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CASE REPORT

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Distraction osteogenesis application in bone defect caused by osteomyelitis following mandibular fracture surgery: a case report and literature review

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Abstract

Background Osteomyelitis secondary to mandibular fracture surgery is rare and complete surgical debridement of necrotic infected tissues is an optimal treatment for it. Subsequent reconstruction is required for bone defect caused by operation. Autogenous, allograft and synthetic bone graft substitutes have become widespread in bone defect treatment. Distraction osteogenesis (DO) was also applied in bone defect reconstruction, even it wasn't conventional therapy in jaw.

Case presentation Here we report a case of a 40-year-old aged man who presented with chronic swelling and pain on the right mandibular masseteric region after mandibular angle and Le Fort II fracture surgery. In six weeks after surgery, CBCT images showed that the fracture ends hadn't heal and the fracture gap had widened significantly. The clinical diagnosis of the patient was right mandibular angle osteomyelitis. After controlling the symptoms of pain and infection with local rinses and systemic antibiotic therapy, the patient underwent segmental resection of the infected bone and DO reconstruction for bone defect simultaneously. Encouragingly, well bone healing and normal occlusion restoration was observed finally.

Conclusions DO could be a valuable alternative therapy to bone grafts for bone defect, even in the case of infection.

Keywords Distraction osteogenesis, Mandible, Osteomyelitis, Fracture

Introduction

Osteomyelitis in mandible secondary to open reduction and internal rigid fixation (ORIF) is uncommon in clinical. Osteomyelitis is more prevalent in the mandible than in the maxilla due to the relatively poor vascular supply to the mandible than that of the maxilla. Segmental osteotomy can completely remove infective bone with low recurrence rate and short treatment process, especially in mandible osteomyelitis. Traditionally, mandibular bone defects following segmental osteotomy could be reconstructed by free or non-vascularized osseous flaps [1]. Distraction osteogenesis (DO) is an endogenous

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Fig. 1 The right mandibular angle fracture and bilateral Le Fort II fracture with the third molar just lined on the fracture line

Fig. 2 ORIF with the third molar extraction was implemented

bone regeneration technique usually applied in maxillofacial deformities treatment. Its application in mandible reconstruction was rarely reported in literatures. Here we reported a rare case of mandibular osteomyelitis secondary to angle fracture, which was cured by a surgery with segmental osteotomy and DO at the same time.

Case report

General history

A 40-year-old, well-nourished man presented to the department of oral and maxillofacial surgery, stomatology hospital of Guangxi Medical University with chief complaints of chronic swelling and pain on the right mandibular masseteric region. The patient reported that he was stroked by a motor car, leading to right mandibular angle and bilateral Le Fort II fracture about 2 months ago (Fig. 1). He was immediately sent to hospital and received ORIF and third molar extraction as the tooth was in the fracture line (Fig. 2). A week after the surgery, the patient was discharged from the hospital. One week after discharge, dull pain was presented on the right masseteric region and gradually aggravated. Few weeks later, swelling and progressively restricted jaw opening occurred. He went back to his surgeon for help and the doctor referred him to be hospitalized. The patient had a history of diabetes that was controlled well by insulin injecting four times daily.

Clinical examination

Extraoral examination revealed swelling in the right masseteric region, accompanying severe pain during slight palpation and limited jaw opening. On intraoral examination, a small mucosal fistula distal to the second molar was noted, which would drain pus in masseteric region palpation. CBCT scan revealed that the fracture ends hadn't heal and the fracture gap had widened significantly (Fig. 3).

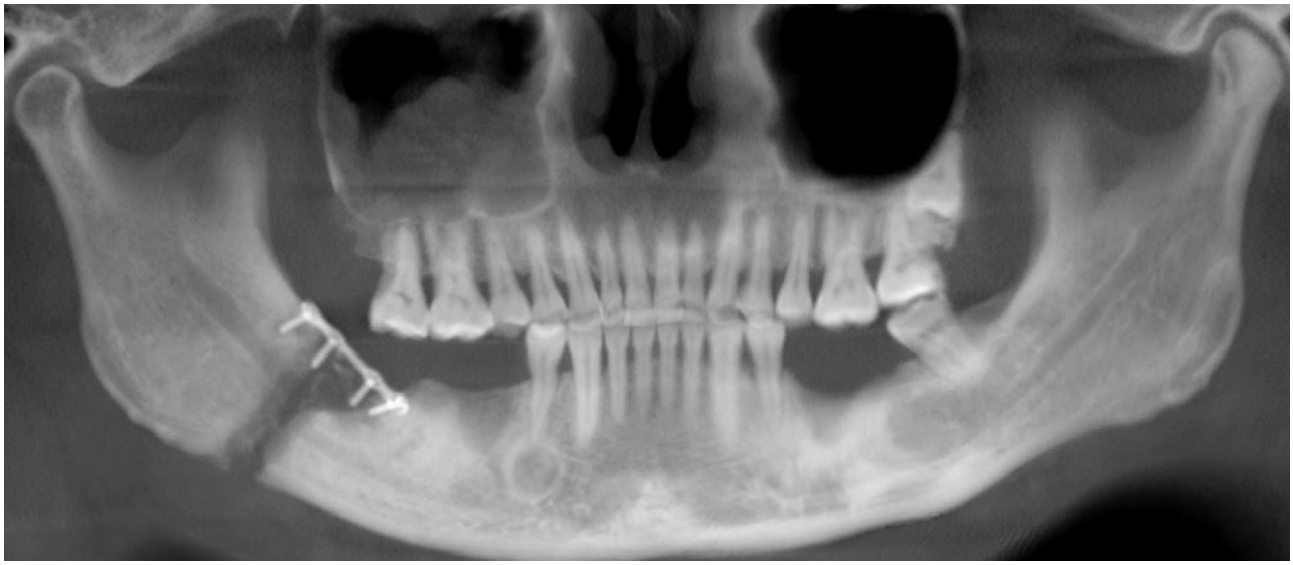

Fig. 3 Mandibular osteomyelitis occurred in the right mandibular angle region

Diagnose and treatment planning

According to the above evidence, the diagnosis of the patient was right mandibular angle osteomyelitis. After sufficient communication with the patient, we planned to use segmental osteotomy and DO in one-stage to settle his osteomyelitis and bone defect once and for all.

Local processing

Before surgery, the intraoral lesion was washed through the mucosal fistula twice a day with 2% chlorhexidine.

Systematic supportive treatments

Ceftazidime and ornidazole was given intravenously to control infection. Dexamethasone 10 mg for 3 days was used to relieve swelling and pain. Pus and infective bone were sent for microbiological culture and sensitive test. Based on the test result, the clinical pharmacist suggested that the ceftazidime and ornidazole was sensitive to the infection of this patient. Five days later, the symptoms of swelling and pus disappeared and the patient was ready for surgery.

Surgical procedure

Under general anesthesia, the mandible was exposed via an intraoral incision. There were numerous sequestrums and granulation tissues in the lesion of right mandibular angle. The necrotic tissues and surrounding affected bone were taken out completely untill the bleeding bone was exposed. A reciprocating saw was used to flatten the bone surface, facilitating linear alignment. Then, an intraosseous distractor device was fixed by screws. CBCT examination was taken the day after operation (Fig. 4).

After 5 days of patency, the distraction device was activated twice daily, at the rate of 1 mm per day until the occlusion was restored to normal and the midline of maxillae and mandible aligned. At the end of distraction, the extended length was 16 mm which was also confirmed by CBCT (Fig. 5).

Consolidation, remodeling, and distractor removal: After six months consolidation, the patient was re-admitted to the hospital. Structural integrated and high-quality new bone in the surgical region was observed on CBCT images (Fig. 6). Under general anesthesia, the distraction device was removed (Fig. 7).

Discussion

In this paper, we reported an uncommon but valuable case of mandible angle osteomyelitis secondary to mandible angle fracture ORIF surgery. Segmental osteotomy and DO was implemented simultaneously to remove the infected tissues and repair the bone defect, which finally harvested high-quality new bone formation and normal occlusion.

Mandibular osteomyelitis is an inflammation involving not only bone marrow but also the cortical and cancellous bone as well as the periosteum. The blood supply of mandible is mainly provided by the inferior alveolar artery and peripheral soft tissues, which is poor than that of maxilla. Therefore, mandibular osteomyelitis is more commonly than the maxilla osteomyelitis. In older people, the infection in mandible would exacerbate because of the age-relative atherosclerosis of the artery [2].

Osteomyelitis of the jaws can be categorized into three types: acute osteomyelitis, primary chronic osteomyelitis and secondary chronic osteomyelitis [3]. According to this classification, the case reported in this article could be defined as primary chronic osteomyelitis as a more than 4 weeks' course, pus exudation, sequestrum

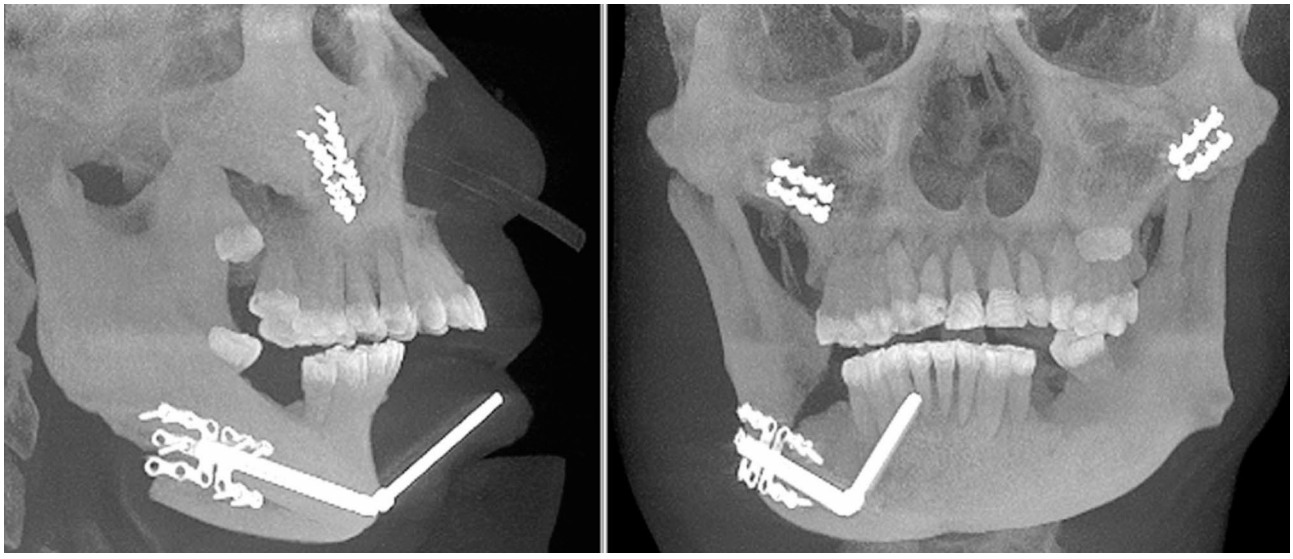

Fig. 4 Segmental resection and DO was completed. The anterior region of the CBCT panoramic graph by can't be reconstructed due to deep overjet

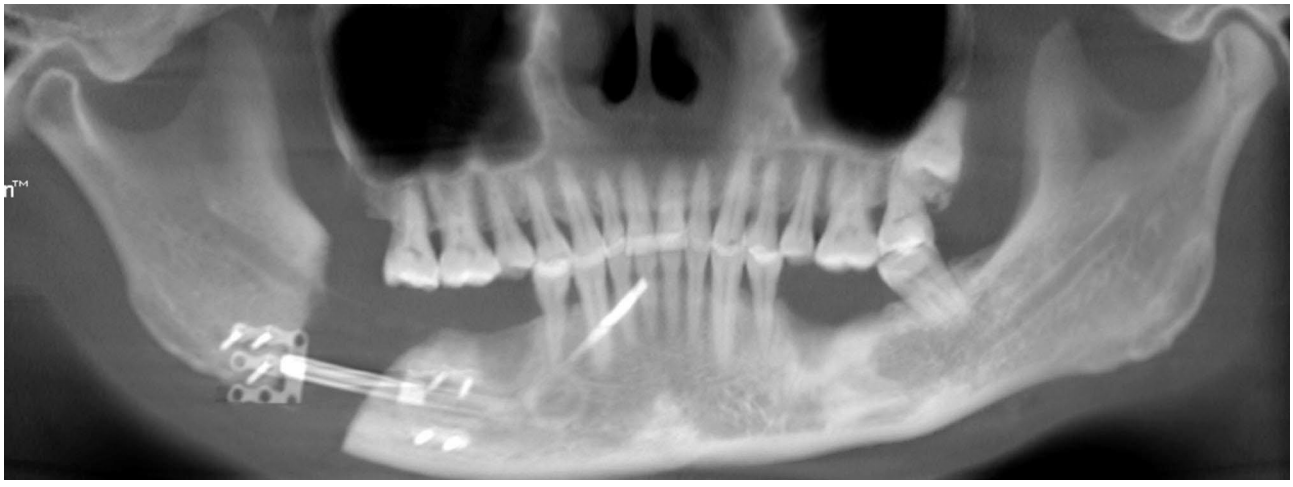

Fig. 5 The distraction length was 16 mm and confirmed by CBCT

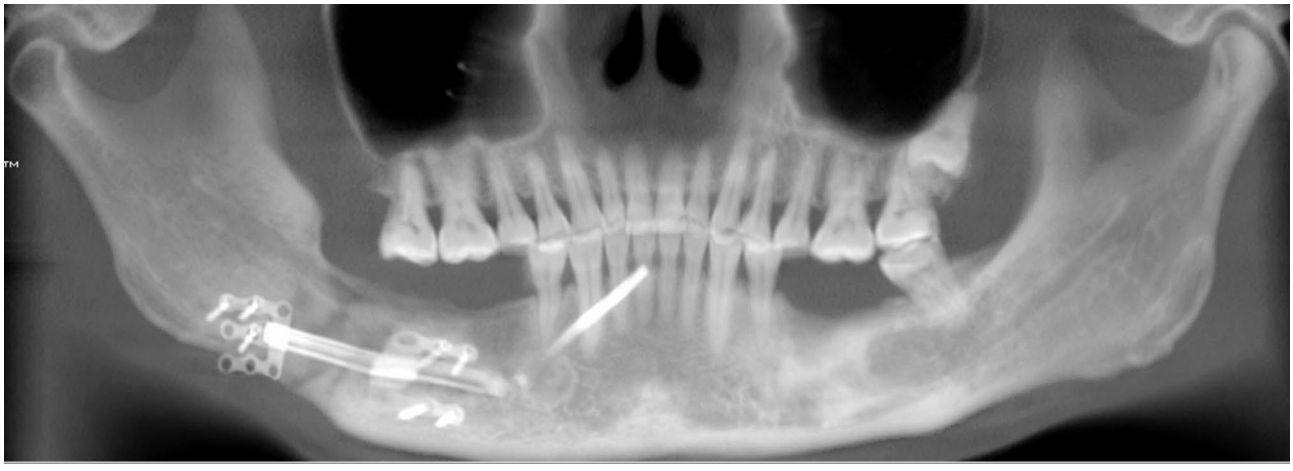

Fig. 6 After 6 months consolidation, new bone in the distraction gap was observed

Fig. 7 The distraction device was removed

formation and fracture origin. Unappropriated treatment of extraction sockets and pathological fractures were the majority causes of mandibular osteomyelitis [4]. The combination of these two factors resulted in mandibular osteomyelitis in the case presented in this article. Systematic supportive treatments including nutrition, intravenous antibiotics and steroids, hyperbaric oxygen, were vital to relieve the symptoms of fever, swelling, pain and restricted opening in osteomyelitis patients once they were admitted [5], these therapies couldn't completely prevent osteomyelitis progression or recurrence. Microbial culture and drug sensitivity test of the infective bone and marrow should be carried out as early as possible and the antibiotics should be changed according to the test result and the suggestion of pharmacist.

Surgical interventions are unavoidable once sequestrum is observed in osteomyelitis by clinical and radiographic examination, as the use of antibiotics alone has no effect on the necrotic tissues [6]. Debridement, decortication and segmental osteotomy or combination therapy can be chosen for osteomyelitis according to clinical disease features [7]. Debridement, which remove infective tissues thoroughly, is effective in mandibular osteomyelitis treatment. Unfortunately, its success rate is not high and may cause a long disease course or progress to certain refractory cases [5]. Mandible decortication would be more effective when it was combined with debridement. Segmental osteotomy can be applied for refractory cases with totally destructed mandible, as it can remove the infective bone thoroughly and bring about low recurrence rate [8]. The recurrence rate of mandibular osteomyelitis was 50% when it was treated by marginal resection, while it was reduced to 5.5% by segmental osteotomy [9]. Segmental osteotomy shows excellent clinical effects on mandible osteomyelitis. It has an obvious disadvantage that mandible defects should be managed simultaneously or secondarily. Reconstruction of bone defects that secondary to osteomyelitis is simpler than that caused by malignancy, as the length is short and soft tissue is intact. Therefore, it is preferable that segmental osteotomy and mandibular reconstruction is implemented simultaneously by using vascularized or non-vascularized bone grafts [8]. In this case, we provided another choice, utilizing the distraction osteogenesis (DO) technique instead of bone graft.

DO is well-known as an endogenous bone regeneration process in which distraction bone and surrounding soft tissue can be lengthened simultaneously with high-quality new bone formation within the distraction gap. It was applied in massive clinical and reached fairly good clinical results by Ilizarov and Soibel et al. in 1969 [10]. Traditionally, bone graft or osseous free flap would be used to reconstruct the mandibular bone defects caused by pathological mandibular fracture [11] or mandibular fracture nonunion [12]. However, these treatments have risks of pain, structural damage, dysfunction and even infection in donor site, especially in the diabetic patient just like the case in this report. Compared with bone grafts, DO can regenerate new bone tissue with normal histological structure and similar mechanic properties for critical-size bone defects [13] and reduce the side effects of bone grafts [14]. Mandibular DO can be divided into 4 categories: body DO, ramus DO, condylar DO, and symphysial DO [15]. It was reported that the applications of trifocal DO on mandibular body defects which caused by excision of ameloblastoma or keratocystic odontogenic tumor could gain satisfactory new bone formation and dental rehabilitation [13]. Lengthening the mandible by 6.5 cm, DO have also successfully treated mandibular body defects secondary to oncologic head and neck

surgery [16]. Transport distraction technique combined with the help of 3D model was used to reconstruct and functionalize the condyle in a bilateral temporomandibular joint ankylosis patient [17]. Distraction of mandibular was easy and simple to handle and it was amazing that the new forming bone had the same growth potential to normal infant bone [18]. From the above content, it can be concluded that DO was a preferable alternative for bone defects in mandible.

The application of DO to reconstruct mandibular defects exhibits excellent advantages in forming good quality continuous new bone. However, complications such as regression, fibrous union, infection and device failure would occur in some cases [19]. The occurrence of regression or relapse depends on the length of distraction in mandibular DO, which is unavoidable in some cases [20]. Mild infections are more prevalent than severe infections within four weeks post-surgery, with a cumulative incidence of 13.6%. These infections can be cured through local washing with sterile saline. Perioperative red-blue irradiation therapy can significantly reduce the infection rate [21]. Fibrous union, usually results from lack of latency period and excessively frequent distraction rhythm, which needs second DO surgery to remove the fibrous tissue in distraction gap [22]. The device failure is characterized by the fracture of distractor, and the treatment approach is determined by the stage of DO. The incidence of device failure range differently in applied modalities (intraorally or extraorally) and instrument material [23]. DO in mandible might cause a range of complications, both mild and severe, which can be prevented by careful planning and surgery.

The reconstruction strategy for mandibular defects has gradually changed with the development of free bone flaps, yielding positive clinical outcomes [24]. Vascularized autograft was considered as a golden criterion for mandible reconstruction and free fibula flaps were most common chosen in clinical. Mandibular reconstruction needs both restoration of occlusal function and esthetic facial. Steven and his colleagues reported that mandibular defects could be repaired by free fibula flaps with the help of virtual surgical planning and surgical cutting guide, which achieved excellent surgical precision and favorable clinical outcomes [25]. Bone flaps such as iliac crest, scapula and rib are also used for mandible vascularized reconstructing while the metatarsus, medial femoral condyle, distal radius are seldomly considered [26]. These bone free flaps can be easily shaped to duplicate the mandibular contour and provide stable supports for dental rehabilitation to obtain functional and cosmetic results. The uncertain success rate of bone flaps and side effects of donor site such as hemorrhage, infection, paresthesia, persistent pain, esthetic problems still limit their clinical application especially in skeletal defects and pediatric population [27, 28].

Conclusion

Segmental osteotomy is an effective therapy for mandibular osteomyelitis with low recurrence and short treatment course. DO can reconstruct bone defects after osteotomy, obtaining high-quality regenerated bone and optimal occlusion without the need for a donor site. DO is a continuously development surgical technique and has significant vantages over traditional bone flaps in reconstruction fields.

Author contributions

Xuanping Huang and Qingtiao Xie completed the entire treatment. Qingtiao Xie wrote the first draft of this manuscript. Xianfang Jiang and Xuanping Huang revised the manuscript. All authors read and approved the final manuscript.

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Data availability

No datasets were generated or analysed during the current study.

Declarations

Ethics approval and consent to participate

This study was approved by the ethical standards of the Affiliated Stomatology Hospital of GuangXi Medical University. Written informed consent was obtained from the patient for the publication of this case report.

Consent for publication

The patient in this article has signed a consent form for the agreement to use his photographs.

Competing interests

The authors declare no competing interests.

Clinical trial number

Not applicable.

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