

# Fibular strut graft for humeral aneurysmal bone cyst with varus deformity

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## Abstract

**Purpose** Proximal humerus is a common site for ABC and frequently associated with varus deformity that limits shoulder abduction. A prospective study was conducted to evaluate the use of intramedullary non-vascularised autogenous fibular strut graft for reconstruction without internal fixation.

**Methods** A total of 20 patients (12 girls, 8 boys) were managed for proximal humeral ABC with varus deformity by extended curettage, osteotomy, intramedullary fibular graft and composite bone substitute. Their ages ranged from 10 to 17 years (average, 13.3 years). The lesion was juxtaphyseal in 16 patients and metaphyseal in 4. All cysts were active and centrally located type 2. The modified Enneking scoring system was used for final functional evaluation. Radiological assessment was done for the extent of defect healing, incorporation of the fibula and correction of the deformity.

**Results** After a mean follow-up of 41.2 months (range, 24–74) most of patients were satisfied and resumed daily activities without pain and with good range of shoulder movement. One patient complained of shoulder pain 10 months after surgery and was attributed to local recurrence. Limitation of recreational activity was experienced by one patient. There were no cases of deep infection, nerve deficit or pathological fracture. No cases of failed healing or incorporation of the fibula was

detected. The improved shoulder abduction was closely related to the mean correction of the neck shaft angle.

**Conclusions** The technique is proved to be effective in controlling disease, correcting deformity and improving function.

**Keywords** Fibular strut · Aneurysmal · Varus deformity

## Introduction

Aneurysmal bone cyst (ABC) is a rare benign expansile cystic lesion characterised by blood-filled cavernous spaces separated by septae containing osteoid tissue and osteoclast giant cells [1]. These lesions are not uncommon in the proximal humeral metaphysis and humeral shaft, but because of their widespread occurrence as a secondary lesion on a pre-existing tumour such as simple bone cyst, giant cell tumour, chondroblastoma, osteoblastoma, fibrous dysplasia and osteosarcoma, the true incidence is unknown [2, 3]. Although benign, ABC can be locally aggressive and cause extensive bone destruction and weakening predisposing the patient to repeated pathological fracture. The muscle pull about the proximal humerus allows healing of pathological fractures in progressive varus deformity with the usual complaint of marked limitation of shoulder abduction and flexion.

The management of such condition should be directed not only to eradicate the disease but also to improve function by correcting the deformity. The recently introduced less invasive treatment options such as percutaneous sclerotherapy and selective arterial embolisation have been reported for ABCs, particularly in difficult to reach locations, with variable success rates. However, these procedures are operator dependant and may not be available in every hospital. Also, the long treatment course, the need for repeated operations and the associated soft tissue morbidity limit their use [4–11].

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Furthermore, varus deformity of the proximal humerus could not be corrected after these techniques, maintaining the limited overhead activity. Intralesional excision by extended curettage with or without bone grafting has been used for primary cysts, while en bloc resection has been reserved for recurrent lesions or lesions located in expendable bones that can be removed without the need for bone reconstruction [12–16]. Abrasions of all surfaces using a high-speed burr and local adjuvants such as phenol, liquid nitrogen, argon beam coagulator or concentrated ethanol have been shown to decrease the rate of local recurrence [3, 11, 17, 18]. The commonly used filling materials such as autogenous cancellous or corticocancellous bone graft, allogenic freeze-dried cancellous bone graft, demineralised bone matrix and ceramic bone substitute usually take the shape of the lesion maintaining the angular deformity and the limitation of shoulder movement [19]. On the other hand, correction of the deformity by osteotomy usually requires the application of internal fixation devices which may be difficult and risky for proximal humeral location in children.

The current study was conducted to evaluate the results of using fibular strut graft as an intramedullary splint supporting the corrective osteotomy and avoiding the need for internal fixation after extended curettage of proximal humeral ABCs with varus deformity.

## Materials and methods

From September 2005 to February 2013, 24 patients with aneurysmal bone cysts of the proximal humerus were selected and managed by extended curettage and intramedullary fibular strut graft at the King Saud Hospital, Unizah, KSA and Mansoura University Hospital, Mansoura, Egypt. All patients had sustained at least one pathological fracture through the cyst that healed in varus angulation of the proximal humerus. Pathological fractures were treated conservatively to full union before any surgical intervention. Patients with recent pathological fractures, recurrent cysts after other methods of treatment, secondary aneurysmal bone cysts or pathologically proved simple bone cysts were excluded from the study. All patients or their guardians were informed about the benefits and the possible complications of the procedure and a signed consent was obtained to participate in the study. Of the 24 patients managed in this way, 4 were lost to follow-up and were excluded from the study. The remaining 20 patients included 12 girls and 8 boys with an average age of 13.3 years (range, 10–17) at presentation. The main complaint was limitation of shoulder abduction in ten patients, pain in seven and swelling in three with a mean duration of symptoms of 6.7 months (range, 3–10). The lesion was located on the left side in 11 patients, juxtaphyseal within 5 mm of the proximal humeral physis in 16 patients and mainly metaphyseal in 4.

The diagnosis was based on the typical clinical and radiological criteria and confirmed by histopathological examination of tissues obtained at the time of surgery. For patients with atypical features, a preoperative needle biopsy was performed. The typical magnetic resonance imaging (MRI) findings of multiloculation, complete internal septation and fluid-fluid level formation were helpful for diagnosis. Furthermore, the extent of proximal humeral physeal involvement by the lesion, which may reflect the possibility of premature fusion and the subsequent shortening of the humeral segment, was evaluated preoperatively by MRI [20, 21]. Broaching of more than one-third of the proximal humeral physis was considered risky for postoperative epiphyseal fusion and limb shortening. The approximate volume of the cyst was measured using the preoperative plain radiographs and more accurately by computed tomography (CT) scan by multiplying the maximum length and width in anteroposterior projection and depth in lateral projection. This could give an impression about the aggressiveness of the lesion and the amount of the graft required. According to the classification of Capanna et al. [22] and the grading system of Enneking [23], all lesions were centrally located type 2 and active. The lesions were extensive, involving from 25 to 75 % the humeral length and associated with either thinning or involvement of the outer cortex.

## Operative technique

All operations were performed under general anaesthesia, with patients in the beach chair position. A deltopectoral approach was used to reach the lesion. A large rhomboid window was made at the anterolateral wall lateral to the biceps tendon. The periosteum was kept attached along the medial side of the window. In this way the periosteal flap can be used to seal the cavity at the end of surgery. After exposure of the whole cavity, the inner walls were curetted using different sizes of curettes and the tissues obtained were sent for histopathological examination. A power burr was used to extend the margin of excision and to open the medulla in some cases but was avoided at the broached areas of physis. All septae and ridges were excised to flatten the inner surface. An argon beam coagulator (Birtcher 6000, Electrosurgical generator+ Argon Beam Coagulator; , Irvine, CA, USA) was used as a local thermal adjuvant in nine patients and phenol as a chemocauterisation adjuvant in 11 patients. The approximate length of the fibular graft was decided preoperatively from imaging and intraoperatively by adding an appropriate intramedullary length to the length of the defect. The graft was obtained subperiosteally from the proximal fibula of the ipsilateral extremity with care to avoid injury of the common peroneal nerve. The diaphyseal end of the fibular graft was contoured to the shape of the humeral medullary canal and inserted intramedullary from proximal to distal. The angulation at the cyst allowed intramedullary placement of the fibula

without much soft tissue obstruction. Subperiosteal osteotomies of the anterior and posterior walls of the cyst were performed at the site of the maximum angulation that is the point where the intramedullary fibula met the humeral shaft. A cortical wedge based laterally was removed from the anterior and posterior walls in most of the cases, leaving the medial osteoperiosteal wall of the cyst intact, which was osteoclased gently while trying to reduce the humeral head over the proximal end of the fibula. For juxtaphyseal lesions, the proximal end of the fibula was inserted to the centre of the proximal humeral physis, and for metaphyseal lesions it was impacted into the remaining cancellous bone of the proximal humeral metaphysis. The stability of the impacted fibula was then checked, and if it was in question, interosseous sutures through the cyst walls were used to stabilise the osteotomy and to prevent the fibula from being displaced laterally out of the cyst. No implant was used for fixation. After gently collapsing the outer cortex, a composite of synthetic bone substitute (ceraform; calcium phosphate hydroxyapatite 65 % and tricalcium phosphate 35 %; Teknimed, Vic en Bigorre, France) and autogenous bone marrow aspirate was used to fill the gaps around the grafted fibula. Finally, the periosteal flap, that was elevated initially, was used to cover the window, keeping the granules of bone substitute inside (Fig. 1). After surgery, the shoulder was immobilised in 30–40° abduction with an arm sling and arm-to-body immobiliser for 4–6 weeks, followed by passive pendulum movement of the shoulder. During the period of immobilisation, patients were instructed to move fingers, wrist and elbow out of the sling without active shoulder abduction 2–3 times a day. Passive shoulder abduction was started 8 weeks after surgery and gradually progressed to assisted active then active range of motion with the progression of the radiological healing.

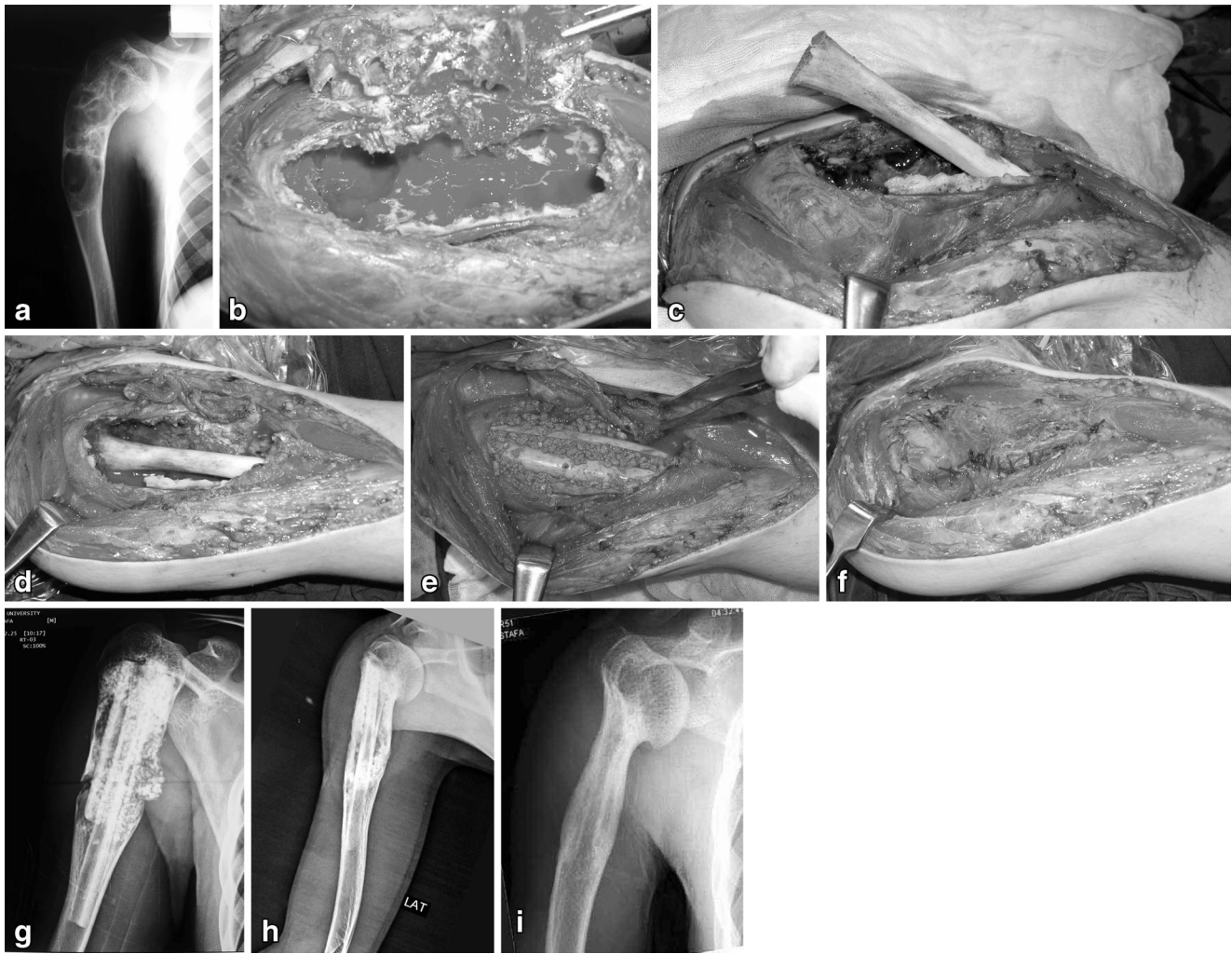
All patients were evaluated clinically and radiologically every month for the first 6 months, every 3 months till completing the 2nd year after surgery, then every 6 months thereafter and at the final follow-up. Clinical assessment was done for pain, alignment, limb shortening, limitation of shoulder motion, functional and recreational activity restriction and complications especially at the donor site. All clinical measures obtained at the time of final follow-up were compared with measures taken preoperatively and that of the contralateral uninvolved limb. The modified Enneking scoring system [24] was used for final function evaluation. Plain radiographs of both arms were obtained preoperatively, immediately after surgery and at follow-up with the limbs at the same position and same magnification distance to decrease the measurement errors. All radiographs were independently reviewed by the authors and two musculoskeletal radiologists to decrease the interobserver errors. Radiographs were examined for trabeculations, internal callus formation, bone density, residual radiolucent areas, evidence of local recurrence and borders between the fibular graft and the cavity. The criteria proposed

by Shih et al. [25] were adopted for final radiological evaluation of cyst healing and fibular graft incorporation. The lesion was considered healed if the preoperative cavity was completely obliterated with or without small radiolucent areas (less than 1 cm) and partially or incompletely healed if large radiolucent areas persist. The healing failed if the cavity was not obliterated and no evidence of trabecular bone formation or the graft was resorbed. Recurrence was diagnosed if lytic lesion reappeared in a previously obliterated area or a residual radiolucent area has increased in size. Incorporation of the fibular strut graft was considered complete if the graft was completely obliterated and partial if the graft was still visible but its borders were blurred. The fibular graft was considered not incorporated if its outer contour was unchanged from that of the initial radiograph. The time to healing was defined as the time required to achieve cortical thickening as well as consolidation of the lesion or the time at which no further healing progress was noted on follow-up radiographs. Radiological evaluation also included the measurement of neck shaft angle (the angle between the longitudinal humeral axis and the perpendicular to the articular margin plane) on plain radiographs before surgery, immediately after surgery and at final follow-up. These measures were compared with that of the normal uninvolved humerus to determine the extent of correction of the varus deformity.

The data of patients and the results were tabulated and analysed using the Statistical Package for Social Sciences (version 22; IBM SPSS, New York, USA). Pearson chi-squared test, independent sample *t*-test and one-way ANOVA test were used to evaluate factors that may influence cyst healing and fibular graft incorporation, and to define relations between clinical and radiological results and the final outcome. Probability values of less than 0.05 were considered significant.

## Results

Although the deltopectoral approach and the technique of extended curettage were familiar to the surgeon, subperiosteal harvest and intramedullary impaction of the fibular graft was technically demanding and time consuming, with the mean duration of surgery 105.3 min (range, 90–145). The mean size of the lesion at the time of presentation was 175.2 cm<sup>3</sup> (range, 62–483), while the mean length of the fibular graft was 14.5 cm (range, 8–19). All patients were followed-up for a mean of 41.2 months (range, 24–74). At the time of final follow-up most of patients were enthused by the surgery, had resumed their daily activities without pain and with a good range of shoulder movement. Only one patient experienced dull aching of the affected shoulder 10 months after surgery, which was attributed to local recurrence. This patient required a second intervention by extended curettage and bone



**Fig. 1** **a** An 11-year-old boy with ABC of the proximal right humerus. **b** Intraoperative photo showing the wide window lateral to the biceps tendon after elevation of periosteal flap. **c** The fibular strut graft implanted intramedullary. **d** Impaction of the proximal end of the fibula

under the humeral head with correction of the deformity. **e** Composite bone substitute filling gaps around the fibula. **f** Window closure using the periosteal flap. **g–i** Follow-up radiographs showing complete healing of the lesion and incorporation of fibula

graft to relieve pain. The residual varus deformity of the proximal humerus was enough to limit recreational activity in one patient. The mean active shoulder abduction was improved from 119.4° (range, 55–160°) preoperatively to 164.5° (range, 145–180°) at final follow-up. Shortening of the humeral segment and subsequent limb length discrepancy was detected in 17 patients, with a mean shortening of 30.6 mm (range, 10–100). Large-sized and juxtaphyseal lesions and lesions which broached two-thirds or more of the proximal humeral physis were significantly associated with more shortening of the humeral segment than small-sized and metaphyseal lesions or lesions away from the growth plate by sufficient cancellous bone ( $p=0.001$ ). The mean functional score (rating percentage of normal) at treatment completion was 98.3 % (range, 87–100 %).

Radiological healing of the defect was seen at a mean time of 6.5 months (range, 4–10) after surgery. Healing was

complete in 18 patients and partial in 1 patient, while recurrence was detected in 1 patient. Consolidation and remodeling of the implanted fibula were observed between 2 and 4 years after operation. At the final follow-up, radiographic evaluation revealed complete obliteration of the fibular strut graft in 11 patients and incomplete incorporation with blunting of the borders between the fibula and the host bone in 9 patients. The mean neck shaft angle of the proximal humerus improved from 106.6° (range, 90–122°) preoperatively to 136° (range, 125–145°) at final follow-up. The mean correction of the humeral neck shaft angle was significantly correlated with the improved shoulder abduction and the high functional score at final follow-up ( $p=0.01$  and 0.05). There were no cases of deep infection, nerve deficit or pathological fracture. Only one patient developed numbness and tingling in the distribution of the common peroneal nerve that improved spontaneously after 3 weeks. This was explained by excessive

traction on the nerve during fibular graft harvest. Superficial infection of the operative wound was seen in one patient and was controlled with repeated dressing and systemic antibiotic. Subperiosteal harvesting of the fibular graft allowed restoration of new fibula with reconstruction of its trabecular structure and bone marrow canal in most of patients, all of them were younger than 15 years. Restoration of the fibula at the donor site was not related to the length of the harvested graft.

## Discussion

Aneurysmal bone cysts of the proximal humerus pose a particular problem in management because of the close proximity of the lesion to the proximal humeral physis and the associated varus deformity that required correction. Currently, curettage and bone grafting with or without adjuvant therapy is the accepted treatment option for ABCs despite a relative rate of recurrence ranging from 10 to 59 % [4, 12, 16, 17, 26]. En bloc resection has been associated with the lowest rate of recurrence compared with other management methods. However, complex reconstructive procedure is often necessary and the technique looks too much for the treatment of such benign lesions. Moreover, en bloc resection is not always feasible due to location of the lesion and its close proximity to important structures. Therefore, en bloc resection is reserved for expendable bones and in the event of multiple recurrences [15, 27]. Intralesional injection of sclerosants was reported to be simple and effective for inaccessible sites which are difficult to treat surgically due to the risk of heavy bleeding. However, the need for multiple injections and prolonged treatment course, as well as the local soft tissue complications such as inflammation, dermatitis, indurations, cutaneous fistula formation and hypopigmentation, are obvious disadvantages. Pulmonary embolism and fatal case of cerebellar infarct following Ethibloc injection have also been reported [5–7]. Recently, embolisation of the feeding arteries has been used as a safe, minimally invasive and effective alternative to surgery for the treatment of ABCs. The main advantage of selective arterial embolisation is the avoidance of radical surgery. However, the procedure is technically demanding and is not applicable to all cases as ABC often lacks large afferent vessels. The main limitations of the procedure are the long time of treatment protocol, the need for repeated imaging and control angiography and the risk of embolisation of adjacent normal vessels in proximal limb girdles with subsequent skin breakdown, nerve palsy, muscle necrosis and infection [8–11]. Despite the promising effect of these less invasive techniques in controlling the disease, the associated varus deformity of the proximal humerus could not be appreciated without surgical intervention. Allograft cortical strut grafts have been used successfully to reconstruct humeral bone defects after excision of benign cystic lesions. In addition to acting as bone graft supplementing bone

healing, cortical struts provided immediate mechanical support, eliminating the need for any internal fixation devices. Although allografts have the advantage of unlimited supply without additional donor site morbidity, their incorporation process is slow and probably less complete compared with autograft due to a low-grade immune response or a lack of osteocytes in the graft. Furthermore, allografting is not available in every centre and carry a small risk of disease transmission [25, 28]. Autogenous fibular graft either vascularised or non-vascularised has been reported for the treatment of large benign humeral lesions [29, 30]. Vascularised grafts do not rely on revascularisation and become fully incorporated sooner than non-vascularised graft. This technique avoids necrosis and resorption of the incorporated graft and the new osteogenic process; hence, it does not weaken and remodels in a similar way to the normal bone. However, vascularised fibular graft is technically demanding and time consuming, with a high rate of thrombosis of the graft vessels [31].

In the current study, authors selected a group of patients with ABCs and significant varus deformity of the proximal humerus limiting the overhead activities. They proposed the use of free non-vascularised fibular graft as an intramedullary strut after extended curettage and they corrected the deformity by moving the humeral head over the graft like a hat on a head after osteotomy. The osteotomy through the thin cortex to correct the varus deformity further weakens the bony structure and metallic internal fixation devices are often needed for stabilisation. However, conventional internal fixation devices such as intramedullary nails or plates and screws are difficult to apply at this location and are associated with morbidity in children [25, 32, 33]. In the present series, autologous non-vascularised fibular strut graft provided an excellent structural bone support to the bone defect and the osteotomy site, and there was no need for any fixation device. Bone stabilisation was obtained by means of graft impaction. The diameter of the bone marrow cavity of the humeral bone was large enough to allow insertion of the cortical end of the fibula into the canal after slight adjustment, while the metaphyseal end was impacted into the proximal humeral metaphysis. The mechanical loading of the strut grafts is primarily in compression and some bending or torsion in certain situations. These mechanical demands are much less in upper limb than in lower limbs. Healing of the strut graft to the adjacent host bone further reduced the mechanical demands on the strut. In the first few weeks after surgery the mechanical strength of the graft is reduced, but the full mechanical properties return after 6–12 months [34]. No fracture or resorption of the fibular graft was encountered and the time to healing was comparable with that of other studies using cortical strut allograft [25, 28]. This could be explained by the good blood supply in the proximal humerus and preservation of the

periosteum, which allow rapid revascularisation and healing of the fibular graft.

Shortening of the humerus after surgical treatment of ABCs was reported to be related to the close proximity of the lesion to the proximal humeral growth plate [29, 32]. Lin et al. [35] suggested an association between growth impairment and

damage to the growth plate during surgery. Ramirez and Stanton [14] noted on the basis of MRI results that damage to the growth plate caused by the cyst is directly responsible for growth impairment. In the current study, humeral shortening was closely related to large-sized lesions, juxtaphyseal location and broaching of two-thirds or more of the growth plate



**Fig. 2** **a** A 17-year-old girl with ABC involving about 75 % of left humerus. **b** Apparent deformity and shortening of the arm and marked limitation of shoulder abduction. **c** Three-dimensional CT image. **d** Broaching of more than two thirds of the growth plate. **e, f** Intraoperative photos showing placement of the fibular strut graft

intramedullary and correction of the deformity. **g-i** Postoperative and follow-up radiographs displaying progressive healing of the defect and near complete incorporation of the fibula. **j** Improved function despite shortening of 10 cm

by the cyst. On the other hand, iatrogenic injury to the growth plate after extended curettage could not be excluded as a cause of growth impairment. Despite the relative shortening of up to 10 cm, no patient was keen to have limb lengthening because of the improved range of motion and function of the affected arm (Fig. 2). The improved shoulder abduction at the time of final follow-up was significantly related to the mean correction of the varus deformity of the proximal humerus. This gave the importance of correcting the deformity in addition to eradicating the tumour.

Radiological assessment of cyst healing and incorporation of the fibular strut was difficult. However, the use of radiographic signs such as blunting or complete obliteration of the fibular borders, as signs of graft remodelling, was quite subjective but reasonably qualitative. Abuhassan and Shannak [36] reported the results of non-vascularised fibular graft for reconstruction of bone defects after en bloc resection of giant ABC in three patients. They observed insufficient graft incorporation at the distal part of the fibular graft in the humerus case and advised rigid internal fixation to prevent graft insufficiency. Grzegorzewski et al. [29] analysed the long-term results of surgical en bloc resection of benign humeral lesions and reconstruction with autogenous non-vascularised fibular graft without fixation. They noted that whenever the periosteum was excised with the cystic lesion, the process of fibular graft incorporation was much slower. In the present study, placing the fibular strut graft intramedullary facilitated union because of the large contact area between the fibula and the endosteal surface of the humerus. Moreover, preservation of the outershell and the periosteum further enhanced the defect healing.

## Conclusions

Management of ABC of the proximal humerus with varus deformity is challenging. Although the primary goals of surgery are to eradicate the tumour and prevent recurrence, correction of the varus deformity is important to improve shoulder abduction. The intramedullary impacted autogenous fibular strut graft acted as a template of the humeral shaft to correct the deformity and mechanically as an internal splint to increase structural stability and biologically as a bone graft material to enhance defect healing. Despite the limited number of patients, the technique is proved to be effective to cure the lesion, improve function of the shoulder and avoid morbidities associated with the metallic implant used for internal fixation.

**Conflicts of interest** No benefits of any form have been or will be received from a commercial party related directly or indirectly to the subject of this manuscript.

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