Outcomes of osteoarticular ulna allograft for the reconstruction of proximal ulna tumour

a retrospective series of 13 patients

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Aims

The ulna is an extremely rare location for primary bone tumours of the elbow in paediatrics. Although several reconstruction options are available, the optimal reconstruction method is still unknown due to the rarity of proximal ulna tumours. In this study, we report the outcomes of osteoarticular ulna allograft for the reconstruction of proximal ulna tumours.

Methods

Medical profiles of 13 patients, who between March 2004 and November 2021 underwent osteoarticular ulna allograft reconstruction after the resection of the proximal ulna tumour, were retrospectively reviewed. The outcomes were measured clinically by the assessment of elbow range of motion (ROM), stability, and function, and radiologically by the assessment of allograft-host junction union, recurrence, and joint degeneration. The elbow function was assessed objectively by the Musculoskeletal Tumor Society (MSTS) score and subjectively by the Toronto Extremity Salvage Score (TESS) and Mayo Elbow Performance Score (MEPS) questionnaire.

Results

The mean follow-up of patients was 60.3 months (SD 28.5). The mean elbow flexion-extension ROM was 95.8° (SD 21). The mean MSTS of the patients was 84.4 (SD 8.2), the mean TESS was 83.8 (SD 6.7), and the mean MEPS was 79.2 (SD 11.5). All the patients had radiological union at the osteotomy site. Symptomatic osteoarthritic change was observed in three patients (23%), one of whom ended up with elbow joint fusion. Two patients (15.4%) had recurrence during the follow-up period. Surgical complications included two allograft fractures, two plate fractures, three medial instabilities, and two infections.

Conclusion

Osteoarticular ulna allograft reconstruction provides acceptable functional outcomes. Despite a high rate of complications, it is still a valuable reconstruction method, particularly in skeletally immature patients who need their distal humerus physis for the rest of hand growth.

Take home message

- Osteoarticular ulna allograft reconstruction yields satisfactory functional outcomes.
- Although it is associated with a high rate of complications, it remains a valuable reconstructive option, especially for skeletally immature patients who require preservation of their distal humeral physis to allow for continued hand growth.

Introduction

Primary bone tumours of the elbow are rare, accounting for almost 1% of all skeletal tumours.[1] Considering the complex interplay between the osseous and capsuloligamentous structures in the elbow, restoration of the elbow joint stability and function following the resection of the tumours is a significant challenge.[2]

The ulna is an extremely rare location for primary bone tumours of the elbow, and tumours of the proximal part of the ulna are even rarer.[3] Various reconstruction options have been introduced for proximal ulna tumours, including arthrodesis, pseudoarthrosis, autografts, allografts, reimplantation of sterilized tumoural bone, and endoprosthesis.[2] However, owing to the rarity of this tumour and the lack of high-quality evidence, there is no clear consensus regarding the optimal reconstruction method for proximal ulna tumours.[2]

In skeletally immature populations, the use of biological reconstruction allows the preservation of the distal humerus physis, which is responsible for most of the hand growth. However, both of the elbow physes are sacrificed when a megaprosthesis is used for such reconstruction, leading to an imminent reoperation after the closure of physics.[4,5] For this reason, biological reconstruction could be a more viable option in the paediatric population.

Although several biological reconstruction techniques have been used for the proximal ulna tumours,[2,3,6-10] outcomes of osteoarticular ulna allograft reconstruction after resection of the proximal ulna have not been reported in earlier studies. In this study, we aimed to report the outcomes of osteoarticular allograft reconstruction in a series of 13 patients with tumours of the proximal ulna.

Methods

This study was approved by the review board of our institute. Medical profiles of the patients with proximal ulna tumours who underwent surgical treatment in our tertiary referral hospital (Shafa Orthopaedic Hospital, Iran University of Medical Sciences, Tehran, Iran) between March 2004 and November 2021 were retrospectively reviewed. Inclusion criteria were primary bone tumour involving the olecranon, treatment with wide resection, and reconstruction with an ulna osteoarticular allograft, and a minimum follow-up of two years. Patients who were treated for a recurrence, patients who were referred with concurrent metastasis, patients who died before two years of operation, and patients lost to follow-up were excluded from the study. Overall, 12 patients who met the study criteria were included in the final analysis. The diagnosis of the tumours was histologically confirmed via preoperative core needle biopsy. All patients with malignant tumours received neoadjuvant and adjuvant chemotherapy. The flow diagram of the patient selection is demonstrated in Figure 1.

Surgical procedure and postoperative protocol

All the surgeries were done by the same senior musculoskeletal tumour surgeon (SHS). Under general anaesthesia and the application of a pneumatic tourniquet, the patient was placed in a lateral position with the whole forearm draped free. A direct posterior incision was used to approach the lesion, beginning at 2 cm proximal to the olecranon fossa and continuing over the subcutaneous border of the ulna, entailing the biopsy tract. After adequate exposure, the ulnar nerve was identified and protected. Then, the patients underwent en bloc resection with safe margins, which were planned on pre-treatment imaging modality (radiographs, MRI, and CT scans) and regarded to contain at least 2 cm of bony margin. The medial collateral ligament, annular ligament, and triceps tendon were cut at a maximum of 1 cm left to the insertion site and tagged with sutures to allow reconstruction. Muscle coverage to the proximal ulna was sacrificed as margins. Osteoarticular allografts of the proximal ulna with appropriate size and length were provided by our university bone bank, which harvests and prepares allografts according to the standard tissue banking protocols. The allografts were harvested within 12 hours post-mortem from the bodies of young adults who had died in traffic accidents.[11] The allografts were stored at 85°C for at least two weeks before their use. Before fixation of allograft to the host bone, a frozen section analysis of the distal ulna intramedullary sample was done to make sure of a clear margin. Then, the remnants of ligaments and capsules were sutured to the allograft. The triceps tendon was sutured to the olecranon with adequate tension using No. 5 (7.0 metric) polyester braided Ethibond sutures. The stability of the elbow joint was checked after reattachment of the triceps and ulna fixation. In case of instability (n = 4), we used an ulnohumoral cross pin to augment the stability, which was removed four weeks after the surgery. The distal part of the osteoarticular allografts was then aligned to the remaining ulnar stump, and osteosynthesis was performed using a seven-hole dynamic compression plate or stainless steel rush nails (Figure 2). An elbow brace was applied for four weeks after the operation. Passive assisted motion was initiated after brace removal. Elbow supination-pronation movements were encouraged as tolerated. A gradual increase in the range of motion (ROM) with stretching and muscle-strengthening exercises was advised. Lifting weights and loading were allowed once radiological union was observed across the osteotomy junctions. Follow-up visits were carried out every three months for the first two years, every six months for the next five years, and yearly thereafter.

Outcome measures

In the last follow-up visit, the outcomes of the patients were evaluated clinically and radiologically. Clinical evaluations included the assessment of elbow stability, ROM, and function. The elbow lateral and medial stability was checked with a varus and valgus stress test, respectively.[12] Elbow ROM evaluation included the assessment of elbow flexion, extension, supination, and pronation with a standard goniometer. The elbow function was evaluated objectively by the Musculoskeletal Tumor Society (MSTS) score,[13] and subjectively using the Toronto Extremity Salvage Score (TESS).[14] The elbow function was also subjectively evaluated by the Mayo Elbow Performance Score (MEPS) questionnaire, which is specifically designed to check the performance of the elbow joint.[12] All the questionnaires were presented on a 0 to 100 scale, and a higher score was indicative of better function. MEPS was also categorized into the excellent (≥ 90), good (75 to 89), fair (60 to 74), and poor function (< 60). Clinical evaluations were carried out by an orthopaedist (WA) who was not directly involved in the patient's care. One patient who had elbow joint fusion due to severe osteoarthritis and instability was excluded from the clinical evaluation.

In each follow-up visit, anteroposterior and lateral elbow radiographs were used for the evaluation of radiological outcomes, which included the assessment of tumour recurrence, union across osteotomy junction, and osteoarthritic changes. Clinical return of pain and swelling at the

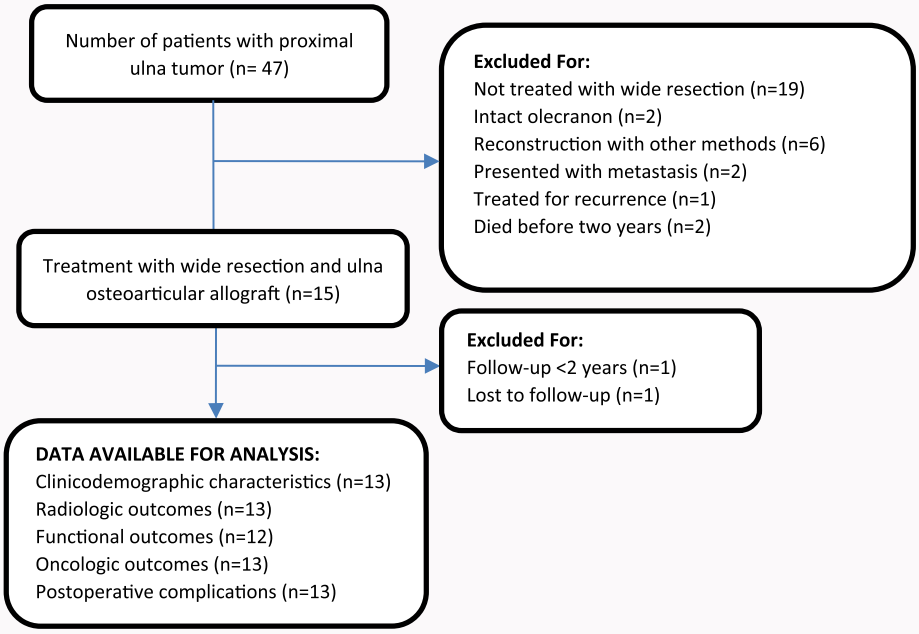

Fig. 1
Flow diagram of the study.

site of surgery was regarded as a sign of recurrence, which was followed by a radiological examination to confirm the recurrence. Union of the osteotomy (healing time) was defined as the time to observe four bony cortices at the osteotomy site in elbow radiographs. Osteoarthritic change of the elbow was evaluated using the Broberg and Morrey classification of elbow arthritis.[15] Accordingly, the elbow joint was graded into four grades (0, 1, 2, and 3), in which grade 0 showed the normal elbow joint, and grade 3 was indicative of severe degenerative change with significant destruction of the joint.[15]

Baseline characteristics

The study population included six males (6/13, 46.2%) and seven females (7/13, 53.8%) with a mean age of 12.3 years (SD 3; 8 to 18). The majority of the tumours were malignant (n = 12; 92.3%). The most frequent tumour type was Ewing's sarcoma (7 of 13, 53.8%) followed by osteosarcoma (5/13, 38.5%). The mean resection length was 8.1 cm (SD 2.2; 5 to 12). The mean follow-up period of patients was 64.1 months (SD 30.5; 24 to 120). The baseline characteristics of the patients are detailed in Table I.

Postoperative complications, including nonunion, fracture, infection, and instability, were extracted from the patient's profiles. Descriptive data were demonstrated with mean and SD for quantitative variables, and with numbers and percentages for qualitative variables.

Results

Clinical outcomes

The mean elbow supination/pronation ROM was 87.9° (SD 21.3°; 50° to 110°), and the mean flexion-extension ROM was 95.8° (SD 21°; 50° to 120°) (Figure 3). The mean MSTS was 84.4 (SD 8.2; 68 to 94), the mean TESS was 83.8 (SD 6.7; 70 to 90), and the mean MEPS of the patients was 79.2 (SD 11.5; 60 to 90). Accordingly, the elbow function was categorized as excellent, good, and fair in four (33.3%), five (41.7%), and three (25%) patients, respectively. Clinical outcomes are demonstrated in detail in Table II.

Radiological outcomes

All the patients had a radiological union at the junction of the allograft and host bone. The majority of the unions (10/13; 76.9%) were observed within 12 months of the operation. In two patients, the union was observed after 14 months of the operation. The mean time to radiological union was 9.8 months (SD 2.4; 7 to 14). Radiological osteoarthritic change was observed in ten patients (76.9%) who were clinically asymptomatic (grade 1) in seven patients and clinically symptomatic in three patients (Table II). Among patients with clinically symptomatic elbow osteoarthritis, eight patients had grade 2 degeneration, and one patient had grade 3 degeneration (Figure 4). This patient finally underwent elbow joint fusion due to severe pain and instability.

Oncological outcomes

Two patients (15.4%) had recurrence during the follow-up period that was managed with tumour resection and implantation of another osteoarticular allograft. Three patients had metastasis during the follow-up; one of them was concurrent to recurrence. Metastases were managed with metastasectomy. Two of these patients died of metastasis.

Postoperative complications

Based on the valgus stress test, three patients had medial instability. An elastic elbow band was used for two of these patients. The other one, who had severe medial instability concurrent to grade 3 elbow joint degeneration, underwent elbow joint fusion. None of the patients had lateral instability. Allograft fracture occurred in two patients and was managed with auto graft from pelvic and plate fixation (Figure 5). Plate

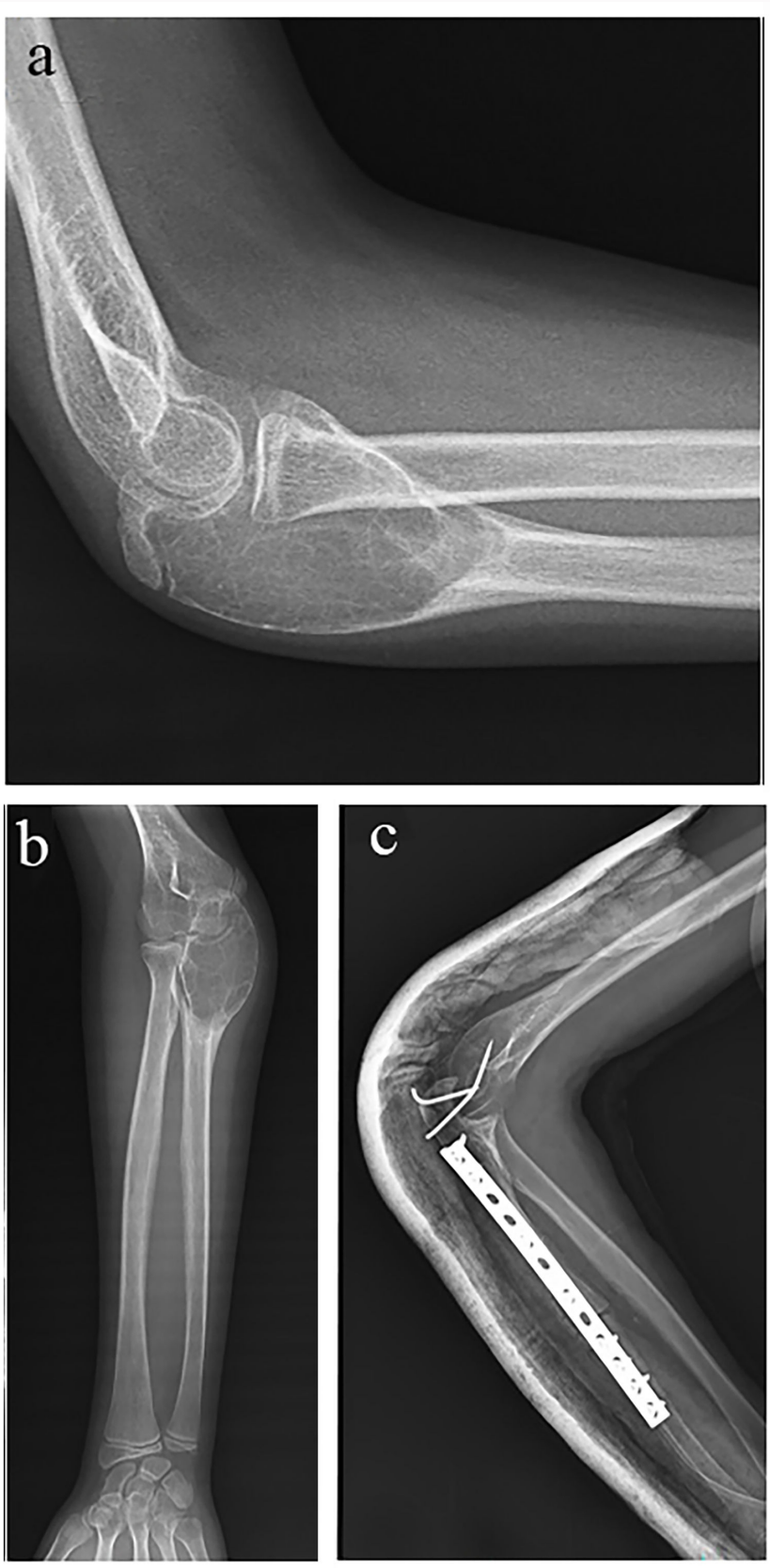

Fig. 2
a) Preoperative lateral and b) anteroposterior elbow radiograps of an 11-year-old female with proximal ulna telangiectatic osteosarcoma. c) Postoperative lateral radiograph of the same patient immediately after the operation.

fracture occurred in two patients and was managed with a new plate. Superficial infection occurred in two patients and was managed with debridement and oral antibiotics (Table II).

Discussion

In this study, we evaluated the outcomes of osteoarticular allograft reconstruction following the wide resection of

Table I. Baseline characteristics of patients with a proximal ulna bone tumour treated by wide resection and osteoarticular ulna allograft.

No.	Age, yrs	Laterality	Sex	Tumour type	Resection length, cm	Necrosis, %	Follow-up, mths
1	12	Right	Male	Ewing's sarcoma	8	95	70
2	10	Right	Female	Ewing's sarcoma	12	60	24
3	8	Right	Female	Ewing's sarcoma	6	80	64
4	8	Left	Female	Osteosarcoma	7	95	84
5	11	Left	Female	Ewing's sarcoma	9	95	72
6	14	Left	Male	Osteosarcoma	10	90	84
7	17	Right	Female	Osteosarcoma	8	75	30
8	18	Left	Female	Osteosarcoma	5	95	66
9	12	Right	Male	Ewing's sarcoma	6	55	32
10	10	Left	Male	Osteosarcoma	8	90	120
11	15	Left	Male	Desmoplastic fibroma	12	-	44
12	13	Right	Female	Ewing's sarcoma	8	95	34
13	14	Left	Male	Ewing's sarcoma	6	85	110

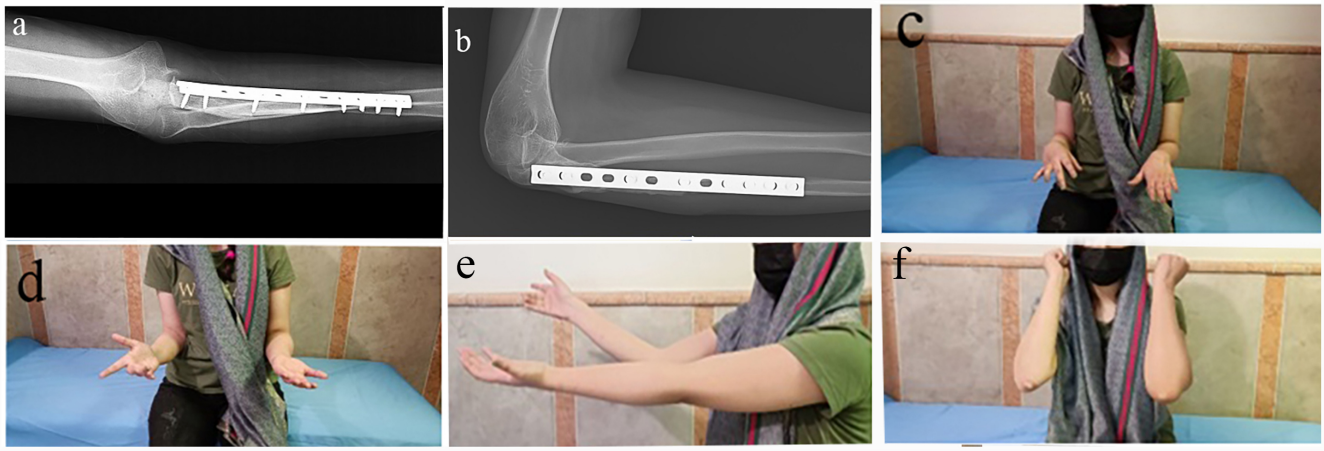

Fig. 3
a) and b) Anteroposterior and lateral radiographs of the elbow in a 12-year-old female patient showing union at the junction of the allograft and host bone four years after the resection of proximal ulna telangiectatic osteosarcoma and osteoarticular allograft reconstruction. c) to f) Clinical photographs of the same patient showing the elbow range of motion.

proximal ulna tumours. Osteoarticular allograft reconstruction provided an acceptable elbow function, ROM, and stability. Union of the allograft-host junction was observed within 12 months in 11 patients; two patients had a delayed union at 14 months. The radiological osteoarthritic change was observed in ten patients. However, only three of them were clinically symptomatic, leading to elbow joint fusion in one patient. Nine surgical complications were observed in a total of six patients, which included two allograft fractures, two plate fractures, three cases of medial instability, and two infections.

Various osteoarticular allograft reconstruction methods have been used for the reconstruction of the proximal ulna after tumour resection. Goyal et al[16] used a non-vascular autologous fibular graft for the reconstruction of proximal ulna in a 15-year-old male diagnosed with desmoplastic fibroma. At two years follow-up, flexion-extension elbow ROM was 40° to 130°. The patient had no restriction in daily living activities. Kimura et al[8] used a vascularized fibular autograft for the reconstruction of the elbow in an eight-year-old female with a proximal ulna tumour. Four years after the surgery, the patient has excellent elbow function and active movement of the elbow.[8] Ogose et al[10] used a free vascularized fibula graft in combination with an extracorporeally irradiated osteochondral graft for the reconstruction of the proximal ulna after resection of osteosarcoma. Ten years after the surgery, the patient was able to play golf with no elbow instability or pain. Megas et al[9] used a free, non-vascularized fibula graft for the reconstruction of the proximal ulna after the resection of metastatic clear cell renal carcinoma. The patient had no problem during the 25-month follow-up period, with a flexion/extension ROM of 90° and a MEPS of 75 points. The

Table II. Outcomes of patients with a proximal ulna tumour treated with wide resection and reconstruction.

No.	Supination/ pronation ROM, °	Flexion/ extension ROM, °	MSTS, %	TESS	MEPS	Union, mths	Degenerative joint disease, grade	Recurrence, mths	Metastasis, mths	Complication
1	100	110	92.7	88	90	8	-	-	-	-
2	65	65	74.4	70	65	14	2	8	-	Medial instability
3	85	100	91.3	89	80	9	1	-	6	-
4	50	50	80.4	80	60	12	1	-	-	Allograft and plate fracture, medial instability
5	110	110	85.2	88	80	8	1	-	-	Plate fracture
6	110	100	94.3	90	90	10	1	-	-	-
7	60	80	67.7	75	60	11	2	6	6	Infection, allograft fracture
8	105	105	81.5	83	80	10	1	-	-	-
9	75	90	77.3	78	80	14	1	-	-	Infection
10	90	120	88.8	84	85	8	1	-	8	-
11	110	110	88.2	90	90	7	-	-	-	-
12	100	110	91.1	90	90	8	-	-	-	-
13*	-	-	-	-	-	8	3	-	-	Severe medial instability

*Patient who had fusion.
MEPS, Mayo Elbow Performance Score; MSTS, Musculoskeletal Tumor Society; ROM, range of motion; TESS, Toronto Extremity Salvage Score.

Fig. 4
a) and b) Nine-year follow-up radiographs of a 23-year-old male showing severe degenerative joint disease at the left elbow joint causing limited flexion and supination (c and d), finally requiring elbow joint fusion.

use of fibular graft for the reconstruction of the proximal ulna has also been reported by several other authors, and the outcomes have been generally reported to be acceptable.[6,7,17] Gundavda et al[6] aimed to restore the elbow joint by reimplantation of the resected osteochondral segment of the proximal ulna after extracorporeal irradiation. They managed three patients with this method. At a follow-up of 28 to 42 months, all patients had a full range of elbow prono-supination and flexion-extension. Muscle power at flexion and extension was full. MSTS and MEPS were both 100, and the Disabilities of the Arm, Shoulder, and Hand questionnaire score[18] was zero for all patients. Union of the osteotomy junctions was observed in all patients within eight months without the need for any intervention. No postoperative complications were recorded.

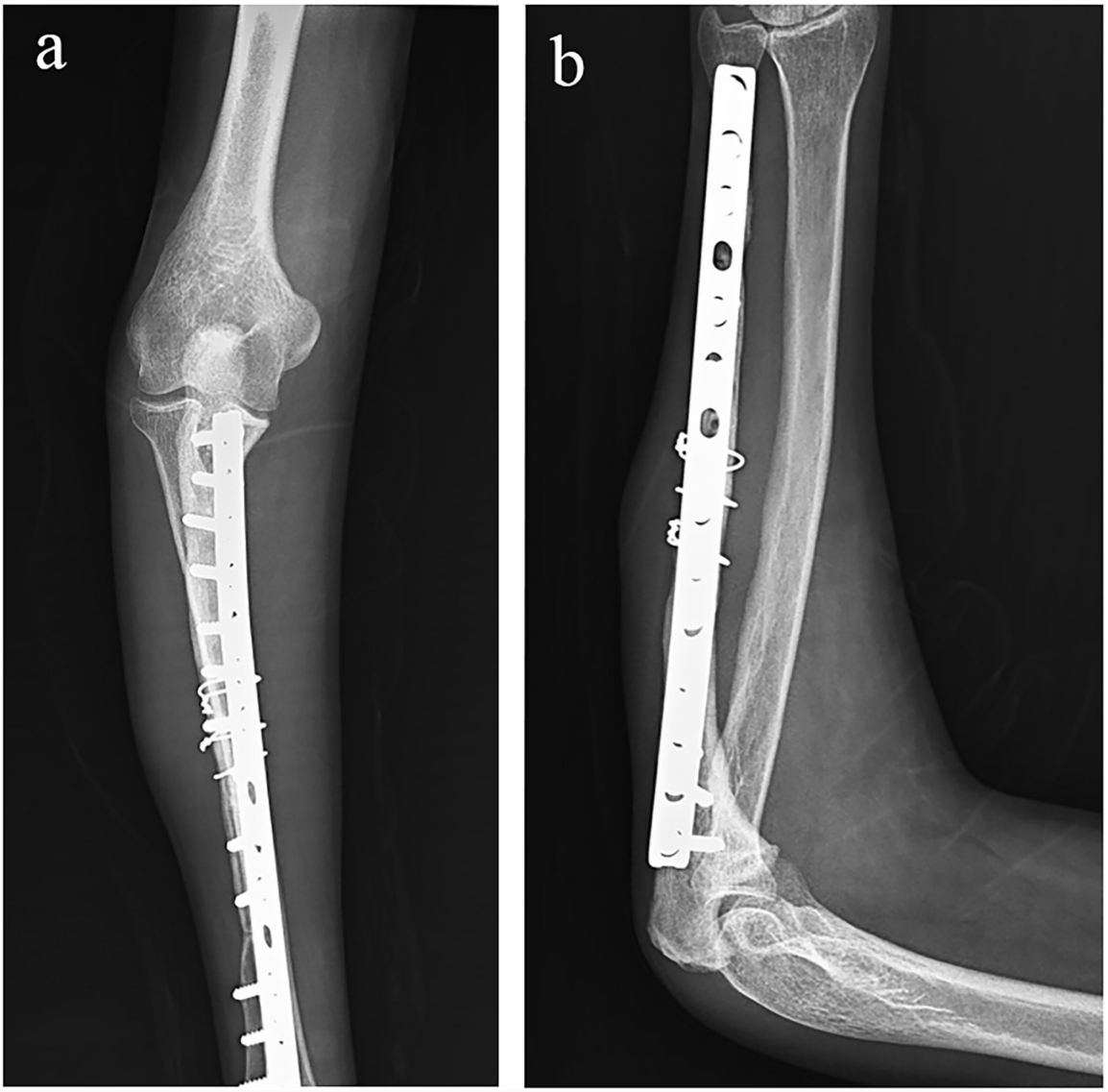

Fig. 5
Three-year follow-up radiographs of a 17-year-old female showing infection and allograft fracture after osteoarticular reconstruction of the elbow that was treated with repeated debridement and pelvic autograft fixed with wire.

None of the patients had clinical or radiological signs of joint arthritis.[6] Similar to the results of the present study, a review of the literature reveals acceptable outcomes of osteoarticular allograft for the reconstruction of the proximal ulna. However, almost none of the earlier studies report a serious postoperative complication after osteoarticular allograft reconstruction, while the present study shows that osteoarticular allograft is susceptible to a variety of postoperative complications such as allograft fracture, plate fracture, elbow instability, and joint degeneration. The same complications have been reported when osteoarticular allograft was used in other bones.[19,20] This difference could be attributed to the small number of patients in earlier studies or the short follow-up period.

Recently, proximal radius transfer, as a non-graft biological method, has been introduced as a reconstruction option for proximal ulna following tumuor resection. The functional outcomes of the patients managed with this procedure have been generally favourable, and no specific complication has been reported within a follow-up period of almost two years.[21,22] With these promising results, proximal radius transfer is worth considering for patients with malignant tumours of the proximal ulna in the future, providing that larger studies with longer follow-ups confirm the results of previous reports.

Non-biological reconstruction of the proximal ulna, mainly the megaprosthesis reconstruction, provides excellent early function. However, they are also prone to late complications such as implant loosening and failure, requiring revision surgeries.[23,24] Sewell et al[25] reported the outcomes of endoprosthetic arthroplasty in four patients with proximal ulna tumours. One patient required a transhumeral amputation for local recurrence after one month. Fixed flexion deformities of the elbow occurred in two patients, enforcing radial-head excision in one of them.[20] Non-biological reconstruction of the proximal ulna is also prone to late complications such as implant loosening and failure requiring revision surgeries.[19,20] In addition, the use of megaprosthesis in skeletally immature patients scarifies the growth plate at both ends of the elbow.

This will result in considerable limb shortening, requiring arthroplasty by a new prosthesis in the near future. Using an expandable megaprosthesis could potentially prevent limb shortening. However, it is not available in many orthopaedic centres, and when available, its complications are even more than a routine megaprosthesis.[26,27] The use of osteoarticular allograft seems a more reasonable option in paediatric with proximal ulna tumours. Since the distal humerus physis, which is responsible for most of the hand growth, remains intact, reconstruction with an osteoarticular ulna allograft does not considerably affect the child's growth.

The present study was not without limitations. The main limitation was its retrospective design, along with the small number of patients, which was posed by the rare incidence of proximal ulna tumours. The absence of a control group treated with another reconstruction method, such as megaprosthesis, could be regarded as another limitation of the study.

In conclusion, osteoarticular ulna allograft provides an acceptable function and elbow joint movement for patients with a proximal ulna tumour. Although it is prone to several postoperative complications, this reconstruction method is still of certain value for proximal ulna tumours, particularly in skeletally immature patients. It allows the preservation of distal humerus physis, which is responsible for most of the hand growth in the paediatric population. Therefore, the patients will not need a reoperation for limb shortening, which is generally required after the closure of elbow physes in patients who are managed with a megaprosthesis.

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The authors have no conflicts of interest to declare.

Data sharing
The data that support the findings for this study are available to other researchers from the corresponding author upon reasonable request.

Ethical review statement
This study was approved by the review board of Iran University of Medical Sciences under the code IR.IUMS.REC.1401.377.