Treatment of relapse ameloblastoma after hemimandibulectomy and mandibular reconstruction

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ABSTRACT

Background. Ameloblastoma is a benign odontogenic tumor with an aggressive biological behaviour, and the surgical treatment frequently results in failure for the post operative recurrence. Purpose. The aim of this study was to evaluate the clinical result of the patient with aggressive recurrent ameloblastoma who underwent two times radical surgery to get recurrence free. Case. We report a patient who was diagnosed with ameloblastoma of the mandible 13 years ago and had undertaken operation hemimandibulectomy and mandibular reconstruction. However, recurrence occured and secondary surgical treatment (particularly radical) offers the best chance to the patient. The recurrence of an ameloblastoma mainly displays the ineffectiveness or perhaps lack of success of the main surgical treatment. Conclusion. The method has to be intense as well as radical in order to steer clear of reccurence. For ameloblastoma an ineffectiveness of initial surgical treatment indicates that more radical strategy led to minimum recurrence rate.

Keywords: Ameloblastoma; recurrence; radical surgical treatment

ABSTRAK

Latar Belakang. Ameloblastoma adalah tumor jinak odontogen dengan sifat biologis yang mampu menjadi agresif, bahkan perawatan dengan pembedahan seringkali mengalami rekurensi setelahnya. Tujuan. Mengevaluasi hasil klinis pada pasien dengan ameloblastoma rekuren yang telah dua kali dilakukan pembedahan secara radikal untuk mengatasi rekurensi. Laporan Kasus. Laporan kasus pasien dengan diagnosa ameloblastoma pada mandibula sejak 13 tahun yang lalu dan telah dilakukan hemimandibulektomi dan rekonstruksi mandibula, tetapi kembali mengalami rekurensi sehingga diperlukan pembedahan secara lebih radikal untuk mengatasi rekurensi. Rekurensi ameloblastoma menunjukkan tidak efektifnya dari terapi pembedahan yang telah dilakukan sebelumnya. Kesimpulan. Terapi pembedahan pada ameloblastoma sebaiknya lebih radikal untuk meminimalisir kemungkinan rekurensi.

Kata Kunci: Ameloblastoma; rekurensi; pembedahan radikal.

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INTRODUCTION

The ameloblastoma is a benign odontogenic tumor of epithelial origin that exhibits a locally aggressive behavior with a high level of recurrence, being believed theoretically to come from dental lamina remains, the enamel organ in development, epithelial cover of odontogenic cysts or from the cells of the basal layer of the oral mucosa.[1] Although it is considered a benign tumor, its clinical behavior may be regarded as lying between benign and malignant. It is characterized by slow but persistent growth, local infiltration into adjacent tissues and recurrences; however, metastases are rare. Diagnosis mainly from tissue biopsy and characteristic finding on plain X-rays does assist in differentiating between types of ameloblastoma.[1]

The surgical treatment frequently results in failure for the postoperative recurrence, especially larger, aggressive lesions require a more radical surgical approach resulting in large jaw defects.[2]The challenges in the management of this tumor are to provide complete excision as recurrence may occur in incomplete removal and also to reconstruct the bony defect in order to give reasonable cosmetic and functional outcome to the patient.

Management of ameloblastoma has been controversial because of the unique biological behavior of this disease as a slow-growing, locally invasive tumor with a high rate of recurrence.[1] Recurrence rates of ameloblastoma are reportedly as high as 15-25% after radical treatment and 75-90% after conservative treatment.[3] The therapeutic challenge is to achieve a complete lesion excision with the least possible morbidity. For this purpose the surgeon is required to assess the location, size and subtype of the ameloblastoma, as well as age of the patient. A number of different treatment strategies have been previously reported including local techniques (curettage, enucleation or marsupialization) or radical treatments (marginal or en-bloc segmental resection with safety margins and reconstruction of bone defect).[4,5] Therefore, wide resection of the jaw in accordance with the treatment of malignant tumors is usually recommended for ameloblastomas. Recent advancements in understanding the biological behaviors of ameloblastoma have led to more rational surgical approaches.

This paper presents a case of a 33 year old woman who was diagnosed ameloblastomain the right body region of mandible since 13 years ago and was operated in 2013, when he was 31year old. The surgical treatment was hemimandibulectomy; reconstruction of the defect was performed with a titanium reconstruction plate. After 2 years, ameloblastoma relapsed in the symphysis region involving the whole region of right mandible.

CASE REPORT

A 33-year-old female patient reported to the Department of Oral and Maxillofacial Surgery in March 2015 with a chief complaint of swelling on the right side of lower jaw. The swelling was hard and had a duration of 13 years. The swelling first operated in 2013, treatment consisted of hemimandibulectomy, the defect was performed with titanium reconstruction plate, surgical margins were free of tumor, twoyears later, thelesion was relapse and gradually progressing in size and was associated with pain and paresthesia for the past 2 months. Clinical examination revealed a diffuse swelling over the left body-ramus region of the mandible, which extended antero-posteriorly from the left corner of mouth to the pre-auricular region. Superiorly, the extent was up to the infra-orbital region and inferiorly to the submandibular region. The overlying skin was normal in color and texture. On palpation, the swelling was mildly tender, smooth, and uniformly bony hard in consistency, with no local rise in temperature, the margins were ill defined (Figure 1).

Intra-oral examination revealed a diffuse swelling of the left lower buccal vestibule extending from the canine region to the retromolar area. Posterior extension was not visualized clinically. A discontinuity of the overlying mucosa was noted with irregular rolled margins posteriorly.There was expansion of buccal and lingual cortex. Expansion had caused the obliteration of buccal vestibule and lingual region (Figure 2a). On intra-oral palpation, the swelling was firm to hard in consistency and mildly tender. A panoramic radiograph revealed a multilocular radiolucency (Figure 2b).

Figure 1. Pre-operation, extra oral clinical appearances.

A B

Figure 2. Pre-operation (A) intra oral condition, (B) panoramic radiography.

The recurrence was confirmed by a second biopsy. A histopathological examination of the specimen showed a well-differentiated neoplastic proliferation. This appeared as strands of peripheral columnar cells in palisading orientation. The fibroblastic tumor-associated stroma was dense with collagen fibers and highly infiltrated by inflammatory mononuclear cells. No histological signs of malignancy were observed, and the diagnosis of follicular ameloblastoma was confirmed (Figure 3).

On the basis of the histopathology report, radical resection was done taking a safe margin of 2 cm (Figure 4).

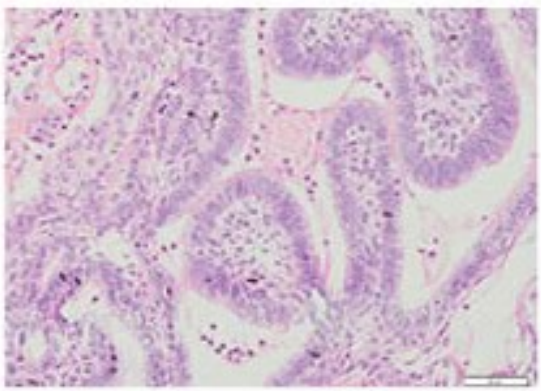
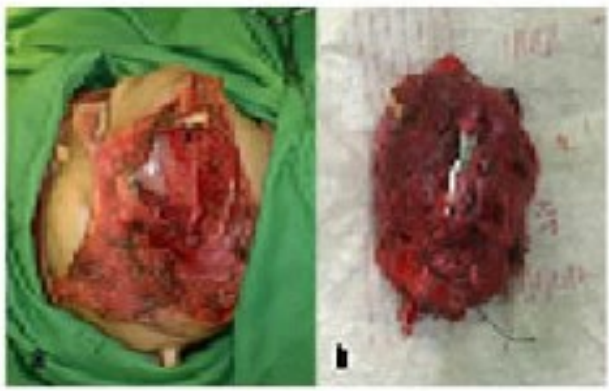


Figure 3. Histopatological finding, follicular ameloblastoma.



A B

Figure 4. A. Radical resection of right mandible, B. mass of tumor.

Figure 5. Clinical pictures,6 months after surgery.

Chemotherapy and radiotherapy was not advised. The patient was kept under regular observation for a period of 6 months. No complications in the postoperative period were reported till date. No recurrence or metastases reported during the follow-up (Figure 5,6).

After 6 months of resection, we've plan to perform the second stage of surgery for reconstruction with nonvascularized fibular bone graft and stabilized with AO reconstruction plate and oral implant rehabilitation in post-oncologic reconstructed mandible. Bone resection due to surgical treatment of large mandibular neoplasm can cause long-span defects. Currently, mandibular fibula free flap graft is widely considered as a reliable technique for restoring this kind of defect. It restores the continuity of removed segment and re-establishes the contour of the lower jaw.[7,8,9,10]

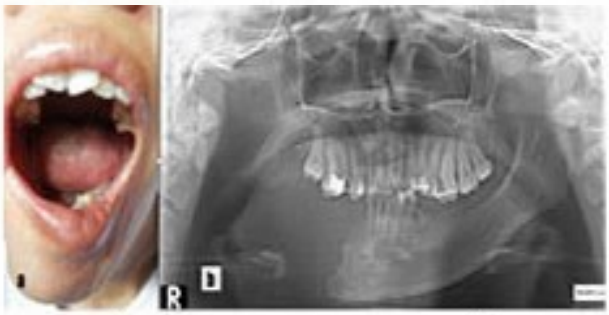


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Figure 6. Post-operative, (a) Intra oral condition,6 months after surgery, (b) panoramic radiography.

DISCUSSION

Ameloblastoma is a tumor with well-known propensity for recurrence. Several factors that may influence the rate of recurrence have been identified. The first and the most important is clinicopathologic variant of tumor. It is generally accepted that there are three variants of the benign ameloblastoma, designated as solid or multicystic, unicystic and peripheral. The solid variety has the greatest propensity for local infiltration and therefore the highest potential for recurrence. The second factor that should be considered is the anatomic site.[7] Up to 95% of ameloblastomas occur in the mandible. The dense cortical bone of the mandible prevents the tumor from spreading extensively for several years, although spread in the central cancellous bone is beyond the radiographic margins of the tumor. The third factor contributing to recurrence is the adequacy of surgery. To ensure that lesion is completely removed, the anatomic extent of the tumor needs to be carefully assessed. The lesions that are completely intraosseous can be adequately assessed with standard radiography. Radiologically, the lesions are expansile, with thinning of cortex in the buccal-lingual plane. The lesions are classically multilocular cystic with a "soap bubble" or "honeycomb" appearance. Finally, the histological variant of the ameloblastoma has been suggested to be of prognostic significance in terms of recurrence. Treatment of ameloblastoma varies from enucleating and curettage to *en bloc* resection.[8,9] The treatment of choice depends on several factors. Multilocularameloblastomas have higher recurrence rates than unilocular ones. Age is another important factor when considering the treatment options. The best treatment is still controversial. Since ameloblastomas infiltrate within the cancellous spaces more, the tumor margin goes beyond the apparent clinical and radiographic margin. The attempts to remove the tumor by curettage may leave small tumor islands in bone, which may later occur as recurrences.[6]

The management of ameloblastoma places the oral and maxillofacial surgeons in a dilemma whether to take a conservative or a radical

treatment. For a long time, surgeons deemed that the ameloblastoma was a benign odontogenic neoplasm, and could be treated by the curettage or enucleation. If the tumor recurred, it could be treated with a secondary surgery in a smaller region than the original lesion.[10] Although it is considered a benign tumor, ameloblastoma has aggressive behaviors including local recurrence, cancerization or even distant metastasis. Therefore, many surgeons tended to take a radical surgery when facing this disease. Our research showed that the prognosis of ameloblastoma was associated mainly with the method of surgical treatment, which meant that patients receiving a conservative treatment had a worse prognosis than those who received a radical one. It seemed that the best way for treating the ameloblastoma was extensive resection of the tumor with a wide region in the normal bone.We believe that the radical surgery is a best choice for many patients of ameloblastoma.

When planning the treatment of ameloblastoma, it is important to understand the growth characteristics and to remove the full extent of the tumor, including the surrounding tissues. Otherwise, the remaining tumor cells may lead to multiple morbidities of recurrence. Wide resection of the jaw is usually the recommended treatment for ameloblastoma, should priority be given to the recurrence rate. However, radical surgery often means that the patients have serious complications including facial deformity, masticatory dysfunction, and abnormal jaw movement. Considering the characteristics of ameloblastoma as a locally invasive but slow-growing and extremely rare metastasizing benign tumor, the priority of the treatment method should be discussed from the points of morbidity and quality of life of the patients, noting that the recurrence rate is not always the primary factor.

CONCLUSION

Ameloblastoma has a high rate of local recurrence if it is not adequately removed. In our opinion, radical surgical resection of ameloblastoma is the treatment of choice. Especially in cases of large, expansive tumors a radical surgical protocol is a very good option to prevent relapse of the tumor on a long-term basis. Successful treatment is the one that renders an acceptable prognosis, causing minimal disfigurement and is based on the behavior and potential of the tumor.

REFERENCES

1. Ram R, Ameloblastoma relapse after 16 years of resection in symphysis of mandible sparing the bone graft, Natl J Maxillofac Surg. 2010 Jul-Dec; 1(2): 190-193.
2. Yi L, Bo H, Long-Jiang L, Prognostic and proliferative evaluation of ameloblastoma based on radiographic boundary, Ijos 2012;4:30-33.
3. Miloro M, Peterson's Oral and Maxillofacial Surgery, 3rd ed. Pmph Publishing, USA,2011.
4. Pedro IC, Treatment of recurrent mandibular ameloblastoma, ExpTher Med. 2013 Aug; 6(2): 579-83.
5. Fregnani ER, da Cruz Perez DE, de Almeida OP, Clinicopathological study and treatment outcomes of 121 cases of ameloblastomas, Int J Oral Maxillofac Surg. 2010 Feb;39(2):145-9.
6. Ramakant D, Atul G, Swati P, Surgical management of ameloblastoma: Conservative or radical approach, Natl J Maxillofac Surg. 2011 Jan-Jun;2(1):22-7.
7. Olaitan AA, Arole G, Adekeye EO, Recurrent ameloblastoma of the jaws: A follow up study, Int J Oral Maxillofac Surg. 1998 Dec; 27(6):56-60.
8. Bataineh AB, Effect of preservation of the inferior and posterior borders on recurrence of ameloblastomas of the mandible, Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 2000 Aug; 90(2):55-63.
9. Montoro JR, Tavares MG, Melo DH, Mandibular ameloblastoma treated by bone resection and immediate reconstruction, Braz J Otorhinolaryngol, 2008 Jan-Feb;74(1):155-157.
10. Sammartino G, Zarelli C, Urciuolo V, Effectiveness of a new decisional algorithm in managing mandibular ameloblastomas: a 10-years experience, Br J Oral Maxillofac Surg, 2007;45(4):306-10.