Treatment of benign lesions of humerus with resection and non-vascularised, autologous fibular graft

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Abstract The aim of this study was to analyse the long-term results of surgical en bloc resection and replacement with non-vascularised, autologous fibular graft for the treatment of large benign humeral lesions without fixation. We retrospectively reviewed data of seven females and 13 males with unilateral benign lesions where steroid injection, curettage and bone grafting or pathological fracture failed to restore integrity. Subperiosteal, en bloc resection of the cystic lesion with a margin of the normal bone was performed. The average age of the patients at the time of operation was 11.8 years (range 4–28 years). All patients were skeletally mature at last follow-up. Aneurysmal bone cysts were histologically identified in seven cases, solitary cysts in 11 and fibrous dysplasia in two cases. No recurrence of the pathology, pain, graft fracture or limitation in range of motion was noted. In three patients in whom the cyst was adjacent to the proximal growth plate of the humerus, there was shortening of the bone at the last follow-up examination measuring 2 cm, 4.5 cm and 6 cm, respectively. Two cases had a valgus deformity of 10° and 15°, respectively, which was evident radiographically at the time of last follow-up. The results of en bloc resection with non-vascularised, autologous fibular graft for the treatment of large benign humeral lesions without fixation are encouraging. The risk of associated complications is low. In our opinion this should be the method of choice in the treatment of large, multi-chamber benign bone lesions of the humerus which fail more “conservative” treatment.

Introduction

Aneurysmal bone cysts, solitary bone cysts and fibrous dysplasia are benign lesions most often diagnosed in humeral bones of children, adolescents and young adults. These are frequently found in the proximal humeral metaphysis or in the humeral shaft. Pathological fracture is often the first and sometimes the only sign of the disease [1]. These benign lesions usually do not initially generate either pain nor limitation of joint movement. These lesions are often incidental findings when X-ray films are taken for assessment after trauma or for the investigation of limb asymmetry. For most of these lesions the treatment of choice includes intralesional steroid injection and curettage with or without bone grafting. However, despite this treatment these cystic lesions are associated with a high percentage of complications. Bone cyst recurrence, refracture and pseudarthrosis have all been observed and reported in the literature [2–5]. Adjunctive therapy such as concentrated ethanol or the use of a high-speed mechanical burr has been shown to decrease the rate of local recurrence [6, 7].

The limited success rate of current management options has prompted further research to be undertaken in order to identify new therapeutic approaches. Steroid injections targeted directly into the cyst cavity resulted in a very high recurrence rate in the treatment of aneurysmal and solitary bone cysts [8–10]. Wright et al. reported that 42% of simple bone cysts treated with methylprednisolone injection and 23% treated with bone marrow healed [11]. However, recent reports of treatment with platelet growth factors injected into bone cysts have shown high therapeutic efficiency in dental surgery [12]. In the case of failure, treatment of multi-chambered bone cyst with cortical layer destruction and non-union after pathological fracture by en

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bloc resection gives the lowest rate of recurrence but is a more extensive procedure and should be reserved for selected cases. Bone reconstruction with vascularised or non-vascularized fibula graft is also an option for management of bone cysts refractory to first line treatment with bone marrow injection or steroid injection or curettage with bone grafting [3, 5, 13–15].

We present our long-term results of surgical en bloc resection with non-vascularised, autologous fibular graft for the treatment of benign humeral lesions after failure of conservative or operative treatment. Fixation was not used in any of our cases.

Materials and methods

We retrospectively reviewed all cases (159) in our unit of proximal humeral or humeral shaft benign cysts treated with bone marrow injection, steroid injection or curettage with bone grafting. We found a group of patients with humeral lesions treated in a nonstandard way who underwent en bloc excision and a non vascularised fibular autograft. There were 20 patients included in the study—seven female and 13 male—treated between 1983 and 2004 at our institution. All lesions were extensive, involving from 25% to 40% length of the humeral bone shaft and the proximal metaphysis. These lesions were associated with either thinning or involvement of the cortical layer. In three cases bone cysts were adjacent to the proximal humeral growth plate. We additionally calculated the radiographic ratio of the lesion according to the method of Kanellopoulos et al. [16]. The extent of the lesion on the longitudinal axis was divided by the normal expected diameter of the long bone at the site of the lesion. The radiographic ratio ranged from 3 to 5.8.

The age of the patients at the time of operation ranged between four and 28 years (mean 11.8 years). Three patients were over 18 years old at the time of surgery. In 13 of these cases pathological fracture was the presenting sign of the disease (Table 1). During follow-up we did not observe healing or remodelling signs of benign lesion from six to 12 months after fracture. Next, all patients from this group (three patients with lesion adjacent to growth plate) underwent at least two steroid injections into the cyst without any response (minimum three months interval). The maximum follow-up after the last injection was six months. In seven patients en bloc excision was not the primary surgical procedure. These patients had recurrence of the cyst after previous curettage and bone grafting with either thinning or involvement of the cortical layer. Patients who had en bloc resection after steroid injection without curettage and bone grafting were operated in the early years of our institution. Nowadays, en bloc resection is performed

only in cases with failed previous conservative and surgical treatment.

The first stage surgery consisted of subperiosteal resection of the fibula graft, the length of which varied between 8 and 20 cm. We attempted to preserve the periosteum in order to provide the opportunity of rebuilding the fibula in the future. The bone graft was taken ipsilaterally. When the graft length was below 10–12 cm, one skin incision was made. For longer grafts a two skin incision technique was performed. The reason for this technique was cosmetic appearance. A proximal incision of 5 cm length was made above the proximal part of fibula and the same length incision above the distal part of fibula. Next, fibula bone was exposed, periosteum was carefully dissected and subperiosteal resection of the fibular graft was made. In patients over 15 years of age at the time of operation we had problems with preserving periosteum during dissection. We stabilised the tibiofibular syndesmosis with one AO screw, when the distance between the distal end of the fibular osteotomy and the distal tibiofibular joint was less than 5 cm. This was necessary in seven of our patients. The periosteum was then closed to facilitate fibular reconstruction. Post operative leg immobilisation was not necessary in any of the patients. The screw was removed after six weeks.

In the second stage of the operation, we subperiosteally resected (en bloc) the cystic lesion. The ends of the fibular graft were contoured to the shape of the humeral medullary canal and the fibula was then inserted from proximal to distal. In three cases of resection of metaphysal cystic lesions extending to the physis and involving the physis, the bone graft was inserted to the epiphysis. We left growth plate in these cases and performed chemical resection by using 96° alcohol. The stability of impacted fibula was checked. No implants were needed to fix the bone fragments. The humeral length after graft insertion was the same as before the operation. After the operation, immobilisation in cast was applied for a period of six to 12 weeks, followed by physiotherapy for functional rehabilitation immediately after removal of the cast. No arm length difference was ascertained between the operated and non-operated upper limb. During follow-up after operation we did not observe any nerve injury or palsy in the area of the upper and lower extremities. In two cases there was superficial wound infection (one arm and leg) which resolved after two weeks of antibiotic treatment.

All our patients were skeletally mature at last follow-up. On clinical examination range of motion (ROM) of shoulder, elbow, knee and ankle joints was evaluated. Pain, limb ailment and shortening were also assessed. On radiological examination we looked for recurrence of cystic changes. We assessed the humeral bone axis status, graft remodelling and fibula restoration.

Table 1 Clinical details of 20 patients with benign lesions of humerus

Patient number	Age (years)	Solitary bone cyst	Aneurysmal bone cyst	Fibrous dysplasia	Side	Length of follow-up (years)	Pathological fracture	Curettage and bone grafting	Number of steroid injections	Involvement of growth plate	LLD (cm)	RTG ratio
1	11	x			Right	25	x		2			3
2	4		x		Right	25		x				4.2
3	8			x	Left	22		x		x	6	3.7
4	10	x			Right	25	x		2			5
5	7	x			Right	25	x		3			4.1
6	10	x			Left	24	x		2	x	4.5	3.9
7	9	x			Right	24	x		2			3.4
8	12			x	Right	23	x					4.8
9	11	x			Right	23	x		2			3.5
10	14	x			Right	23	x		2			4.7
11	6		x		Right	22		x	1			4.1
12	19	x			Right	22	x		2			3.4
13	27	x			Left	4	x		2			5.8
14	10		x		Left	21		x	1			3.2
15	16		x		Right	19		x				4.3
16	15		x		Left	18	x		2	x	2	3.6
17	19	x			Right	13		x				4.1
18	9		x		Right	22	x		2			4.4
19	5		x		Right	12		x				4.1
20	14	x			Right	11	x		2			3.8

LLD leg length discrepancy, RTG ratio radiographic ratio

Fig. 1 **a** Humeral solitary cyst. **b** Immediately after surgery. **c** Last follow-up 20 years after surgery

Results

Histopathological studies of resected humeral bone fragments revealed aneurysmal bone cyst in seven cases, solitary cyst (unicameral) in 11 cases and fibrous dysplasia in two cases. The mean follow-up period after the surgery was 19 years (range 4–25 years). No recurrence of the pathology was noted during the entire follow-up period after the operation. In follow-up examinations, none of the patients reported any pain in the affected limb—arm or leg. There was no limitation in the range of motion in either the shoulder or elbow joint nor of the knee or ankle joint. The normal mobility range in shoulder and elbow joints was regained one to four months after plaster cast removal. No difference in leg length was noted immediately after the surgery nor after regaining full mobility. Pathological fracture did not occur in any patient postoperatively. There was shortening of the humerus in three cases at the time of last follow-up examination measuring 2 cm, 4.5 cm and 6 cm respectively. Humeral shortening was observed only in those patients in whom the cyst was adjacent to the proximal growth plate of the humeral bone. Two cases had a valgus deformity of proximal humeral bone evident radiologically at the time of last follow-up measuring 10° and 15° respectively. Neither of these cases had functional impairment as a result of the valgus alignment of humeral bone and there were no cosmetic concerns. Neither limb shortening nor any axial disorders of the knee or tarsal joint were observed within the lower limb. All patients reported satisfaction after surgery, including three patients with arm shortening, during last follow-up (Figs. 1 and 2).

Periosteal reaction was noted ten to 14 days postoperatively on AP and lateral humeral radiographs. Bone healing between the ends of the humeral bone or the proximal epiphysis occurred three to four months after the operation. No graft atrophy was observed in any of the cases. Complete remodelling of the fibular graft in the proximal humerus occurred between two and four years. Where the

periosteum was excised with the cystic lesion the process of fibula graft incorporation was much slower.

At the donor site gradual restoration of the fibula occurred even in cases where a 20 cm graft was harvested. In six patients complete fibula rebuilding was observed, together with reconstruction of its trabecular structure and of the bone marrow canal (Fig. 3). All these patients were under 12 years of age at the time of surgery. In the remaining cases some degree of bone regeneration was found. The older the patients were during operation, the slower was the process of fibula restoration.

Discussion

There is a wide spectrum of treatment for benign cystic lesions of the humerus; in cases of pathological bone

Fig. 2 **a** Humeral aneurysmal bone cyst extended to the physis. **b** Ten months after surgery with periosteal reaction, growth plate is still open. **c** Last follow-up 22 years after surgery, humeral shortening of 6 cm

Fig. 3 Fibula restoration
20 years after taking bone graft

fracture within the cyst area, the simplest immobilisation technique can result in healing of the cyst. This observation prompted a new concept of treatment to be designed involving the application of autologous bone marrow or platelet growth factors [9, 12]. Use of platelet growth factors had been popularised by dental surgeons for the treatment of dental cysts [12]. A very well known method of treatment which has been widely accepted is a steroid injection into the bone cyst. The steroid agent is administered directly into the cystic area, taking advantage of its anti-inflammatory and anti-neoplastic effects. This technique of puncture with injection is simple and minimally invasive. However, this too may be associated with a high risk of failure [8–10]. Wright et al. presented a 42% success rate after steroid injection of simple bone cysts [11]. Another operative technique involves opening a cortical window in the wall of the cyst, debridement and impaction of osteogenic material. The success of these treatments is limited to the management of large bone lesions. At the extreme end of the treatment spectrum there are techniques consisting of radical cystectomy coupled with reconstruction of the bone defects. The application of microsurgical techniques and the use of a pedunculated fibular implant improves the healing time of the implant. This technique avoids necrosis and resorption of the incorporated graft and of the new osteogenic process; hence, it does not weaken. It remodels in a similar way to the normal bone and is superior to the non-vascularised bone graft [14, 15, 17]. However, the operative technique is demanding and time consuming, and it is not available in every centre. Additionally, anomalies of the vessels leading from the anterior tibial artery, which vascularise the proximal end of the fibula, makes this method even more difficult [17]. The autologous non-vascularised cortical graft provides excel-

lent structural bone support at the recipient side [17, 18]. The good blood supply of the recipient side in the proximal humerus allows revascularisation of the fibular graft. In the first few weeks immediately post-operatively the mechanical strength of the graft is reduced. The full mechanical properties return after six to 12 months [17, 18].

The choice of method of treatment is often difficult and depends on the experience of the surgeon and tradition of a given centre. A good therapeutic outcome requires patience and collaboration with the patient. The method outlined requires careful consideration when patients are being listed for surgery. The technique itself is relatively simple for an experienced surgeon but is based on subperiosteal dissection of both the pathological bone lesion and the graft. Bone stabilisation is obtained by means of graft impaction. At this stage of the operation, the dimensions of anastomosed elements are of key importance, while the goal is their stable connection whilst maintaining the length of the limb. No special fixation techniques for the fibular graft were used. The diameter of the bone marrow cavity of the humeral bone is large enough to allow insertion of the fibula ends into the canal, following slight adjustment. The fixation obtained was so stable that no graft loosening was observed in the follow-up observation. In the course of the healing process of the graft, neither fracture nor pseudarthrosis was found. No limb shortening was observed in our material immediately after the surgery, while 2–6 cm shortening was found in three patients during follow-up. In this group the cystic lesion was adjacent to the proximal humeral growth plate, and we now exclude patients with this cyst location from this operation, instead trying to treat them using less invasive methods. However, none of the patients were keen to have limb lengthening because they had excellent range of motion and full function in that arm. Similarly, the two patients with valgus alignment of the proximal humerus did not have any concerns regarding the limb appearance. We did not perform the operation immediately after pathological fracture of the humeral bone. The surgical intervention was applied only when the process of bone healing was complete (minimum six to nine months after pathological fracture). In cases of pathological fracture and a wide bone lesion, multicameral cyst lesion, with involvement and damage of the cortical layer, we undertook the risk of the surgery, taking into account possible problems with periosteum dissection or the necessity for total removal, associated with extended time for fibular graft healing.

We observed restoration of ROM in shoulder and elbow joint in all patients one to four months after plaster removal. The reasons for good functional results could be that the surgery did not involve a joint, before operation there was full ROM, patients were young and physical therapy was performed immediately after plaster removal.

Full incorporation of the fibula at the donor site was mainly observed in patients before the tenth to 12th years of life. This may be explained by the fact that in these children, the periosteum is thick, resistant and easily detachable from the bone, while in adolescents and young adults, the periosteum is thin and strongly bound with the bone, thus more easily damaged in the course of dissection. As a result the fibular regeneration process is either partial or absent.

The long-term clinical outcomes show that the results of this method are encouraging and that there is a low risk of associated complications. It should be underlined that patient's engagement, compliance and contribution in the rehabilitation process of the operated limb are key factors for therapeutic success. However, the general value of this method is unquestionable. In our opinion it is the method of choice, providing a great chance for very good results in treatment of large, multi-chamber benign bone lesions of the humeral bone with cortex layer destruction, where other types of treatment fail.

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