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ORIGINAL ARTICLE

Treatment of persistent large cystic lesions of the humerus with vascularized fibular grafts

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ABSTRACT

Objectives: In this study, we aimed to evaluate the short-to-mid-term results of the resection and reconstruction of large cystic lesions of the humerus.

Patients and methods: Eight male patients (median age: 22.9±10.4 years; range, 12 to 42 years) with large cystic lesions of the humerus operated between January 2017 and December 2019 were retrospectively analyzed. The age of the patients, their previous treatments and follow-up periods, the size and location of the cysts, postoperative functional scores, presence of a union, recurrence of the cyst, and graft resorption were examined.

Results: The mean follow-up was 42.8±7.5 (range, 34 to 54) months. Preoperatively, the mean length of the cystic lesions was 15.1±2.6 (range, 10 to 18) cm. At the final follow-up, the patients had a normal range of shoulder flexion-extension, internal rotation-external, abduction-adduction, and elbow flexion-extension, pronation-supination. The patients had a mean DASH score of 1.13±1.1 (range, 0 to 3.3) and MSTS score of 28.75±1.8 (range, 26 to 30) postoperatively. Complications such as pseudoarthrosis, graft resorption, or cyst recurrence were not observed in any of the patients.

Conclusion: Although the risk of recurrence is low in small cystic lesions of the humerus, it increases as the size of the lesion increases. This reconstruction technique using vascularized fibular grafts, which we applied, seems to be extremely successful in ensuring biological healing and preventing recurrence and complications in patients with large cystic lesions of the humerus.

Keywords: Cystic lesions, humerus, vascularized fibula graft.

The most common benign bone lesions of the humerus in adolescence and childhood are aneurysmal bone cysts (ABCs), unicameral bone cyst (UBC), and fibrous dysplasia (FD). These lesions are often located on the metaphysis and diaphysis of the humerus.[1] Studies on large series of patients with cystic lesions that cause eggshell-shaped cortical deformations (Figure 1) and involve more than one-third of the humerus are limited in the literature. Simple bone cysts are defined as reactive or dysplastic lesions rather than true tumors. While these cysts mostly asymptomatic lesions and are usually detected incidentally, they sometimes manifest as pathological fractures, especially in the proximal humerus.[2] Unicameral bone cyst usually starts at the metaphysis of the bones and extends to the diaphysis.[3] Aneurysmal bone cysts are osteolytic, expansile, and hemorrhagic lesions.[2] While one-third of the lesions occur secondary to lesions such as giant cell tumor, chondroblastoma, osteoblastoma, chondromyxoid fibroma, FD, or non-ossifying fibroma,[4,5] they may very rarely emerge secondary to malignant tumors.[5]

Following the treatment, curettage, methylprednisolone injection, isolated bone powder, or bone substitutes injections, 10 to 30% recurrence has been reported and the only method that warrants cure is wide resection.[4,6] Currently, marginal resection is performed particularly in large lesions;[2] however, the main handicap of this method is the reconstruction problem. It is obvious that fibular grafts would cause

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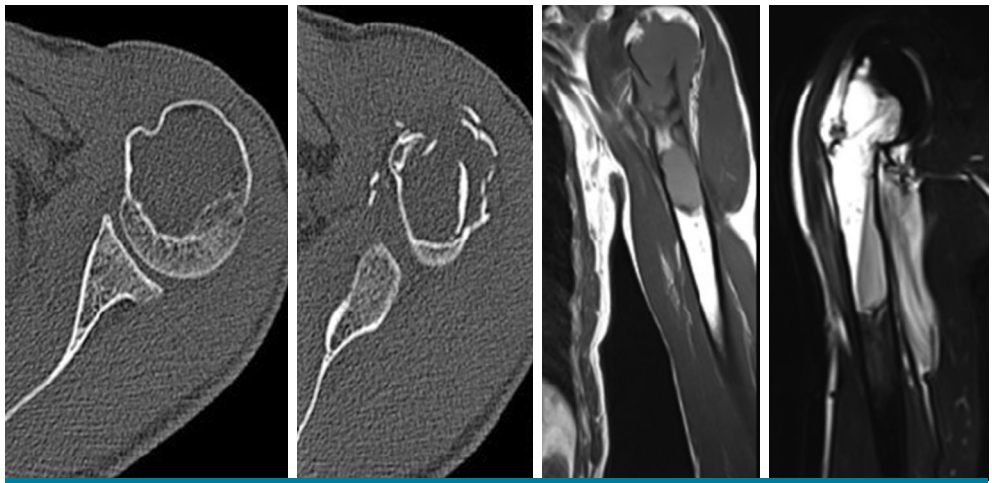

FIGURE 1. Large cystic, eggshell-shaped humeral lesion on computed tomography and magnetic resonance imaging scans.

problems in terms of bone consolidation and union when applied in lengths over 12 cm compared to vascularized grafts.[7]

In the present study, we aimed to evaluate the short-to-mid-term results of the resection and reconstruction of large cystic lesions of the humerus and to attempt to answer how we can reduce recurrence and complication rates.

PATIENTS AND METHODS

This single-center, retrospective study was conducted at Yeni Yüzyıl University Department of Orthopedics and Traumatology between January 2017 and December 2019. A total of 10 patients with large cystic humerus lesions who underwent reconstruction with wide resection and vascular fibular grafting (VFG) were reviewed. Of these, eight males (median age:

FIGURE 2. Preoperative and postoperative radiographs of the previously operated cystic humerus lesion.

22.9±10.4 years; range, 12 to 42 years) who regularly attended to follow-up visits and had complete records were included in the study.

Four of our patients previously underwent two to five operations due to cystic lesions of the humerus (Figure 2). Two other patients were followed conservatively after the fracture and were not cured (Figure 3), while two patients were at risk of pathological fracture due to the size and symptoms of the cyst (Figure 4). Six of the patients had pathological fractures; two of them were treated conservatively, while four received surgical treatment. At the time of admission to the clinic, the large humeral cyst persisted in all patients after conservative or surgical treatment and that pseudoarthrosis developed in five of them. Lesions were localized to the diaphysis in two patients, extended from the distal metaphysis to the diaphysis in two, and extended from the proximal metaphysis to the diaphysis in four. Five of these lesions were diagnosed as ABC, two as UBC, and one as FD. The posterior incision was performed in one of the patients with a diaphyseal cyst since his previous interventions were posterior. Three patients with diaphyseal and distal cysts were performed a lateral incision (due to previous surgeries, usually old incision sites were preferred), while the other four patients with proximal cysts were performed an extended deltopectoral incision.

Four patients who underwent cyst resection and reconstruction with VFG previously underwent at least two unsuccessful surgical interventions such as curettage-grafting, intra-cystic injections, and curettage-internal fixation.

Surgical technique

The cyst site was entered with 20 to 25 cm incisions over the previous surgical incisions. The lateral or anterior cortex was elevated over the humerus.

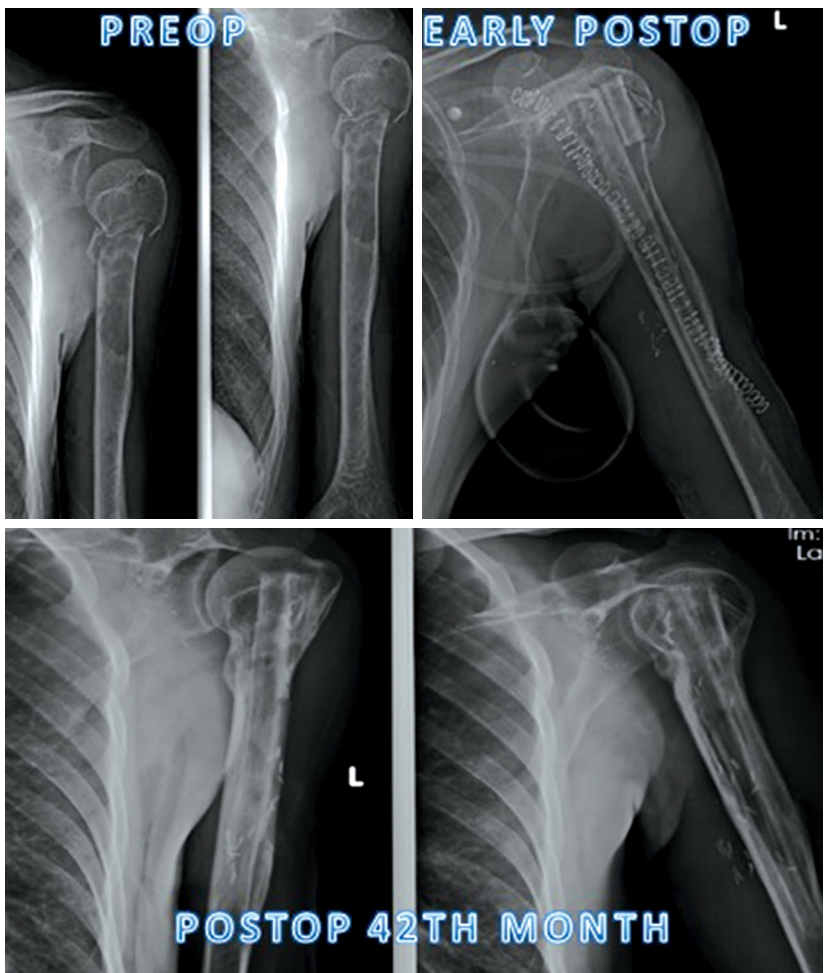

FIGURE 3. Preoperative and postoperative radiographs of the patient in whom conservative treatment failed.

The cystic lesion in the entire humerus was curetted and sent for pathological examination. Our aim was to preserve the length of the extremity. The inside of the cyst was curetted and cleaned thoroughly from the proximal and distal, until intact bone tissue was seen (Figure 5). The intact part of the humerus in the proximal and distal was reached, and the site in the intramedullary canal where the fibular graft would be placed was prepared with flexible reamers and the length of the graft to be used was determined. Based on the size of the cyst, a 12 to 20-cm free bone flap was elevated from the fibula on the same side, leaving appropriate safety margins around the fibula, knee, and ankle. During this procedure, at least 5 cm of the proximal and distal parts of the fibula were preserved, and no additional fixation was performed for tibiofibular syndesmosis (Figure 6). The flap donor site was closed appropriately. Postoperatively, a below-knee plaster splint was applied.

The harvested vascularized fibula was intramedullary placed in the defect area in the humerus. Other defects observed in the humeral head in two patients were filled using a 2-cm free fibular graft in one patient and a cancellous graft was taken from the iliac bone in the other. The harvested cortical grafts were closed on the flap, and cerclage wire was applied from the proximal or distal humerus, when necessary (Figure 5). No additional fixation materials were used in any of the patients. The branch of the brachial artery was anastomosed with the flap's artery using an 8-0 non-absorbable surgical suture.

Prior to intramedullary placement of the fibular graft, its placement in the distal or the proximal was decided and the recipient site was prepared. After deciding on the location of the recipient's vein, the recipient artery was passed through the window opened in the bone that enables the recipient artery to reach that region medially, and anastomosis was provided in this region. Proper planning and graft placement were essential to prevent complications. At this stage, the appropriate length of the graft was determined for precise harvesting, and if the graft was desired to preserve its vascularity, the planning was done according to the position of the veins. In particular, in cysts that are located in the proximal metaphysis and that beget large bone defects, both the application of a double-barrel fibular graft and an iliac bone graft was necessary to fill this region and to ensure the stability of the graft. The main fixation in the diaphysis was provided by intramedullary placement of the graft.

The Velpeau bandage was applied to our patients after dressing. The patients that were followed in the ward were discharged on the third postoperative day, since no complications were observed. The sutures were removed on Day 15 of follow-up. The Velpeau bandages were removed after three weeks, and passive elbow and shoulder exercises were initiated. Again, at the end of the third week, the patients were clinically and radiologically evaluated. In addition, the active movement was started, but the patients were not allowed for lifting weights and applying axial loading.

All patients were evaluated at the final follow-up visit. Bilateral radiographs of the humerus were taken and evaluated for cyst recurrence, presence of graft resorption, and union of the graft. Again, at the final follow-up, any pathological movement was assessed in terms of joint range of motion and muscle strength, and the Disabilities of the Arm, Shoulder

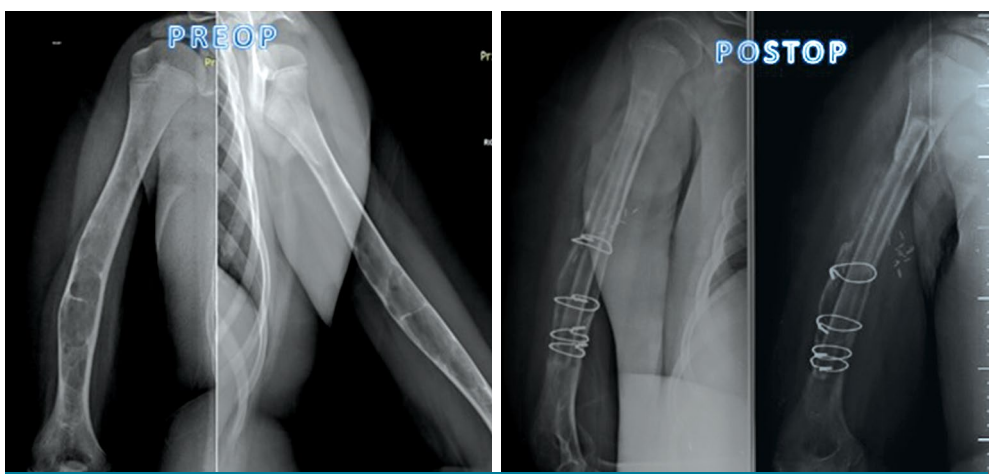

FIGURE 4. Large symptomatic humeral cyst.

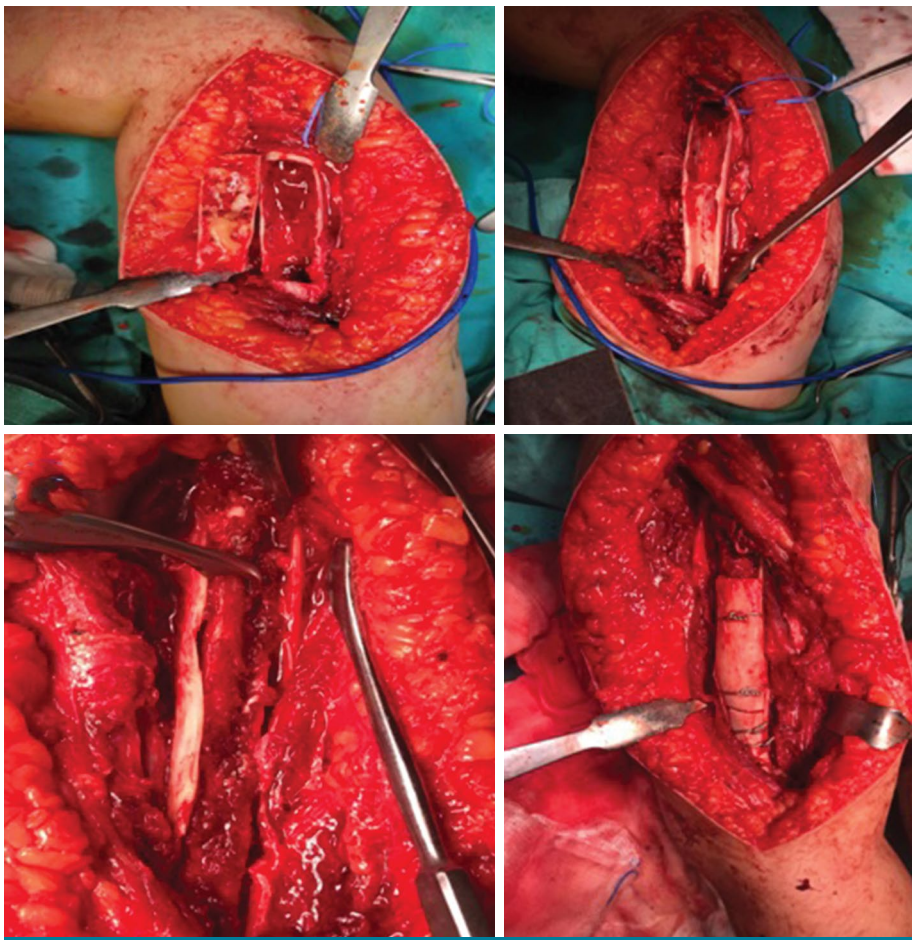

FIGURE 5. Removal of the cyst and preparation of the intramedullary cavity.

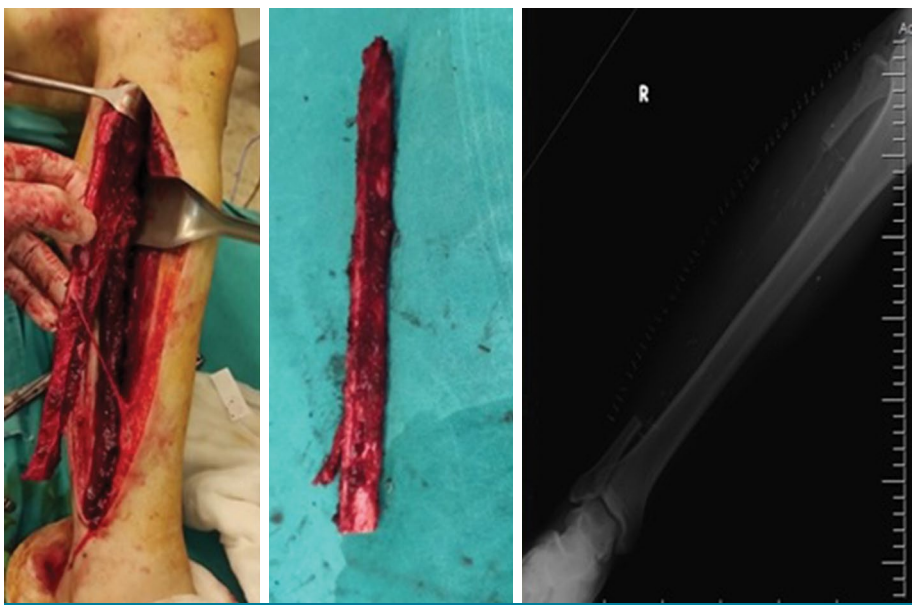

FIGURE 6. Vascularized fibular graft harvesting.

TABLE I
Postoperative ranges of motion of patients

	Mean (°)	Range (°)
Shoulder flexion	174.3	170-180
Shoulder extension	55.6	50-60
Shoulder internal rotation	67.5	58-70
Shoulder external rotation	86.2	79-88
Shoulder abduction	175	165-180
Shoulder adduction	54.3	50-60
Elbow flexion	143.1	135-150
Elbow extension	-4.2	0-10
Elbow pronation	76.9	70-80
Elbow supination	81.9	73-85

and Hand (DASH) scores and the Musculoskeletal Tumor Society (MSTS) scores were calculated.

Statistical analysis

Statistical analysis was performed using the NCSS version 2007 software (NCSS LLC, Kaysville, UT, USA). Descriptive data were expressed in mean ± standard deviation (SD), median (min-max), or number and frequency.

RESULTS

Three of the patients were younger than 18 years old. The mean follow-up was 42.8±7.5 (range, 34 to 54) months. At the final follow-up, one of our patients under 18 years of age had shortness of 2.5 cm in the operated extremity. The reason for this was thought to be physeal damage due to the extension of the cyst to the proximal humeral physis.

Preoperatively, the mean length of the cystic lesions was 15.1±2.6 (range, 10 to 18) cm. Vascularized fibular grafts at least 2 cm longer than the cyst size were used to contribute to intramedullary fixation. In six patients, cerclage wire was applied from the proximal or distal part of the humerus. Also, no internal or external fixation materials were used.

The ranges of motion at the final follow-up are given in Table I. The patients had a mean DASH score of 1.13±1.1 (range, 0 to 3.3) and MSTS score of 28.75±1.8 (range, 26 to 30) postoperatively. No cyst recurrence or graft loss was observed in our patients, while all attained bone continuity with no findings of nonunion (Table II).

DISCUSSION

Although various methods are used in the surgical treatment of benign cystic lesions, the main purpose in all treatments is to achieve a high rate of recovery and union, a low rate of recurrence, and return to previous daily activities as early as possible.[8]

Morbidity after surgeries performed due to cystic lesions of the humerus is low, yet surgical treatment is particularly preferred in patients with pathological fractures. Although the risk of recurrence is low in small lesions, this risk increases as the lesion size increases.[9] These lesions are usually described as those that do not require specific treatment and disappear spontaneously with bone maturity. Although it is assumed that for many cases the fracture stimulates healing, it has been reported only in 5 to 10% of the cases.[10,11]

Until now, several methods of treatment have been described. The irradiation method, which was described in the past years, is no longer used due to its low success rates and the risk of malignant transformation.[12] Again, intralesional corticosteroid or bone marrow injection has been performed as

TABLE II
Data and scores of patients

Patients	Age (year)	Diagnosis	Location	Cyst size (cm)	Complication	MSTS (postoperative)	DASH (postoperative)
1	42	UBC	Proximal to diaphysis	15	No complication	30	1.7
2	35	UBC	Proximal to diaphysis	16	No complication	26	0.8
3	19	ABC	Proximal to diaphysis	15	No complication	30	0.8
4	12	ABC	Proximal to diaphysis	13	2.5 cm shortness	28	1.7
5	21	ABC	Distal to diaphysis	18	No complication	30	0.8
6	17	ABC	Distal to diaphysis	10	No complication	30	0
7	23	ABC	Diaphysis	18	No complication	26	3.3
8	14	FD	Diaphysis	16	No complication	30	0

UBC: Unicameral bone cyst; ABC: Aneurysmal bone cyst; FD: Fibrous dysplasia.

the conventional method.[13] *En-bloc* or partial cyst resection, which have been used for many years, are considered as other alternative methods.[14] However, considering the low chance of success mentioned with the classical methods and the increasing rate of failure with the increase in lesion size, wide resection of the cyst may be the preferred treatment method. Meanwhile, how to fill the remaining the bone space still remains a question.

The iliac or other cancellous autograft stock would not be sufficient to fill the remaining defect after *en-bloc* resection or curettage, particularly in patients who present with pathological fractures and who cannot recover, despite repeated surgeries and long follow-up periods, as was the case in our series. In this context, fibular grafting may be the desired treatment method in such patients, since it is both a cortical bone and allows harvesting of large amounts of grafts. In a related study, successful results have been reported in large cystic lesions of the humerus with *en-bloc* resection followed by non-vascularized fibular graft transfer.[15] Although fibular grafts between 8 and 20 cm were used in this study and no graft loss was mentioned, free vascularized fibular grafts are a good option, particularly in defects larger than 6 to 7 cm, and that full union is achieved in about six months.[16] In a study describing the reconstruction of segmental defects with non-vascularized fibular grafts, the authors found a significant relationship between mechanical complications such as fracture and delayed union/nonunion and the presence of a defect >12 cm.[17]

Based on our experience with this series, we had some implications for the intramedullary placement of the fibula and fracture stabilization. Particularly in patients with large cysts, very proximal or very distal localization of the cyst close to the joint may cause problems for proximal and distal osteosynthesis. Intramedullary placement can prevent these problems and enable the fibula to be used as an aid for fracture fixation.

The use of non-vascularized fibular grafts is advantageous in terms of operative time, and it requires less experience. While it is undeniable that vascularized fibular grafting requires experience, it is a known fact that it provides better osteoconduction, osteoinduction, osteogenesis, mechanical strength, and vascularity when assessed regarding all other graft options.[18] Since it is known that non-vascularized fibular grafts have no biological activity and possess a high risk of resorption, vascularized fibular grafts have been used for approximately 40 years in the reconstruction of defects due to their advantages.[19,20] It has also been shown that vascularized grafts are superior to non-vascularized grafts in terms of hypertrophy, resistance to bacterial infection, and union.[21,22] Considering all the aforementioned advantages, the vascularized fibular graft should be preferred to minimize the possible complications, particularly in cases similar to ours where the cyst size is large. Therefore, the fibula graft to be applied should be large. Also, in cases with a previous history of osteomyelitis and still at risk of osteomyelitis, as was the case in two patients in our series, the vascularized fibular graft is another reason for preference due to its resistance to bacterial infection.

In a meta-analysis examining 110 articles on the use of vascularized fibula grafts in orthopedic surgery between 1983 and 2020, a total of 2,226 patients were evaluated, while VFG to the humerus was performed in only 9.6% of the cohort.[23] This finding suggests that surgical interventions requiring VFG application in the location in which we present data are not very frequent. An evaluation of the surgical reasons in these patients revealed that 47.3% of them were operated on due to tumor, 0.5% due to previous unsuccessful surgeries, and 0.4% due to FD. In particular, no distinction was made between benign and malignant tumors; however, a review of the aforementioned literature showed that the majority of them were malignant ones (47 studies included only oncological patients). Again, only 17% of these patients were operated on for upper extremity pathologies. In these studies, while the patients were fixed with various methods, we did not come across the use of isolated cables as in our study. On the other hand, there were 294 patients on whom screws were used as a minimal fixation method, similar to our study. Although the number of cases was low in our series, we believe that finding a large cohort with such large defects involving only the humerus is truly difficult. The fact that there is no similar study in the literature regarding large benign bone lesions, particularly involving the humerus, renders this study a valuable one, which would give a clearer idea for future studies with larger case series.[24]

The intramedullary application of the graft and its cut that is larger than the size of the lesion, the preparation of the intramedullary canal, compressed placement of the graft from the proximal or distal, and application of cerclage wire when necessary, as explained in the technique, is the best example of intramedullary fixation. Again, in a study conducted on similar lesions to understand the

benefit of intramedullary fixation and compared curettage grafting and elastic intramedullary fixation to curettage grafting alone, the authors reported the recurrence rates as 10 to 32.2% in favor of additional intramedullary fixation.[25]

In hemicortical reconstruction techniques performed similar to our technique, better results were reported with a greater contact surface between the graft and the host bone and preservation of cortical continuity.[26] Similarly, Lenze et al.[17] reported successful results in benign and early malignant cases in which they applied hemicortical reconstruction with non-vascularized fibula grafts and recommended its use in defects of <12 cm due to the risk of graft resorption. Considering the graft size we used on our patients, the authors had a similar conclusion with ours.

The rates of union and hypertrophy of vascular fibular grafts have been reported as 85 to 100% in the upper extremity, while the success rate of the same technique in the lower extremity has been reported as 75 to 86%, and the difference was associated with the absence of mechanical forces in the upper extremity and long-term load prohibition in the lower extremity.[20,22] Similarly, a union rate of 93% was reported five months after the application of vascularized fibular grafts following resection due to oncological pathologies in the humerus.[27]

Considering complications, this rate was observed to vary between 6 and 100% in current studies. These complications were mostly refractures, nonunions, delayed unions, infections, vascular thrombosis, and donor area complications. The most frequently used scoring system in these studies was MSTS, with an mean score of 25.6 (range, 21.8 to 29.4).[23] In our study group, the mean MSTS score was found as 28.75 (range, 26 to 30), which can be interpreted to be compatible with the literature and satisfactory. The major causes of these complications can be listed as weight-bearing on the extremity and subsequent chemotherapy and radiotherapy. Of note, we believe that our results were more satisfactory, as our patients had upper extremity pathologies, which allowed us to be more protective regarding weight-bearing, while the absence of the need for chemotherapy and radiotherapy also reduced the risk.

This technique, together with its modifications, still stands before us as a technique that has no alternative, particularly in the treatment of large bone defects, despite its high complication rate and risks. Nevertheless, various alternatives have been attempted to be produced recently. The most prominent of these is the use of scaffolds with or without augmentation.[28,29] While this technique creates a new structure for the bone, it allows us to avoid the technical difficulties and complications of VFG. However, it should be kept in mind that whether the technique would provide sufficient success, particularly in large bone defects, is still a controversial issue and that, despite all its negative aspects, VFG is a widely used technique currently.[23]

Due to the low prevalence of such large cystic lesions, the small number of patients, the absence of a comparison group, and the lack of very long-term follow-up results can be regarded as the main limitations to our study.

In conclusion, considering that the patient population we examined underwent repeated surgeries or long-term treatment processes, we can be described as a group that is almost intolerant of failure in terms of surgery, which makes it essential to apply the surgical technique with the highest chance of success to the patients. In this context, the reconstruction technique with vascularized fibular grafts, which we applied after the removal of the cyst in the humerus, was extremely successful in ensuring biological healing and preventing recurrence and complications, such as graft loss in patients with cystic lesions of the humerus. Nevertheless, further studies are warranted.

Ethics Committee Approval: The study protocol was approved by the Tekirdağ Namık Kemal University Ethics Committee (date/no: 30.03.2021/2021.73.03.13). The study was conducted in accordance with the principles of the Declaration of Helsinki.

Patient Consent for Publication: A written informed consent was obtained from each patient.

Data Sharing Statement: The data that support the findings of this study are available from the corresponding author upon reasonable request.

Author Contributions: Idea/concept: İ.B.Ö.; Design: M.Ü.Ç.; Control/supervision: İ.B.Ö.; Data collection and/or processing: M.Ü.Ç.; Analysis and/or interpretation: M.Ü.Ç.; Literature review: M.Ü.Ç.; Writing the article: M.Ü.Ç.; Critical review: İ.B.Ö.; References and fundings: M.Ü.Ç.; Materials: İ.B.Ö.

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