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Case Report

Spontaneous Mandibular Regeneration Following Segmental Mandibulectomy: A Case Report

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ABSTRACT

An 8-year-old male presented with profuse oral cavity bleeding, leading to diagnosis of a large arteriovenous malformation involving the hemimandible and extending into the infratemporal fossa. After optimization and arterial embolization, he underwent hemi-mandibular resection, with spontaneous mandibular regeneration observed at 6-month follow-up. This case highlights spontaneous mandibular regeneration (SMR), a rare phenomenon primarily reported in children, influenced by periosteum preservation. Literature underscores age as a key factor, alongside occurrences in medication-related osteonecrosis of the jaw (MRONJ), with implications for bone tissue engineering (BTE) in mandibular reconstruction. SMR offers potential to overcome traditional reconstructive limitations, warranting further research in oral and maxillofacial surgery.

Keywords: Arteriovenous Malformation, Bone Tissue Engineering, CT Angiography, Hemimandibulectomy.

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INTRODUCTION

Mandibular defects, often resulting from traumatic injuries, benign or malignant lesions, or congenital conditions, pose significant challenges in oral and maxillofacial surgery. Traditional reconstructive options, such as bone grafts or implants, carry risks of donor site morbidity, complications, and variable success rates. Spontaneous mandibular regeneration (SMR) is a rare phenomenon, which the mandible regenerates following resection of a segmental defect. Although documented in several case reports, SMR remains inconsistently observed, with limited understanding of its underlying mechanisms and clinical predictability.

Literature review has reported varying rates of SMR, where a few cases have been documented in South Asia, US, UK, and Japan. Studies have highlighted a gap in awareness and research regarding this phenomenon. Periosteal preservation has been emphasized as a contributing factor. But notably, there is a lack of large-scale studies to fully delineate the contributing factors, such as age, periosteal integrity, and systemic diseases that may influence SMR. Understanding these factors is critical to optimizing reconstructive approaches and improving patient outcomes.

CASE REPORT

An 8-year-old male patient presented to the department of oral and maxillofacial surgery with sporadic bleeding from the oral cavity for the past one year. On intraoral examination, his right lower 1st and 2nd molar deciduous teeth exhibited Grade-III mobility, and there was continuous ooze from the interdental gingiva. Extra oral examination showed an enlarged facial artery with strong pulsation. The intraoral bleeding was controlled using local pressure, and the patient was admitted for optimization, and baseline blood investigations were performed. His Hb level was 5mg/dl. An OPG was performed, which showed a large radiolucency involving right body and ramus of the mandible (figure-1).

Figure-1: OPG of the patient showing radiolucency in the right body and ramus of the mandible

CT Angiography and CT scan were done, and a diagnosis of arteriovenous malformation involving the right mandible was made. The interventional radiologist performed embolization of the feeding vessels (maxillary and facial artery). 24 hours later,

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segmental resection of the right mandibular body and ramus was done, sparing a condylar stump. The periosteum was preserved, and adequate soft tissue was adapted for the closure of intraoral defect. Patient was discharged two days postoperatively and placed on regular follow-up (figure-2). At 6-month follow-up, spontaneous regeneration of mandibular bone was noted in place of resected mandible on OPG (figure-3).

Figure-2: Immediate Post-Operative OPG Showing Segmental Defect From Body of Mandible To The Condylar Stump

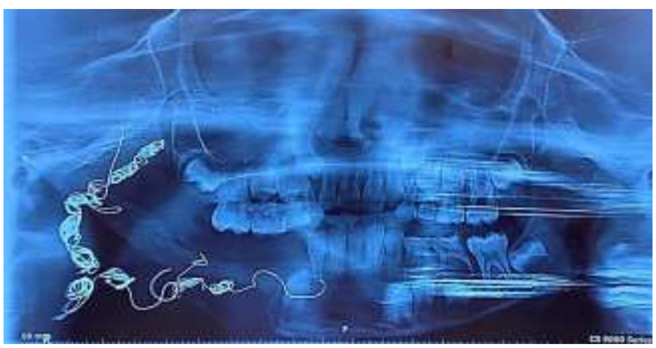

Figure-3: OPG Done After 6 Months Showing Spontaneous Mandibular Regeneration

DISCUSSION

Spontaneous mandibular regeneration was first reported in 1948 by V. H. Kazanjian.[1] This seminal case described bone regeneration following excision of a mandibular section, establishing the phenomenon's recognition in medical literature. Subsequent reports have built on this, often citing Kazanjian's work as the earliest documentation, particularly in pediatric or young adult cases with preserved periosteum.

In our case, an 8-year-old male presented with a large arteriovenous malformation involving the hemimandible and extending into the infratemporal fossa, for which hemimandibulectomy was performed without immediate reconstruction. Notably, at the 6-month postoperative review, spontaneous new bone formation was observed within the resected mandibular region. This finding aligns with a phenomenon that has been only sparsely documented in the literature. Previous reports describe similar regenerative responses following mandibular resections for benign and malignant pathologies, as well as in traumatic scenarios such as gunshot or blast injuries. Our case adds to this limited body of evidence and further highlights the potential for spontaneous mandibular regeneration in select pediatric patients.[2]

SMR is an unexpected, usually rapid formation of cortical bone in a mandibular segmental defect. The size of the neo-generated bone can be a few centimeters to the whole hemimandible.[2] Rai *et al.*, reported 2 cases of spontaneous bone regeneration. One case was reported in a 17-year-old adolescent where resection was done for Ameloblastic Fibroma, and another one in the 7-month-old infant where resection was done for melanotic neuroectodermal tumor of infancy. They compared the outcome of these two cases with 60 other cases of spontaneous regeneration of bone reported till then. Their study concluded that the predictability and consistency of this phenomenon is questionable, and it cannot be taken as a viable option for reconstruction.[2]

Okoturo *et al.*, presented a case series of 8 patients where primarily benign pathologies were removed through mandibular resection while preserving the periosteum. 5 of these cases were of ameloblastoma, 2 of central giant cell lesion, and 1 of fibro myxoma. Most of these resections resulted in large defects, 6 of which were hemi-mandibulectomies. All patients were kept in intermaxillary fixation for 3 to 5 weeks, with in only 2 cases plates were used to stabilize the fragments. Complete bone regeneration was shown in all but 2 cases. They, however, have not mentioned any case of recurrence, particularly in cases of ameloblastoma, where mandibular resections were performed via subperiosteal dissection.[3]

Anyanechi *et al.*, retrospectively analyzed 636 patients who had undergone mandibular resection for varying pathologies over a period of 23 years. A total of 13(2%) patients showed spontaneous bone regeneration. Age range varied from 16 to 51. Periosteum was completely excised in 8(61.5%) patients while partially preserved in 5(38.5%). It took about 9 to 17 weeks to first notice spontaneous bone regeneration. Radiographically, new bone showed same appearance as cortical bone, but it lacked height. They concluded that those patients who show spontaneous bone regeneration may still need bone graft reconstruction.[4]

The phenomenon of SBR is even reported in medication-related osteonecrosis of the jaws (MRONJ)[5,6] Kwon *et al.*, reported 14 cases of MRONJ where mandibular resection was performed. All patients showed formation of a bony bridge, with 71.4% within the first year of resection. They concluded that preservation of periosteum may be one of the most important contributing factors for new bone formation.[7]

A study published by Li *et al.*, demonstrated the role of periosteum in the spontaneous mandibular regeneration. This study determined the dynamic gene expression profile of periosteal cells during bone regeneration in mandibular segmental defects over two weeks in mini-pigs. At week 1, genes associated with immature wound healing processes were up-regulated, indicating the early stages of tissue repair. By week 2, genes involved in skeletal development, ossification, bone mineralization, angiogenesis, and neurogenesis were also expressed, suggesting bone formation and maturation, as well as vascularization and nerve growth in the supporting tissue regeneration.[9]

The findings of this study highlighted the temporal regulation of gene expression during bone healing and identified potential candidate genes that play crucial roles in early stages of intramembranous bone regeneration. This understanding could pave the way for targeted therapeutic interventions to enhance bone regeneration in clinical settings.[10]

CONCLUSION

This case underscores the remarkable regenerative potential of the pediatric mandible when periosteal integrity is maintained. Spontaneous mandibular regeneration offers a significant clinical advantage by potentially reducing the need for complex reconstructive procedures. Further research is warranted to better understand the biological mechanisms behind SMR and to explore its application in mandibular reconstruction, including advances in Bone Tissue Engineering.

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Authors' Contribution

Following authors have made substantial contributions to the manuscript as under:

SJ & ZD: Data acquisition, data analysis, critical review, approval of the final version to be published.

MI: Study design, data interpretation, drafting the manuscript, critical review, approval of the final version to be published.

Authors agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.

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