Aneurysmal bone cysts of the distal fibula in children

LONG-TERM RESULTS OF CURETTAGE AND RESECTION IN NINE PATIENTS

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We report the results of the treatment of nine children with an aneurysmal bone cyst of the distal fibula (seven cysts were juxtaphyseal, and two metaphyseal). The mean age of the children was 10 years and 3 months (7 years and 4 months to 12 years and 9 months). All had open physes. All cysts were active and in seven cases substituted and expanded the entire width of the bone (type-2 lesions). The mean longitudinal extension was 5.7 cm (3 to 10). The presenting symptoms were pain, swelling and pathological fracture. Moderate fibular shortening was evident in one patient.

In six patients curettage was performed, using phenol as adjuvant in three. Three with juxtaphyseal lesions underwent resection. A graft from the contralateral fibula (one case) and allografts (two cases) were positioned at the edge of the physis for reconstruction. The mean follow-up was 11.6 years (3.1 to 27.5). There was no recurrence.

At the final follow-up there was no significant difference in the American Orthopaedic Foot and Ankle Society scores (excellent/good in all cases) and in growth disturbance, alignment, stability and bone reconstitution, but in the resection group the number of operations, including removal of hardware, complications (two minor) and time of immobilisation/orthosis, were increased. Movement of the ankle was restricted in one patient.

The potential risks in the management of these lesions include recurrence, physeal injury, instability of the ankle and hardware and graft complications. Although resection is effective it should be reserved for aggressive or recurrent juxtaphyseal lesions.

Local surgery of neoplastic and pseudoneoplastic lesions of the distal fibula poses problems of the adequacy of resection and of reconstruction of the ankle.^{1,2} Particular difficulties arise in the management of aneurysmal bone cysts of the distal fibula in children. These lesions may replace an entire segment of the bone and impinge on the physis (juxtaphyseal locations). They may recur if not adequately treated. The therapeutic challenge, in conjunction with eradication of the lesion, is to preserve the physis, to maintain fibular length in order to prevent progressive valgus deformity of the ankle and to preserve stability of the ankle.³⁻⁵

The treatment of choice for aneurysmal bone cysts of the distal fibula has been curettage with bone grafting. Resection of the cysts, which has a lower rate of recurrence compared with curettage,^{6,7} has been reported in only a few cases^{2,3,6,8-13} for such lesions in children, and different techniques of reconstruction have been performed.

Our aim was to summarise our experience in the management of aneurysmal bone cysts arising in the distal fibula in children.

Patients and Methods

In a retrospective review of 97 patients with an aneurysmal bone cyst who had been treated between 1976 and 2004, the distal fibula was found to be involved in nine. There were five boys and four girls with a mean age of 10 years and 3 months (7 years and 4 months to 12 years and 9 months). The lesions were located on the left side in five and on the right side in four. The gender, age at the time of diagnosis, history of symptoms, radiological features, type of treatment and outcome were recorded and the details are summarised in Table I.

Radiological evaluation described the relationship of the cysts to the growth plate (juxtaphyseal or metaphyseal) and were classified according to the system of Campanacci, Capanna and Picci¹⁴ and Capanna et al¹⁵ based on the anatomical location.

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Table I. Clinical and radiological details of the nine patients

Case	Symptoms	Duration (mths)	Relation to growth plate	Longitudinal extension (cm)	Type ¹⁴	Staging ¹⁵	Fibular shortening ¹⁶
1	Pathological fracture	*	Metaphyseal (6 mm)	3.0	1	Active	-
2	Pain, swelling	3.0	Juxtaphyseal	3.5	3	Active	-
3	Swelling	3.0	Juxtaphyseal	4.5	2	Active	-
4	Pain, swelling [†]	7.0	Juxtaphyseal	6.0	2	Active	-
5	Swelling (after contusion) and pain on compression	4.0	Metaphyseal (5 mm)	3.5	2	Active	-
6	Swelling	12.0	Juxtaphyseal	9.0	2	Active	-
7	Pain, swelling	3.5	Juxtaphyseal	10.0	2	Active	Moderate (grade II)
8	Persistent pain following a sprain	5.0	Juxtaphyseal	4.5	2	Active	-
9	Pain, swelling	8.0	Juxtaphyseal	7.5	2	Active	-

* biopsy was performed after immobilisation in a cast for 40 days

† primarily treated in another hospital by corticosteroid injection

Table II. Details of the surgery performed

Case	Age at surgery (yrs + mths)	Treatment	Complications
1	7 + 4	Curettage	-
2	9	Curettage + phenol + homologous bone grafting	-
3	9 + 3	Curettage	-
4	10	Curettage + phenol + homologous bone grafting + Ostilit*	-
5	12 + 8	Curettage + homologous bone grafting	-
6	12 + 9	Curettage + phenol + homologous bone grafting + Ostilit*	-
7	8 + 3	Resection + homologous (cortical strut) grafting + intramedullary Kirschner (K)-wire	Valgus deviation of graft after removal of hardware (2 months after surgery) [†]
8	10 + 8	Resection + autogenous (cortical strut) grafting (taken from contralateral fibula) + intramedullary K-wire	-
9	12 + 6	Resection + homologous grafting (2 cortical struts) + 2 screws	Breakage of the proximal screw after 7 months

* tricalcium phosphate and hydroxyapatite (Stryker Howmedica, Osteonics, Newbury, United Kingdom)

† complete remodelling of valgus deformity at follow-up

There were five types of lesion with types 1 and 2 corresponding to central forms and types 3 to 5 to peripheral forms. The activity was graded by the staging system proposed by Capanna et al¹⁵ which involves three stages (aggressive, active and inactive). Alignment of the ankle was evaluated by measuring the tibiotalar angle. The relative levels of the distal growth plates of the tibia and fibula (fibular shortening according to Malhotra, Puri and Owen¹⁶) were evaluated to assess the influence of the lesions on physal growth.

Post-operative evaluation consisted of clinical and radiological monitoring for the extent of healing, recurrence, physal growth, alignment and functional outcome. Weight-bearing radiographs of the treated and of the contralateral ankles were performed for comparison. The American Orthopaedic Foot and Ankle Society (AOFAS) clinical rating system for the ankle and hindfoot¹⁷ was used for the evaluation of pain, function (activity limitations, support requirements, walking ability on uneven ground, maximum walking distance), stability, mobility and alignment.

Results

Pain and swelling were the two most common presenting symptoms. Seven patients complained of swelling which was painful in five and painless in two. Six complained of pain. In two patients there had been minor trauma and one had received a corticosteroid injection, based on the presumptive diagnosis of a simple bone cyst. One patient presented with a pathological fracture through the cyst. Excluding this case, the presenting symptoms had lasted for a mean of 5.6 months (3 to 12).

At presentation, none of the patients had abnormal alignment or limitation of movement of the ankle. In all, the cartilaginous growth plates were seen to be open. Lesions were in contact with the distal fibular physis in seven patients (juxtaphyseal lesions) and in two cases were at a distance of 5 mm and 6 mm, respectively, from the growth plate (metaphyseal lesions). The mean length was 5.7 cm (3 to 10) and the mean width was 2.7 cm (1.5 to 4.5).

Using the classification of Campanacci et al¹⁴ one case was type 1, seven were type 2 and one was type 3. All lesions were active, none being inactive or aggressive. Two

Fig. 1a

Fig. 1b

Fig. 2a

Fig. 2b

Case 6 – Anteroposterior radiographs showing a) a juxtaphyseal type-2 aneurysmal bone cyst (longitudinal extension, 9 cm), and b) the distal fibula 16.5 years after curettage with phenol and allogenic bone grafting mixed with tricalcium phosphate and hydroxyapatite granules.

Case 8 – Post-operative anteroposterior radiographs after seven weeks, of a) a juxtaphyseal lesion treated by resection and autograft taken from the contralateral fibula preserving bone continuity and b) 27.5 years after surgery.

patients had MRI and four had CT which showed double-density fluid levels in five. In none was there epiphyseal involvement with disruption of the growth plate. Thus no patient had significant varus or valgus tilt of the ankle on radiographs. There was moderate fibular shortening (distal fibular physis in line with distal tibial growth plate) and a slight increase in the tibiotalar angle in one juxtaphyseal lesion.

Various surgical procedures were performed (Table II) and all cases had histological confirmation of the diagnosis. In five an incisional biopsy preceded surgery and an intra-operative frozen-section biopsy was performed in two. Two patients underwent curettage without prior histological confirmation of the diagnosis and were followed up routinely. There were no secondary aneurysmal bone cysts.

Intralesional excision and curettage were performed in six patients (four juxtaphyseal and two metaphyseal) at a mean age of 10 years and 3 months (7 years and 4 months to 12 years and 9 months) (Fig. 1). These lesions had a mean length of 4.9 cm (3 to 9). In juxtaphyseal lesions the procedure was performed with care taken to preserve the growth plate. In three active and juxtaphyseal lesions, phenol was used as adjuvant in the proximal side of the physis. There were no complications after this procedure. In two cases treated by curettage there was no grafting. In four allogenic bone grafting (with an added fibular strut graft in one) was used. In two cases bone-graft substitutes (tricalcium phosphate and hydroxyapatite (HA) (Ostil; Stryker Howmedica Osteonics, Newbury, United Kingdom), were added to allogenic bone.¹⁸

Three patients with active and juxtaphyseal lesions underwent resection of the lesion. Their mean age was 10 years and 5 months (8 years and 3 months to 12 years and 6 months) and the mean length of these lesions was 7.3 cm (4.5 to 10). In all cases the distal cut was performed above the growth plate and resection was completed at the edge of the physis by blunt curettage.

In a girl aged 10 years and 8 months (Fig. 2), fibular continuity was restored by autogenous bone grafting. A cortical strut was taken from the diaphysis of the contralateral fibula, preserving bone continuity, and it was stabilised using an intramedullary wire.

In two cases allografts were placed at the edge of the physis after resection of large lesions, 10 cm and 7.5 cm long respectively. In one patient (case 7) an allogenic cortical graft stabilised by an intramedullary wire was used (Fig. 3). In another (case 9) two cortical strut allografts were used, one for bridging the fibula and one from the tibial meta-diaphysis to the bone above the distal fibular physis, fixed by two screws, for stability (Fig. 4). The proximal screw broke after about seven months and was removed. The two wires were removed two months after surgery.

A cast was applied post-operatively in all the patients for a mean of seven weeks (4 to 10) after curettage and 14 weeks (10 to 17) after resection. A rigid ankle-foot orthosis was used thereafter for a mean of four months (2 to 5) in the patients who had resection and when a cortical strut had been added to allogenic grafting.

Post-operative complications included breakage of a screw, and minor valgus deformity after removal of the

Case 7 – a) Radiograph showing a juxtaphyseal type-2 lesion with moderate fibular shortening and a slight increase in the tibiotalar angle, b) MR scan showing the relationship of the lesion to the distal fibular physis, c) post-operative radiograph after resection and allografting at the edge of the growth plate, and d) radiograph 8.5 years after surgery.

Case 9 – Post-operative anteroposterior radiographs after four months, a) of a juxtaphyseal lesion after resection and allografting with two cortical struts and b) at three years and one month after surgery.

intramedullary wire. The latter corrected completely with growth.

The mean follow-up was 11 years and 7 months (3 years and 1 month to 27 years and 6 months). All patients had closed growth plates at the final follow-up and there was no recurrence. In those who had curettage, radiological evalu-

ation of bone healing showed complete bone reconstitution in five and persistence of a static cystic area¹⁹ in one in which curettage without grafting had been performed. Of the three patients treated by resection, two showed bone reconstitution and fibular continuity and one had a heterogeneous area of bone bridging between the margins of the resection and the distal fibula and tibia.

There was no significant change in the tibiotalar angle and very minor changes in the relationship of the distal tibia and fibula, in comparison with the contralateral ankle (Table III). In cases in which follow-up was performed periodically until skeletal maturity, the fibular growth plates remained active after surgery. In those in which stigmata of the lesions were still evident radiologically, these appeared to be more proximal than the lesions themselves.

The functional results were excellent in eight patients with a mean AOFAS score of 99.1 (96 to 100/100) and good (78/100) in the patient who had been treated by curettage, phenol and grafting. One patient had occasional pain and instability and two had some discomfort when walking on uneven ground. The range of movement of the ankle was reduced by 20% in the patient in whom distal tibio-fibular fusion by two cortical struts has been undertaken.

Discussion

Aneurysmal bone cysts are expansile osteolytic lesions³ consisting of blood-filled spaces of variable size, separated by connective tissue septa containing trabeculae of bone or osteoid tissue and osteoclast giant cells.²⁰ In approximately 80% of cases they occur within the first two decades of life,

Table III. Details of the clinical and radiological outcome

Case	Follow-up (yrs + mths)	Clinical result*	Main clinical complaints	Bone reconstitution	Relationship of distal fibula to tibia†
1	10 + 6	100	-	Complete	Normal length
2	10	100	-	Complete	Fibular overgrowth (2 mm)
3	10	98	Difficulty on uneven ground	Static residual cyst	Normal length
4	14 + 3	99	Reduced maximum walking distance	Complete	Fibular shortening (3 mm)
5	4 + 2	100	-	Complete	Normal length
6	16 + 7	78	Occasional pain, subjective instability, difficulty on uneven ground	Complete	Normal length
7	8 + 6	100	-	Complete	Normal length
8	27 + 6	100	-	Complete	Normal length
9	3 + 1	96	Limited movement	Heterogeneous area of bone bridging	Fibular overgrowth (2 mm)

* American Orthopaedic Foot and Ankle Society clinical rating¹⁷

† measured on weight-bearing radiographs and compared with the contralateral ankle

arising typically in the metaphysis and the metadiaphysis¹⁴ of long tubular bones and in the posterior osseous elements or the vertebral bodies of the spine.^{3,21} The juxtaphyseal location has a relatively high incidence in patients who have not reached skeletal maturity, 20% according to Campanacci et al¹⁴ and 26% according to Rizzo et al.³ This fact may be explained by one of the proposed pathogenic mechanisms which is that aneurysmal bone cysts arise secondarily to an anomalous arteriovenous fistula leading to an alteration in local haemodynamics, with resulting increased venous pressure, augmented resorption of spongy bone and endosteal erosion of cortical bone.²² The initial development of the arteriovenous malformation may be caused by trauma (or repetitive microtrauma), or by the growth of a pre-existing lesion,²³ either benign or malignant, more commonly a giant-cell tumour, non-ossifying fibroma, chondroblastoma, osteoblastoma and osteosarcoma, or as in the case of juxtaphyseal locations, as a result of the intense neovascularisation drawn to the growth plate with increased potential for arteriovenous malformations.^{3,22,24}

The incidence of a distal fibular location varies between 1.5%¹⁴ and 7.1%¹¹ in different series. In a review of paediatric aneurysmal bone cysts reported in the literature,²⁵ a location in the fibula was found in 8.3% of patients, which is lower than that which we recorded (16.4%).

According to the radiological classification of Campanacci et al¹⁴ any type of aneurysmal bone cyst may occur at or near the growth plate, but in the distal fibula type 2 in which the lesion substitutes and expands an entire segment of bone, is the most common, like in other long thin bones such as the ulna, metacarpals and metatarsals, where the distance needed to involve the entire width of the bone is decreased.^{11,14} This type is reported to invade the growth plate^{3,4} more readily and to have, with type-1 lesions in which the cyst occupies the centre of the bone and the bone profile is intact or slightly expanded, a higher rate of recurrence than that of the peripheral forms (types 3 to 5).²⁶ The weakening of bone induced by these central lesions is more

likely to lead to the development of a pathological fracture,⁷ which is not usually seen in the more common off-centred presentation.^{11,21,26} We have recorded one case. Other clinical features of presentation are nonspecific pain, sometimes mistaken for the consequences of ankle sprain, and a bony swelling, given the propensity for expansion of a superficial bone.²¹

The most common differential diagnoses in children at this site are simple bone cysts, fibrous dysplasia, non-ossifying fibroma, chondromyxoid fibroma and osteoblastoma, but the differential diagnosis should include telangiectatic osteosarcoma and giant-cell tumour.²¹ The diagnosis can usually be suggested by a number of features characteristic of aneurysmal bone cyst, including the age of the patient, the location, clinical and radiological findings, but only pathological examination can provide diagnostic safety. We agree that a suspected aneurysmal bone cyst should not be treated without biopsy.^{19,24} Magnetic resonance imaging can be highly suggestive, but not diagnostic.^{19,21}

Juxtaphyseal aneurysmal bone cysts abutting the growth plate may lead to its damage, caused either by the growth of the cyst itself or by an iatrogenic injury.

Penetration of the cyst through the physeal plate has been described with a frequency varying between 23%^{4,14,27} and 27%²⁸ of juxtaphyseal lesions. Disruption of the growth plate may become clinically evident if lesions are left to their natural course.³ The only case of epiphyseal involvement from a juxtaphyseal lesion at the distal fibula which we were able to find in the literature was in a ten-year-old patient without signs of growth disturbances.²⁹ In the seven cases of juxtaphyseal lesions which we reported, none showed gross invasion of the growth plate and in none was there deformity of the ankle. However, moderate fibular shortening with a slight increase of the tibiotalar angle was present in one eight-year-old child.

Treatment. Some authors^{7,20} have stressed the importance of physeal preservation in the treatment of aneurysmal

bone cysts, especially in young children even if lesions are likely to recur. By contrast, when a fibular location is considered, the expression 'expendable bone' is sometimes used,^{12,20,26,30} justifying more aggressive treatment. However, when growth of the lower fibular physis is arrested for any reason, a valgus deformity of the ankle may develop and the degree of valgus depends on how many years of growth are left.³¹ The age over which the risk of secondary valgus deformity is insignificant, seems to be between 10 and 12 years.³¹

Traditional treatment has been curettage and bone grafting.^{6,14,21,26} For juxtaphyseal locations, especially in the case of large lesions, physeal damage may be inflicted by attempts at thorough curettage.^{8,9} When curettage and bone grafting have been performed with care taken to preserve the growth plate³ there have been no cases of major iatrogenic physeal disturbance,^{3,4} despite the low rate of recurrence reported (20%).³ Cases of juxtaphyseal lesions in the distal fibula successfully treated in this way were reported by Rizzo et al³ with only minor complications and no deformity or recurrence. In our experience, there were no cases of recurrence or growth disturbance after curettage. The clinical results were excellent or good in all patients. In three we used phenol as an adjuvant to 'sterilise' the bony wall of the cavity in the proximal side to avoid physeal injury³² and no complication was recorded.

Alternative treatments have been considered to reduce the rate of recurrence reported after curettage and bone grafting.⁶ In a recent review of the literature of series including 20 or more cases, curettage and bone grafting was associated with an average recurrence rate of 31%.²⁴ Cryosurgery, embolisation and radiotherapy are inadequate options for these locations in children because of the complications which include arrest of the growth-plate, skin necrosis, post-operative fracture, sarcomatous degeneration, joint stiffness, and osteonecrosis.^{6,28}

Resection has been considered by several authors as the treatment of choice for aneurysmal bone cysts^{5-7,11} because of its lower rate of recurrence compared with curettage and bone grafting.^{7,11} Recurrence of distal fibular lesions after curettage and bone grafting have been reported for which only resection was successful.⁸⁻¹⁰ Conversely, no recurrence after resection at this location has been described in the literature or in the three cases which we reported. An increased morbidity, however, must be considered.^{8,10,19,30}

In children with open physes the main problems are at the level of the distal cut with the risk of iatrogenic physeal damage or incomplete resection, and the subsequent reconstruction.⁵ For juxtaphyseal lesions the level of resection may be different, according to the age of the patient. In those older than 10 to 12 years,³¹ inclusion of the growth plate in the resection may provide safer margins, as reported by some authors.^{2,8,9} Nevertheless, we advise resection above the growth plate even in patients older than ten years. For younger children, the lesion should be resected subperiosteally to the edge of the physis⁶ and the

distal cut should be a few millimetres above the physis.⁵ Resection adjacent to the growth plate should be completed by blunt curettage.⁷ This procedure combines the advantages of the lower rate of recurrence of resection and the lower risk of physeal injury. Radiological follow-up of the three cases treated by this procedure has shown no secondary growth disturbance.

For lesions arising at a distance from the epiphyseal plate, resection does not pose any particular problems of physeal injury or reconstruction.^{10,13}

Reconstruction. The main reconstructive problems after resection are restoration of fibular length and instability of the ankle. In young children fibular length should be restored to allow the thrust of growth contributed by the proximal physis to be transmitted to the lateral malleolus. This prevents progressive valgus deformity of the ankle.³¹ In order to encourage fibular regeneration, it is essential to leave intact as much of the periosteal sleeve as possible, when subperiosteal resection is feasible.³¹

Autografts taken from the contralateral (case 8) or ipsilateral fibula^{2,6,12,13} may be used. In the latter option, periosteum at the site of the graft is sutured,² and stabilisation of the fibula is performed whilst waiting for bone regeneration to avoid malleolar ascent.^{11,13} Rizzo et al⁵ reported a technique of ipsilateral slide grafting after resection. The fibula is exposed proximal to the defect and bisected longitudinally, and the lateral half is transposed distally.

As an alternative, allografts may be used, particularly after resection of lesions with significant longitudinal extension. Rigid fixation may be used for diaphyseal reconstruction.¹⁰ To our knowledge, we have reported the first two cases of the use of allografts for juxtaphyseal lesions in the distal fibula. Allogenic grafting avoids morbidity at the donor site. An intramedullary wire was used to stabilise the graft at the edge of the growth plate in one case, while in the other the graft was simply wedged between the margins of the remaining bone. In very young children, it has been advised^{7,20} to position the graft into the periosteal bed held away from the growth plate with small-calibre pins, in order to reduce the risk of epiphysiodesis.⁷

Reconstructive procedures should also take into account potential instability of the ankle caused by division of ligaments of the ankle syndesmosis³³ in the case of resection of large lesions.²⁴ Stabilisation of the tibiofibular mortice is required in these cases.^{10,11,34} In our series after resection of a large lesion (case 9) distal tibiofibular fusion by cortical struts was performed, with some subsequent limitation of movement of the ankle.

However, in cases in which resection of the entire distal fibula is performed, instability of the ankle becomes the main concern.³⁵ Although it has been reported that resection without bony reconstruction did not cause instability,^{8,9,35} reconstitution of both the bony support and lateral ligaments is advisable. Reconstruction using bone or bone-and-ligament allografts^{10,34} or using the proximal fibula, rotated and placed distally, has been proposed.¹

In conclusion, when selecting the best procedure for the treatment of aneurysmal bone cysts arising in the distal fibula in children, the type and activity of the cyst, its longitudinal extension and relationship with the growth plate, and the age of the patient must be considered.¹ In active juxtaphyseal lesions, particularly in children younger than ten years of age, curettage should be performed, taking care to preserve the growth plate. Resection is effective, but care must be taken to avoid iatrogenic physeal injury, instability and hardware/graft complications and it should be reserved for aggressive or recurrent juxtaphyseal lesions.

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