



Spontaneous Healing of Mandibular Fracture after Molar Extraction in a Patient with Undiagnosed Osteogenesis Imperfecta: Case Report and Clinical Considerations

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Abstract

Osteogenesis imperfecta (OI) is a group of inherited disorder characterized by low bone mass, bone fragility and bone fracture, which also has several metabolic defects that predispose patient surgical and anesthetic risks. Furthermore, bisphosphonates (BPs) are main part of their therapy and require some surgical considerations. OI patients commonly suffer from long bone fractures due to mechanical loading but facial fractures are very rare in patients with such disorders. Facial fractures in OI patients were generally managed surgically in literature. From the best of our knowledge, we describe a first case of a mandible fracture in a patient with undignosed OI successfully managed by postoperative instructions without intervention.

Keywords: Spontaneous Healing; Mandible Fracture; Osteogenesis Imperfecta

Introduction

Osteogenesis imperfecta (OI) is known as 'brittle bone disease' [1]. OI is a rarely seen genetic heterogeneous disorder that affects collagen metabolism (especially type 1 collagen metabolism). Bone, dentin, ligaments and sclera contain copious amounts of type 1 collagen, so these structures are generally affected [2]. Its' estimated prevalence is 1 in 12000 to 15000 children. Patient with OI is characterized by decreased bone mass, increased bone fragility and increased risk of multiple long bone fractures, but facial fractures are rare [3]. We presented a case of left mandible body fracture in a patient with undiagnosed OI following extraction of left first molar tooth.

Case Report

A 34-year-old man had come to our consultation with severe pain, and paresthesia on his left mandible after the lower left first molar tooth extraction. The patient had history of uncomplicated tooth extraction. Clinical examination shown left mandibular angle swelling, limited range of mouth opening. Radiographic examination showed isolated left mandible body fracture that not causing malocclusion. Fracture pattern was favorable and displaced fractu-

re was not shown in CBCT (Figure 1). Osseous step was not reported upon palpation. The patient had been diagnosed of reflux. He suffered bone fractures with minimal trauma in adolescent (humerus fracture). Humerus fracture was treated surgically. Later, plates had been removed due to infection and had bone deformity on the affected side. There were clear family of history of OI (his uncle and aunt). Physical examination revealed that patient had blue sclera (Figure 2). The patient was consulted to the orthopedic and genetic department for the suspicion of osteogenesis imperfecta. Genetic scan showed mutations in collagen, type 1, alpha 1 (COL1A1) on chromosome 17 and collagen, type 1, alpha 2 (COL1A2) on chromosome 7. These are genes that encode type 1 collagen. Moreover, bone density scan confirmed osteoporosis (Figure 3). The patient was diagnosed with osteogenesis imperfecta (type 1) and placed on zoledronate therapy to prevent further fractures. Protecting mandible from trauma, soft diet, meticulous oral hygiene and deferring from yawning are instructed and patient was followed regularly. A normal sensation of left inferior alveolar nerve and occlusion were reported at the first month. Healing was completely uneventful during 24 months follow up (Figure 4).



Figure 1: CBCT shown left extracted socket and mandible body fracture.



Figure 2: Blue Sclera.

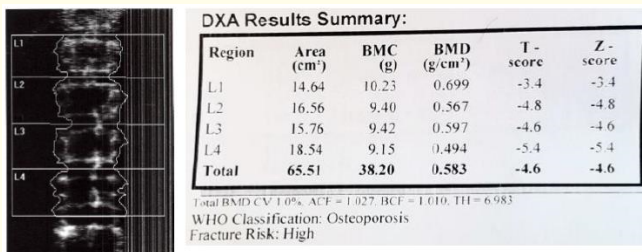


Figure 3: Lumbar Spine Dual Energy X-ray absorptiometry (DXA) revealing bone mineral density reduction.



Figure 4: Uneventful healing (24 months follow-up).

Discussion

OI is a heritable disorder of bone development. OI patients have low bone mass and greatly increased risk of fragility fractures. Sil- lence., *et al.* classified this disorder into four categories and type 1 OI is the most common form of the disease [4].

Patients with OI generally suffer from long-bone fractures, but facial fractures are rare [5]. Fractures of mandible occur at intra- operative or postoperative period and are mostly associated with the removal of wisdom teeth [6].

Management of fracture in OI patient takes into account that they are at risk not only from open reduction and fixation due to fragile bone but also metabolic defects (hyperthermia, platelet dis- orders,). Incidence of hyperthermia was reported high in OI pa- tients receiving general anesthesia. Hyperthermia can lead up to malignant hyperthermia during the general anesthesia [5].

Platelet disorders are another aspect of distorted energy met- habolism and result in crucial bleeding in OI patient. The risk of bleeding related to type of OI. Platelet functions including adhesion and clot formation are also disrupted in OI patients. Thus, history of bleeding episodes should be asked and managed accordingly while managing fractures at OI patients [7].

Another point that should be taken into consideration is diffi- culty of intubation in OI patients. OI patients generally have large tongues and short necks. Anesthesiologist should be careful about fracture by applying force on mandible while intubating OI patients due to generalized osteoporosis. Furthermore, the teeth may be eas- ily torn out due to dentinogenesis imperfecta in OI patients during the intubation [8].

BPs are commonly prescribed for a range of metabolic condi- tions such as hypercalcaemia, osteoporosis. Moreover, BPs are also used in OI patients to increase the bone density, reduce skeletal pain and decrease the incidence of bone fracture. However, BPs therapy results in side effects (osteonecrosis of the jaw). Studies have revealed that osteonecrosis of the jaw was triggered by in-vasive tooth extraction. Invasive surgery involving jaws have been thought as precipitating factor for the development of osteonecrosis of jaw [9].

Clinicians should pay attention while treating mandible fractu- res in OI patients. Both systemic condition and BP use can com- promise bone healing in OI patients. Newly formed bone is inferior quality as the native one in OI patients. Thus, open reduction and fixation can increase the risk of mal-union and non-union due to decreased bone healing capacity in OI patients [10].

Conservative management (intermaxillary fixation) and rigid internal fixation (RIF) are the both treatments of choice which are

used mostly in the treatment of mandibular fractures. RIF achieve three-dimensional stability which provides patient with immediate function. On the other hand, patients' mouths are closed for 3-4 weeks at conservative therapy (intermaxillary fixation) [5]. In our present case, intermaxillary fixation was not the treatment of choice due to patients' airway problems and surgery was not taken into consideration due to anesthetic risks.

Avoidance of mandible fracture should be kept in mind in OI patients when removing tooth. The incidence of lower jaw fracture in OI patients is low. On the other hand, patients with OI was put in increased fracture category when exposed to trauma. Special attention should be given when extracting molar teeth. Sectioning of tooth rather than bone removal is suggested to prevent mandible fracture while removing tooth.

Conclusion

In conclusion, mandible fractures can be minimized by meticulous preoperative assessment of the patient. From clinical point of view, patients with OI suffer not only from fragile bones, but also metabolic conditions. Prevention of fractures is an important factor for OI patients. Fracture of mandible even in OI patients is very low when the surgical principles are fully applied. Conservative therapy should be in surgeons' armamentarium for managing mandible fractures in OI patients.

Conflict of Interest

There is no conflict of interest between authors.

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