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Intramedullary osteosynthesis for fracture associated with osteogenesis imperfecta☆

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ABSTRACT

Our aim was to analyse the complications associated with intramedullary stabilisation of the bone fractures which are a common complication of osteogenesis imperfecta. A total of 12 fractures among six individuals with osteogenesis imperfecta were treated by intramedullary stabilisation. The mean age of the patients (three male and three female) was 19.4 years, range 7–42 years. The most common fracture site was the femoral midshaft (seven fractures). After implant removal, one new fracture and one re-fracture occurred. Operative stabilisation of fractures is a safe treatment option for osteogenesis imperfecta.

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Introduction

Osteogenesis imperfecta (OI) is a genetic disorder characterised by defective type I collagen synthesis leading to frequent fractures, often for little or no apparent reason, and the development of progressive deformity, particularly of the long bones.[6] It has been estimated that 20,000–50,000 people in the USA have the disorder. Definitive diagnosis is based on clinical features, collagen biopsy and DNA tests. The most commonly identified cause is a dominant genetic defect, followed by a few recessive genetic disorders and spontaneous mutations.[19] Usually the characteristic features vary from person to person, even within the same family. Older classifications termed less severe forms 'osteogenesis imperfecta tarda' and more severe forms 'osteogenesis imperfecta congenita'. In 1978, Sillence et al. described a four-type system, which is now extended to an eight-type system (NIAMS), representing different states of the disease.[10,19,22,23]

General treatment consists of drug therapy (bisphosphonates, growth hormones, pain management), physiotherapy, cast therapy, braces and the use of wheelchairs and other mobility aids.[2,16] Fractures are treated either non-surgically or surgically. In stable fractures, non-operative therapy usually based on closed reduction and fixation with casts or splints is preferred.[20] For young children with femoral fractures, Bardenheuer's overhead skeletal traction therapy for 6 weeks is the treatment of choice.

For displaced or unstable fractures, surgical treatment is generally suitable and consists of plate fixation, intramedullary (IM) nail stabilisation or external fixation.[14,20,21] Preventive IM insertion of rigid or flexible rods and telescopic nails (e.g. Bailey nail, extensible Sheffield rod, pins such as the Rush pin) is performed to prevent fractures and to correct deformities.[2,3,13,24] Multiple bone fractures and re-fractures after operative or non-operative treatment are common complications.[11,12]

The standard treatment of fractures of long bones consists mostly of IM stabilisation with various devices, but there are no guidelines for the treatment of acute fractures of people with OI. This study reports the long-term results of surgical treatment methods used for a consecutive series of six individuals with OI over 13 years. Its aim was to analyse bone fracture treatment with IM stabilisation and ensuing complications among people with OI.

Methods

Charts and radiographs of all patients with OI, treated for traumatic fractures at the Department of Trauma Surgery of the Medical University of Vienna between 1993 and 2006, were retrospectively analysed. A consecutive series of 12 people, eight male and four female with a total of 72 fractures, was identified. Within this series a group of six individuals (three male and three female, mean age 19.4 years, age range 7–42 years).) were treated by IM osteosynthesis. A total of 12 bones were affected (Table 1). The mean follow-up after fracture was 78 months (range 17–170 months). Premorbid ability in all cases was good; no one was

☆ Parts of this study were presented as a poster at the Osteology Congress 2007, Vienna, Austria, on 1–3 March 2007.

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Table 1
Long bones treated operatively.

Extremity	Number of bones
Total	12
Femur	7
Radius	2
Ulna	1
Tibia	2

wheelchair dependent. At the last follow-up each person in the series underwent clinical examination by the authors, and plain radiographs were obtained.

We distinguished three types of physical restoration: 1 = excellent or good (able to walk and exercise freely); 2 = fair (walking with mobility aids); and 3 = poor (wheelchair dependence most of the time).

We distinguished between true re-fracture on the same fracture site, a new fracture of the same bone and non-union (Table 2).

Operative technique

A reamed locking nail (diameter 13 mm) was used in two cases, an unreamed locking nail (diameter 9 mm) in one case, a reamed gamma nail (T3) once and various IM rods (diameter 1.4–3.5 mm) as required.

Results

The most common fracture site (29%) was the femoral midshaft. Figs. 1–7 illustrate two typical cases of fracture and re-fracture.

Surgery was required by six people for a total of 12 fractures and was based on IM fixation, with bone augmentation for one fracture (Table 3) and postoperative casting for one person (Table 4). Implant removal was performed for seven fractures; for two people the locking screws were removed and the nail left in place; the re-fracture was detected 10 months after removal of an IM rod; and, in one case, the two Bailey nails (inserted primarily at another hospital) perforated the knee joint and required shortening. Physical restoration was good or very good in most cases, the average score being 1.37.

No early postoperative complications were observed in this series. Our data indicate a clear age distribution of people with fractures, peaking at 19.4 years. In four cases the type of OI was known: one of type 1, two of type 3 and one of type 4.

Discussion

To our knowledge, based on literature search, few studies of the treatment of fractures and associated complications among people with OI exist.[4,7,11] The risks indicated by our data of re-fracture and new fracture of the same bone after operative stabilisation in OI are thoroughly comparable with published reports. Non-operative treatment of femoral and tibial bone in our series was limited to children up to 4 years of age and to older children with undisplaced fractures, and required 6–15 weeks of cast immobilisation.

Table 2
Operative treatment with intramedullary device of 12 fractures: numbers of postoperative new fractures and re-fractures.

Total implants	12
Re-fracture with implant	1
New fracture with implant	0
Re-fracture after removing implant	2
New fracture after removing implant	1

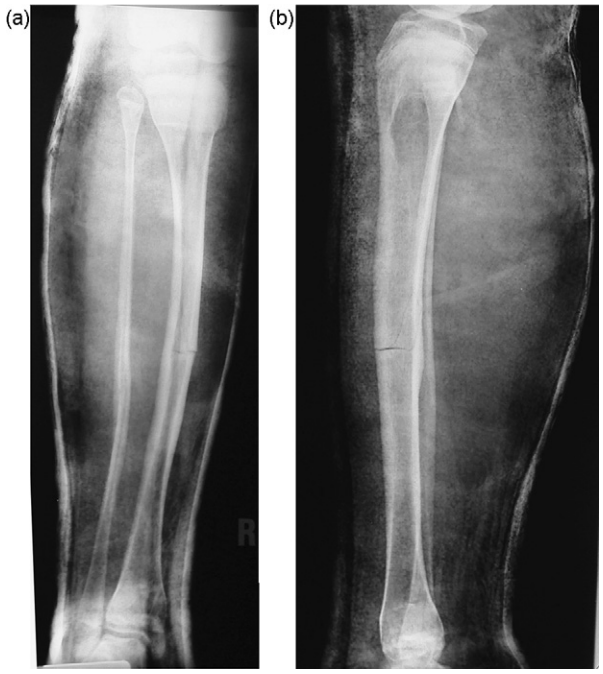

Fig. 1. Case 1 (15-year-old girl). Radiographs: (A) A.p. view. (B) Lateral view. First fracture of right tibia on 16 May 2002, treated by casting.

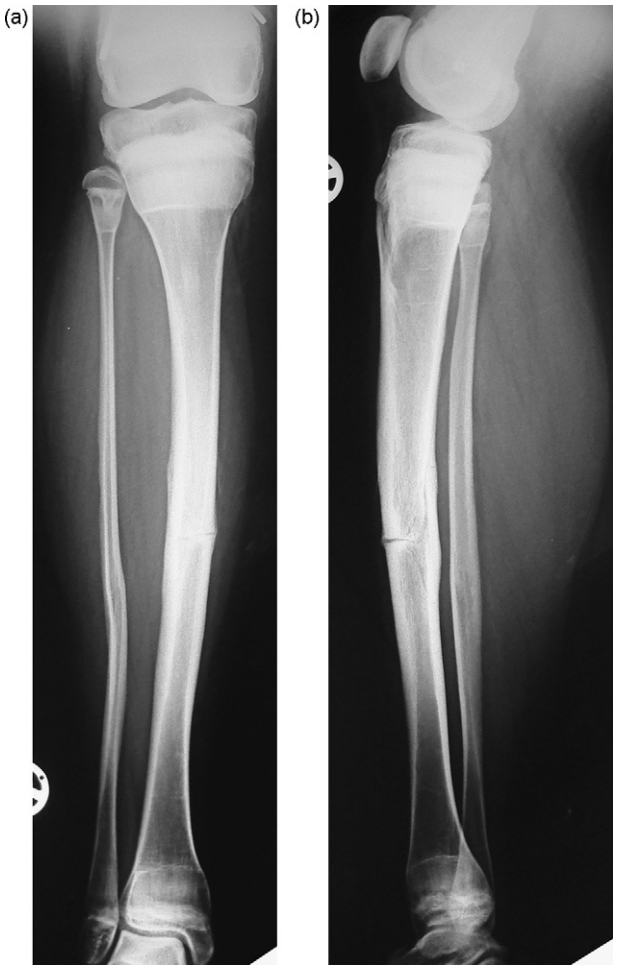

Fig. 2. Case 1. Radiographs: (A) A.p. view. (B) Lateral view. Cast was removed after 15 weeks.

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