed tumor tissues consisting of mucosoid stellate cells surrounded by palisade and basaloid cells infiltrating connective tissues.

Discussion

Ameloblastoma is a benign odontogenic tumor originating from the remnants of the Malassez epithelium. Ameloblastomas grow slowly but could aggressively develop into the trabecular bone causing damage to hard tissue.

The results showed that ameloblastoma recurrence in only 1 out of 28 patients. Ameloblastoma is an invasive tumor. Tumor cells can infiltrate tissues extensively and may not be detected when the tumor is excised clinically or radiologically. Even when adequate excision measures have been carried out, undetected microscopic cells can grow back into new tumors.

Recurrence occurs in acanthomatous ameloblastoma. Among the histopathological types of ameloblastoma, acanthomatous ameloblastoma is the most aggressive and has the highest recurrence rate. By contrast, unicystic ameloblastoma has the lowest recurrence rate.

The high risk of recurrence of acanthomatous ameloblastoma is attributed to its high MMP-2 expression. Studies have shown that high MMP-2 expression and activity are related to increased aggressive and recurrence behaviors in ameloblastoma. The action performed on the patient with recurrent ameloblastoma was hemimandibulectomy.

The results of this study showed recurrence in the soft tissue of only one case. No bone recurrence was observed in the panoramic study. Besides histological type, the risk of ameloblastoma recurrence is influenced by the type of surgery undertaken. Ameloblastoma can recur in remaining tumor tissues in the jaw bone and the surrounding soft tissue or through intraoperative contamination. Er et al. revealed that cell surface antigens, such as CD56 or neural cell adhesion molecule and CD147 or neurothelin, play roles in the aggregation, organization, and metastatic properties of tumor cells. However, in the present case, no tumors left in the jawbone were found in the panoramic radiograph (Figure 2) and recurrence was observed in the posterior region, not in the soft tissue around the osteotomized region. This result agrees with that of Lin et al., who showed six recurrence cases diagnosed as soft-tissue ameloblastoma in their retrospective study spanning a 15-year period.

Recurrence occurs in medium-sized ameloblastomas (6 dental units). The rate of ameloblastoma recurrence generally increases with increasing lesion size. However, size is not the only determinant of recurrence. For instance, a patient’s ameloblastoma type also determines his/her risk of recurrence. In addition, the integrity of the tumor wall and whether resection or enucleation was carried out determine recurrence.

In the present study, recurrence was controlled by radical resection with 1–1.5 cm extension to the healthy tissue. Recurrence occurred 3 years postoperatively in this study. Post-therapy recurrence can occur within 1–45 years after enucleation. All patients who have undergone ameloblastoma tumor removal surgery are advised to conduct lifelong surveillance. Postoperative CT scan must be carried out to serve as a reference. Patients who do not have any symptoms are advised to undergo CT scans within the first 5 years after surgery for surveillance. This measure is useful for the early detection of recurrence.
Excision with extension to the healthy tissue is necessary to treat ameloblastoma cases adequately. However, this route causes disability in postoperative patients. The degree of difficulty of reconstruction also increases with increasing expansion of excision.

Ameloblastoma management requires extensive research because of its risk of recurrence. The most common action taken to manage this tumor is wide excision to the healthy tissue. However, to date, whether this action is the most appropriate one has not been confirmed. Surgeons are guided by the principle of complete tumor excision to prevent recurrence without performing improper or excessive surgery. While conservative action must be increased in young patients, the focus in elderly patients must be prevention of complications. In particular, conservative action must be developed in the management of unicystic ameloblastoma. If the tumor does not spread beyond the wall or the wall of the tumor is intact, conservative action with decompression or marsupialization followed by enucleation may be recommended. Excision with extension to the healthy tissue is only recommended for large extensive ameloblastomas involving cortical bone or soft tissue; such ameloblastomas are generally of a multicystic type. While application of Carnoy’s, cryotherapy, or diathermy solutions could help reduce the frequency of ameloblastoma recurrence, such treatments also present several shortcomings requiring further study.

Conclusions

We conclude that tumor excision with 1–1.5 cm extension to the healthy bone is adequate for treating ameloblastomas. However, surgeons must pay more attention to soft-tissue excision, especially in aggressive ancatomous ameloblastoma.

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Declaration of Interest

No potential conflict of interest relevant to this article was reported.

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