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COSTOCHONDRAL GRAFT FOR PROSTHODONTIC MANAGEMENT AFTER HEMI-MANDIBULECTOMY DUE TO AMELOBLASTOMA.

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ABSTRACT

Prosthetic rehabilitation plays a vital role in functional recovery after hemi-mandibulectomy patients. We report a case of a 17-year old male with ameloblastoma of his left mandible. Following hemi-mandibulectomy, reconstruction was done using an autogenous costochondral graft harvested from his 7th right rib, decorticated and inserted as an interpositional graft. The patient had a satisfactory clinical outcome with no post-operative complications.

Keywords: Ameloblastoma, Autograft, Bone, Costochondral, Mandible, Young adult.

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COSTOCHONDRAL GRAFT FOR PROSTHODONTIC MANAGEMENT AFTER HEMI-MANDIBULECTOMY DUE TO AMELOBLASTOMA.

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INTRODUCTION

Ameloblastoma is a benign tumor of the bone from the embryonic odontogenous epithelium, with progressive growth that might result in facial disfigurement. Approximately 10-15% of ameloblastoma occurs amongst young adults.¹⁻³ A number of different histopathological varietes of ameloblastoma has been reported i.e. follicular, plexiform, acantomatous, granulose cell, basal cell and desmoplastic; solid or multicystic, unicystic or peripheral types were found macroscopically.^{1,2,4} Due to its high recurrence, radical resection of the affected area guided by intra-operative biopsy is mandatory. Prosthetic rehabilitation is then

an important key feature in the management of hemi-mandibulectomy for recovery of function and aesthetics.⁵⁻⁷ We report a case of hemi-mandibulectomy in a young male patient diagnosed with ameloblastoma in his left mandible, followed by reconstruction of defect using a costochondral autograft.

CASE REPORT

A 17-year old male presented with a painless swelling of the left mandible of 18 months duration, which had gradually increased in size. The patient also experienced a gradual sensory deficit around his left mandible. He could not chew well, as several of his left mandibular molars felt unstable. He had facial asymmetry with a surgical scar at his left mandible due to previous surgical removal of a cyst and two teeth. His vital signs and

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complete blood count (CBC) test were normal. His left mandibular tooth of 37-38 were missing; hyperemia of the mucosa of buccal and vestibulum oris were noted in the area of his tooth 32-43 with palpable intra-oral tumor of approximately 6 x 9 x 3 cm in size. An orthopantomogram (OPG) and CT scan examinations showed unilocular-radiolucent area in the left side of his mandible extending from the tooth 32-36, radiolucent spots were also observed from tooth 32-43 area (Figure 1). He was diagnosed with follicular and plexiform type of ameloblastoma of the left mandible based on the histopathology examination on biopsy.

The patient was informed and consented for a left hemi-mandibulectomy with a planned reconstruction using autologous costochondral autograft from his right 7th rib. Under general anesthesia, left hemi-mandibulectomy from the area of 32-43 teeth was performed and intra-operative biopsy was done to ensure complete removal of tumor cells. Elimination of the tumor cells was meticulously performed by scraping the borders of the affected mandible.

Risdon approach was used with a horizontal incision approximately ½ inch inferior to the lower mandibular border. The initial incision was carried through the skin and subcutaneous tissues to the level of the

platysma muscle; then the skin was undermined with careful scissors dissection. The superior portion of the incision was undermined approximately 1 cm and the inferior portion was undermined approximately 2 cm. The pterygomasseteric muscular sling and submasseteric space were then dissected. Incision of intraoral mucosa was done, vertical resection of the mentum in approximately at the area of tooth 32 was conducted using *gigli saw*. The mandible was then released from depressor labii inferior, depressor anguli oris and platysma muscles. Massive bleeding was avoided by keeping the condyle and coronoid process *in situ* when doing the disarticulation of temporomandibular joint. Approximately 20 cm of the 7th costochondral autograft was harvested, decorticated and reshaped accordingly; with the cartilage end of the graft being placed at the mandibular fossa whilst the other end was secured to the native contralateral mandible using 2 titanium mini-plates 2.0 anchored to the bone using 8 titanium screws (Osteonic, Republic of Korea). To shape the angle and preventing occlusion of the upper jaw, the graft was made at an angle and slightly broken off. Layer-by-layer suture was done and 2 radon drains were inserted.

The patient was kept nil per oral during 7 days post-surgery; intermaxillary fixation was maintained for 4 weeks



Figure 1: An orthopantomogram (OPG) view of the patient's dentition and mandible, with a left unilocular cystic lesion. Tumor is seen in the area of tooth 32-36 (White arrow).

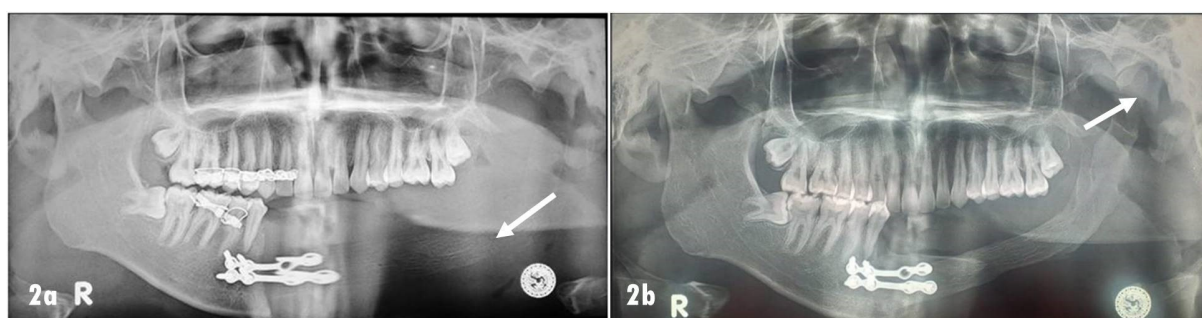


Figure 2: (a) OPG at 16 days post surgery, indicating a successful grafting as evident by the radiopaqueness of the grafted area (white arrow), and (b) at 8 months post surgery, showing good occlusion and no sign of ankylosis of the grafted chondrocartilage left mandibular reconstruction (white arrow). (Click on image to enlarge)

afterwards followed by early mobilization. The autograft showed signs of incorporation as evidenced by the radiographic bridges appearing on the OPG (Figure 2a). Supportive adjunctive physiotherapy to prevent adhesions and soft-tissue contraction in the healing stage was done and 8 months later, the patient had no significant complications, although slight facial deformity from prior operation was still observed. The pareses at the left mandible resolved and he could move his jaw properly. The OPG showed an excellent growth of the graft with no signs of temporomandibular joint ankylosis (Figure 2b).

DISCUSSION

Prosthetic management of a surgically placed decorticated costochondral autograft has been accepted as a sound technique in treating mandible osseous tissue loss.⁵ Other viable options include autogenous fibular flap or iliac crest graft.⁸

A segmental unilocular tumor resection with radical approach to remove the ameloblastoma carefully was followed by left hemi-mandibulectomy, where the gap was reconstructed with a non-vascularized costochondral autograft. Without surgical reconstruction, severe deviation of the mandible to the opposite side would have been likely due to the unopposed contralateral masticatory muscles and the

scar tissue contraction. Recovery of functions like chewing, talking and swallowing is known to be slower with greater disposition for facial deformity.⁴⁻⁶

Costochondral graft has high biological tissue compatibility, good growth potential in juveniles and sufficient anatomic similarities to the condyle although wound healing of the donor area should be carefully taken care of to avoid pneumothorax and / or hemothorax. The growth of the graft should also be monitored continuously; linear overgrowth with malocclusion had been reported.⁶⁻⁸

In recent years, targeted therapy with a clear understanding the pathophysiology at molecular levels is being used for a non-surgical treatment of ameloblastoma. It was reported that in an ameloblastoma, many pathways might lead to increased bone resorption secreted by the tumour cells i.e. RANKL (receptor activator nuclear factor kappa B ligand) that stimulates osteoclastogenesis, the MMPs (Matrix Metallo Proteinase) family including MMP-1, MMP-2 and MMP-9 that may modulate the bone resorption significantly via degradation of the extracellular matrix, IL-1 α , IL-6 and TNF- α that would induce tumour growth and invasion with an interaction to the stromal cells releasing the IL-8. On the other hand, there are types of genetic mutation of B-Rapidly Accelerated Fibrosarcoma / BRAF

(predominantly in the mandibular ameloblastoma) and Smoothed / SMO (more in the maxillary ameloblastoma) genes that codes the hedgehog pathway protein, which is suggested to lead into neoplastic cell growth via the modulation of the Sonic Hedgehog (SHH) antiapoptotic pathway.⁹⁻¹¹ Various agents to modify the aberrant cell growth of ameloblastoma i.e. vemurafenib and dabrafenib which are selective inhibitors of BRAF mutation, itraconazole and arsenic trioxide to inhibit the SHH pathway, have been reported.¹¹⁻¹³

CONCLUSION

The use of costochondral graft improves the aesthetic outcome following hemimandibulectomy, while contributing to faster return to function for the patient. However, this also provides a successful platform upon which prosthodontic work could be carried out so that the patient can regain function for an improved Quality of Life in the long-term.

CONFLICTING INTEREST

We declare that we have no conflict of interest.

INFORMED CONSENT

Verbal informed consent (verbal) was obtained from the patient for publication of the images.

REFERENCES

- 1: Acharya S, Joshi A, Tayaar AS, Gopalkrishnan K. [Extreme ameloblastoma of the mandible with hypoproteinemia: a case report and review of clinicopathological features.](#) J Clin Exp Dent. 2011;3:e343-347. [Accessed on 2021 December 9].
- 2: Avon SL, McComb J, Clokie C. [Ameloblastic carcinoma: case report and literature](#) Journal of the Canadian Dental Association 2003;69:573-576. [Accessed on 2021 December 9].
- 3: Gümğüm S, Hosgören B. [Clinical and radiologic behaviour of ameloblastoma in 4 cases.](#) J Can Dent Assoc. 2005;71:481-484. [Accessed on 2021 December 9].
- 4: Kahairi A, Ahmad RL, Islah W, Norra H. [Management of large mandibular ameloblastoma: a case report and literature reviews.](#) Arch Orofac Sci. 2008; 3:52-55. [Accessed on 2021 December 9].
- 5: Mulyawan I, Ismawati ND. Costochondral reconstruction of the mandibular condyle after hemi-mandibulectomy due to ameloblastoma. International Journal of Oral and Maxillofacial Surgery 2012;42:1250.
- 6: Oliveira LR, Matos BH, Dominguet PR, Zorgetto VA, Silva AR. [Ameloblastoma: report of two cases and a brief literature review.](#) J. Odontostomat. 2011;5:293-299. [Accessed on 2021 December 9].
- 7: Siar CH, Nakano K, Chelvanayagam PI, Nagatsuka H, Kawakami T. [An unsuspected ameloblastoma in the subpontic region of the mandible with consideration of pathogenesis from the radiographic course.](#) Eur J Med Res. 2010;15:135-138. [Accessed on 2021 December 9].
- 8: Tyagi BS, Tyagi S. Long-term results of mandibular reconstruction with autogenous bone grafts and oral implants. Journal of Head and Neck Surgery 2017;1:555-560.
- 9: Fuchigami T, Kibe T, Koyama H, Kishida S, Iijima M, Nishizawa Y, et al. Regulation of IL-6 and IL-8 production by reciprocal cell to cell interactions between tumor cells and stromal fibroblasts through IL-1 α in ameloblastoma. Biochem Biophys Res Commun. 2014;45:491-496.
- 1: Fuchigami T, Ono Y, Kishida S, Nakamura N. [Molecular biological findings of ameloblastoma.](#) Japanese Dental Science Review 2021;57:27-32. [Accessed on

2021 December 9].

- 11: Brown NA, Rolland D, McHugh JB, Weigelin HC, Zhao L, Lim MS, et al. [Activating FGFR2-RAS-BRAF mutations in ameloblastoma.](#) *Clin Cancer Res.* 2014;20:5517-5526. [Accessed on 2021 December 9].
- 12: Kaye FJ, Ivey AM, Drane WE, Mendenhall WM, Allan RW. [Clinical and radiographic response with combined BRAF-targeted therapy in stage-4 ameloblastoma.](#) *J Natl Cancer Inst.* 2015;107:378. [Accessed on 2021 December 9].
- 13: Kim J, Aftab BT, Tang JY, Kim D, Lee AH, Rezaee M, et al. [Itraconazole and arsenic trioxide inhibit hedgehog pathway activation and tumor growth associated with acquired resistance to smoothed antagonis.](#) *Cancer Cell* 2013;23:23-34. [Accessed on 2021 December 9].

