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ORIGINAL PAPER

Locking plate and fibular strut-graft augmentation in the reconstruction of unicameral bone cyst of proximal femur in the paediatric population

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Abstract
Purpose Several therapeutic strategies have been used for managing unicameral bone cyst (UBC) of the proximal femur. However, there is insufficient evidence to support one treatment over another, and the optimal treatment is controversial. This study aims at describing our experience with surgical reconstruction of paediatric UBCs of the proximal femur using a proximal locking plate and fibular strut allograft.
Methods In total, 14 consecutive paediatric patients with Dormans types IB (four cases) and IIB (10 cases) UBC were assessed. Mean patient age was 8.6 ± 2.3 years, and mean follow-up period was 41.7 ± 29.8 months. Six patients (42.8%) were referred with a pathologic fracture. Clinical/radiological outcome and complication rates were evaluated at the final follow-up session.
Results No cysts were Capanna's class III (recurrence) or IV (no response). Complete healing (Capanna's class I) was seen in ten cysts, while four other cysts healed with residual radiolucent areas (Capanna's class II). Mean healing period was 14.1 ± 5.1 (9–24 months). One patient had superficial infection, one heterotopic ossification, and one mild coxa vara, and mean Musculoskeletal Tumor Society (MSTS) score was 99.5%.
Conclusion According to our results, locking plate and fibular strut graft in Dormans classification types IB and IIB results in a favorable outcome in managing UBC of the proximal femur in the paediatric population.

Keywords Unicameral bone cyst · Proximal femur · Surgical reconstruction · Locking plate · Fibular strut allograft

Introduction

Unicameral bone cyst (UBC), also known as solitary or simple bone cyst, is a benign lesion that mostly affects children between the age of four and ten years and represents ~ 3% of all primary bone tumors in the first two decades of life. It is defined as an atrophic, degenerative osteolytic process consisting of a cavity filled with fluid and lined by a membrane. The most common anatomical location is the proximal humerus, followed by the proximal femur. Although its aetiology remains unknown, the intramedullary venous obstruction or disturbance theory is one of the most accepted pathogenesis models [1, 2]. The main treatment objectives are to prevent pathologic fracture, promote cyst healing, and avoid cyst recurrence. Surgical intervention has been advocated for large cysts to prevent fractures and subsequent complications, including skeletal deformities, especially in the weight-bearing long bones. Among the wide range of different therapeutic modalities described for treating UBC are radical excision in the form of subperiosteal partial diaphysectomy and allograft, subtotal resection with and without bone graft, curettage and bone graft, multiple drill holes, intracystic prednisolone injection, and, recently, intramedullary flexible nails [3]. However, search for the development of an optimal therapeutic approach continues. Dormans and Pill introduced a classification system to guide the treatment of lytic lesions of the proximal femur. In Dormans classification, type II is in the femoral neck; type I is not. Moreover, subdivision B refers to cysts

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where insufficient bone stock is present on the lateral side of the proximal femur [4]. While proximal femoral UBC reconstruction is challenging [5, 6], managing types IB and IIB with insufficient bone stock in lateral buttress is even more challenging. The aim of this study is to describe our experience with surgical reconstruction for UBC types IB and IIB of the proximal femur using proximal a locking plate and fibular strut allograft.

Materials and methods

Between 2007 and 2014, 85 consecutive paediatric patients with a confirmed diagnosis of UBC were treated at our centre. In 19 patients, the UBC was in the proximal femur, from which 16 with Dormans types IB and IIB were identified and in whom locking plate and fibular strut-graft augmentation was used for UBC reconstruction. Two patients were lost to follow-up. Consequently, 14 patients were included in the final study, which comprised six female and eight male patients with a mean age of 8.64 ± 2.3 years (range 5–12). Mean follow-up was 41.7 ± 29.8 months (range 24–108). Clinical and demographic characteristics are shown in detail in Table 1.

Based on the Dormans classification system, four type IB UBC were identified in which the lateral buttress was affected and the femoral neck was intact (Fig.1a). The remaining ten patients were classified as type IIB, in which both lateral buttress and femoral neck were involved (Fig.1b).

Six of 14 patients (42.8%) presented to our center after sustaining a pathologic fracture. In five patients (35.7%), thigh pain, and limping were the leading cause of referral. The remaining three patients (21.5%) were detected incidentally.

Surgical technique

All patients were operated using the Watson–Jones approach [7] under general anesthesia on a regular operating table employing an image intensifier fluoroscopy. We first exposed the anterior part of the proximal femur. In cases with fracture, we initially reduced the fracture and created an oval-shaped cortical window over the cyst. Subsequently, curettage and high-speed burring of the cyst wall was done, and the removed material was sent for histological examination. In type IB, the fibular strut graft was inserted in the proximal femoral shaft (Fig.2a), while in type IIB, it was placed in the femoral neck (Fig.2b).

After placing the fibular strut grafts, the remaining voids were filled with cancellous allograft chips. The locking plate was fixed to the lateral part of the proximal femur with intensive care to avoid any physical damage. Fresh–frozen fibular shaft and cancellous allograft were provided by our bone bank, which harvests and stores allografts according to the standard tissue-banking protocols of allograft preparation and processing [8]. We used proximal humerus locking plate (AO philos plate 3.5), which better matches the small size of the proximal femur in this pediatric group of patients. We also sometimes bent the plate to direct the locking screw in line with the femoral neck. Post-operative antibiotic prophylaxis using 50 mg/kg cephazolin was administrated for 24 hours. Partial weight bearing and waking with crutches was started three days after the surgery. Progressive weight bearing was planned considering cortical thinning and pathologic fracture. In patients with pathologic fracture, weight bearing was delayed to three weeks, and progressive weight bearing was encouraged considering the time of radiographic observation of the callous.

The first follow-up was two weeks after surgery. In cases without a pathologic fracture, follow-ups were performed at three month intervals thereafter. In cases with a pathologic fracture, follow-ups were in four week intervals until union, and three month intervals thereafter. At each follow-up, improvement was evaluated using serial plain radiography, which was continued until cyst consolidation. Moreover, all post-operative complications were recorded. Capanna's classification system was used for radiological evaluation of healing at the latest follow-up [9]. The classification system divides the healing process into four categories based on response to treatment:

(1) Healed
(2) Healed with residual
(3) Recurrence
(4) No response

Functional outcome was assessed using the Musculoskeletal Tumor Society (MSTS) score at the latest follow-up.

Statistical analysis

Data was analyzed using IBM SPSS for Windows version 16. Central tendency and variability of numeric variables were assessed using mean and standard deviation (SD), respectively. Mann–Whitney *U* test was used to compare variables across groups. Pearson correlation coefficient was used to evaluate potential correlations. *P* value <0.05 was considered significant.

Results

Ten cysts were classified as completely healed (Capanna's class I) (Fig.3); mean healing period (complete defect filling) was 14.1 ± 5.1 months (range 9–24).

Four cysts were classified as healed with residual radiolucent areas (Capanna's class II) (Fig.4), meaning that residual radiolucency was observed radiographically at the last follow-

Table 1 Patient clinical and demographic characteristics

No.	Age (years)	Sex	Presentation	Dormans classification	Follow-up (months)	Union (weeks)	Cyst healing (months)	Capanna's classification	MSTS (%)
1	9	Male	Fracture	IB	108	6	9	I	100
2	9	Male	Incidental	IIB	30	–	9	I	100
3	6	Female	Fracture	IIB	30	6	18	I	100
4	9	Male	Thigh pain	IIB	24	–	12	I	100
5	11	Male	Thigh pain	IB	24	–	18	I	100
6	12	Female	Thigh pain	IIB	84	–	24	I	100
7	11	Female	Incidental	IIB	25	–	Residual	II	93.3
8	6	Male	Fracture	IB	26	8	12	I	100
9	9	Male	Thigh pain	IIB	30	–	Residual	II	100
10	5	Male	Fracture	IIB	24	6	12	I	100
11	9	Female	Fracture	IIB	28	6	9	I	100
12	7	Female	Incidental	IB	30	–	Residual	II	100
13	6	Male	Thigh pain	IIB	96	–	Residual	II	100
14	12	Female	Fracture	IIB	25	6	18	I	100

MSTS Musculoskeletal Tumor Society

up. No case was eventuated as class III (recurrence) or IV (no response).

The mean healing period was 14.5 ± 5.5 months in male and 13.5 ± 5.1 months in female patients (not statistically significant; $p = 0.6$). Moreover, no significant correlation was observed between patient age and healing period ($r = 0.063$, $p = 0.8$). The healing period was 13 ± 4.6 months in UBC with type IB and 14.6 ± 5.6 months with type IIB (not statistically significant; $p = 0.7$). Average time of fracture union was 6.33 ± 1.2 weeks (range 6–8). All fracture-positive cases healed completely (Capanna's class I). The healing period was 13 ± 4 months in fracture-positive and 15.7 ± 6.6 months in fracture-negative cases (not statistically significant; $p = 0.42$). MSTS score was 100% in all patients, except case no. 7, at 93.3% (mean 99.5%.)

Complications

No major complication such as growth arrest, avascular necrosis (AVN), or device failure was observed. There was one superficial infection (case 3), which was treated orally with antibiotics. There was one case of heterotopic ossification (case 10) of Brooker class I [10] and one of mild coxa vara with acceptable mild deformity (case 7). No complaint was reported by the patient during follow-up examinations, and consequently, no intervention was performed. No pathologic

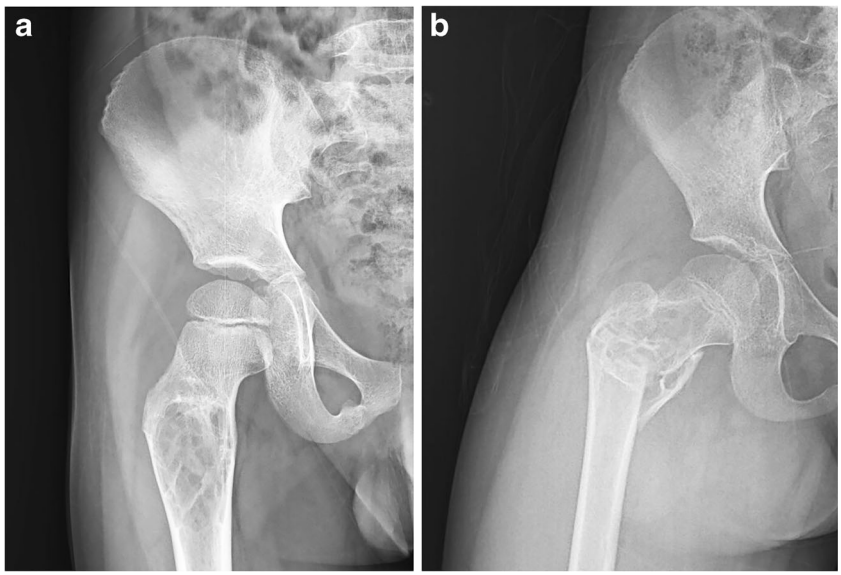

Fig. 1 **a** Anteroposterior radiograph of a Dormans type IB unicameral bone cyst (UBC) with femoral shaft defect. **b** Lateral radiograph of a type IIB UBC presented with fracture

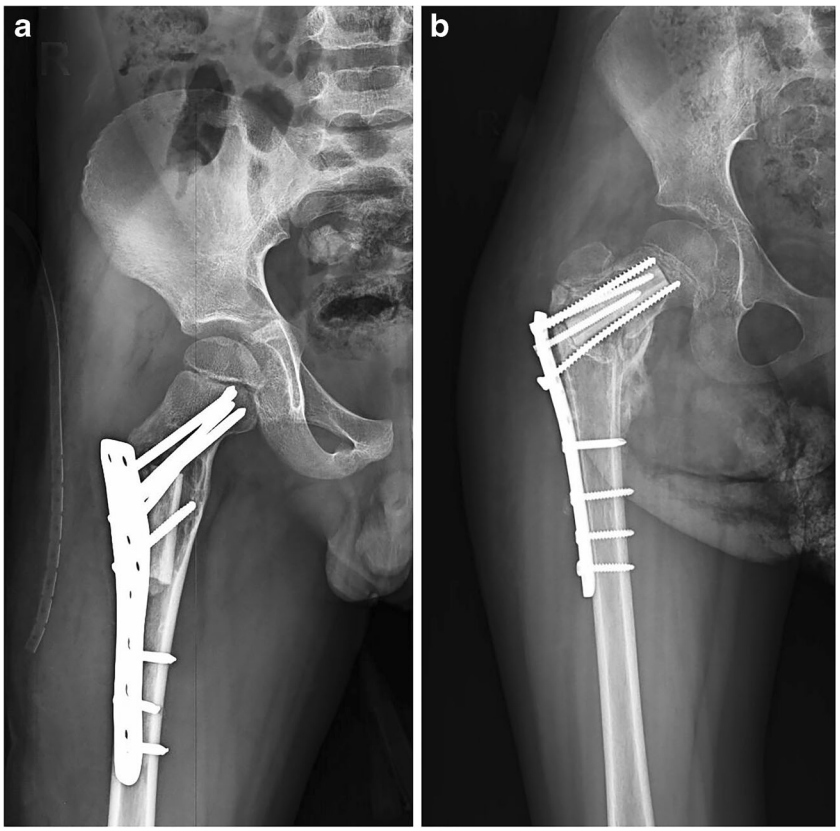

Fig. 2 **a** Anteroposterior radiograph of a Dormans type IB unicameral bone cyst (UBC) showing placement of fibular strut graft in the femoral shaft. **b** Lateral radiograph of a type IIB UBC showing placement of fibular strut graft in the femoral neck

fracture occurred during the healing period or until the end of follow-up.

Discussion

Simple bone cysts are identified as the underlying lesion in 40% of pathologic femoral neck fractures in children [11]. Many of these lesions involve the femoral neck or inter- and subtrochanteric regions (Dormans types IB and IIB), significantly weakening the bony framework [5, 12]. In such cases, the surgeon should deal with two difficult situations, including managing the bone cysts that endanger bone stability and treating the pathologic fracture through the bone cyst once it has occurred [13]. The high risk of recurrence and fixation failure have been noted as major drawbacks of conventional interventions [6, 14]. For this reason, managing UBC of the proximal femur is still challenging.

Several different therapeutic strategies have been applied in the treatment of UBC. However, evidence is not enough to support one treatment over another, and the optimal treatment is controversial [15]. Total or subtotal resection or saucerization have been associated with major drawbacks, including physeal damage, large amounts of intra-operative

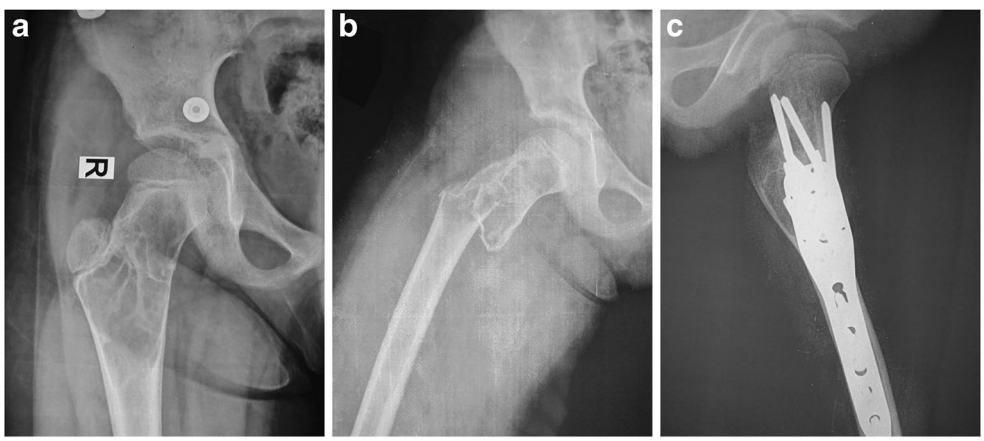

Fig. 3 **a** Anteroposterior radiograph of proximal femur of a six year-old girl showing a simple bone cyst Dormans type IIB (case 3). **b** Pathologic fracture occurred one week after UBC diagnosis. **c** Lateral radiograph showing complete healing 18 months after surgery

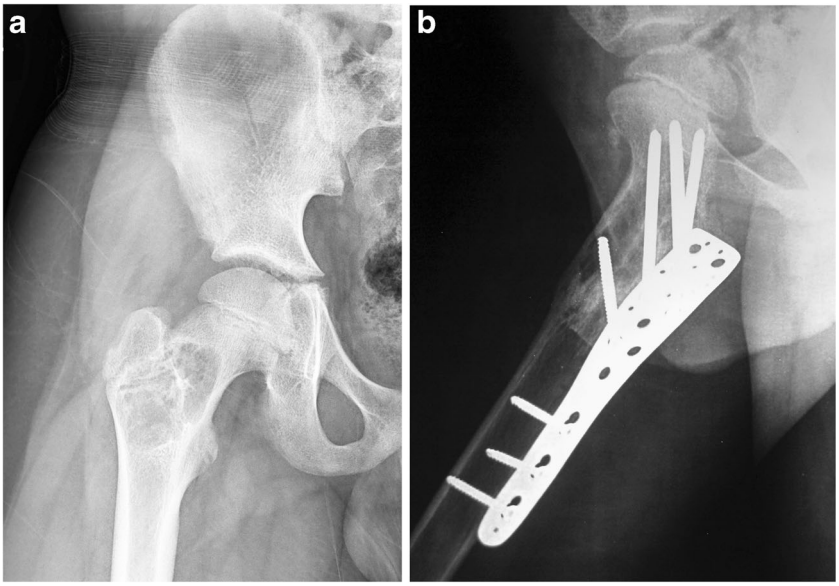

Fig. 4 **a** Anteroposterior radiograph of a proximal femur of a nine year-old boy showing a simple bone cyst Dormans type IIB (case 9). **b** Lateral radiograph showing incomplete healing with residual radiolucency 30 months after surgery

blood loss, intra-operative fracture, and a prolonged period of post-operative immobilization [16, 17]. Classical treatment with curettage and bone grafting causes large-bone defects and predisposes the bone to fracture. A large cortical window is also needed, which causes additional instability to the already weakened bone [18]. Moreover, the cancellous bone graft might be resorbed, leading to lesion recurrence [19]. Neer et al. reported the results of 129 primary UBC treated with intralesional excision and cancellous bone graft. The incidence of re-operation was 17% in the proximal end of the femur. They concluded that UBC recurrence following surgical treatment is significantly more frequent in paediatric patients [20]. Oppenheim and Galleno reported a complication rate of 17%, which included infection, coxa vara, epiphyseal arrest, and limb shortening. They concluded that the procedure had a high morbidity rate and added that curettage and bone grafting should not be considered in UBC treatment [12]. Intralesional steroid injections are also used to treat UBC. However, repeated injections are necessary in most cases, and a higher rate of recurrence is reported [21]. According to Hashemi–Nejad and Cole, the healing response to intralesional steroids is unpredictable and usually ineffective, even after multiple injections [22]. In another attempt, bone marrow from the iliac crest has been injected percutaneously into the cyst cavity to stimulate bone formation in the cyst with the osteogenic elements in the autogenous bone marrow. Yandow et al. reported the outcome of 12 UBC managed with this technique: eight (67%) patients demonstrated substantial healing, two (17%) showed partial healing, and two (17%) did not respond to bone marrow therapy. Pathological fracture occurred in four patients during treatment [23]. Treatment with multiple drill holes has also been attempted. Continuous decompression of the cyst is the essential feature of this method, which is performed by draining fluid through the cyst wall. Shinozakiet al. reported results of 23 UBC treated with this technique. A second surgery was needed in 12 patients due to cyst recurrence. Moreover, a third operation was necessary for three patients due to unsatisfactory consolidation of the cyst and the risk of a pathological fracture [24].

To the best of our knowledge, flexible intramedullary nailing has been proposed as the latest therapeutic strategy for UBC management. The essential feature of this method is the same as that of multiple drill holes and is reported to provide early stability, allowing early mobilization and obviating the need for a plaster cast while decreasing the prevalence of pathological fracture. However, this method has a complication rate of 25%, which includes nail perforation through the lateral cortex of the femoral neck and nail shortening following bone growth [13]. Recently, fibular strut grafts were proposed as an alternative to corticocancelous grafts in treating benign lesions of the proximal femur on the basis that they are less prone to bone resorption [19, 25]. Fibular strut graft also provides a firmer bed for the locking screws and prevents screw cutout, the most common mechanical failure of femoral-neck fixation. Proximal femoral locking plate was recently introduced as an implant that allows angular stability by creating a fixed-angle block for treating complex proximal femoral fractures [26]. We assumed that when fixation is extended to the femoral shaft (in Dormans types IB and IIB), a locking plate could act as a lateral buttress, and the addition of a fibular strut graft could potentially decrease the risk of fracture, graft resorption, and screw cutout. This procedure could also obviate the need for a plaster cast and allow early return to normal activity.

We evaluated the outcome of this procedure in 14 pediatric patients with Dormans UBC type IB or IIB of the proximal femur at a mean follow-up of 41.7 months. No case needed a

re-operation, and there was no recurrence, fracture, or screw cutout. Surgical complications comprised one superficial infection, one heterotopic ossification, and one coxa vara. A longer healing period was necessary in type IIB compared with type IB UBC, which could be explained by the larger defect in type IIB. No residual cyst was observed in fracture-positive cases, the healing period of such cases was shorter than for fracture-negative UBC. Thus, a better outcome could be expected when a UBC patient is referred with a pathologic fracture. These observations could be explained by the concomitant callus osteogenesis, which might accompany the healing cyst. The higher healing potential of the cyst after a pathologic fracture was also noted by Galasko [27].

Since most pediatric orthopedic interventions are intended to prevent further limitation of a child's physical activity or reduce existing physical disability, we believe our strategy of treating UBC using a locking plate and fibular strut-graft augmentation addresses these objectives sufficiently. The strength of our study was the homologous pediatric population with Dormans UBC type IB or IIB of the proximal femur; the main limitation is the small number of patients. Further studies with more patients and longer follow-up are needed to support our results.

Conclusion

Application of a locking plate and fibular strut-graft augmentation in pediatric patients with Dormans UBC types IB and IIB in the proximal femur could be considered an acceptable surgical strategy for managing these lesions and is associated with a favorable outcome and early mobilization.

Compliance with ethical standards

Conflict of interests statement On behalf of all authors, the corresponding author states that there is no conflict of interest.

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