CASE REPORT

SUCCESSFUL MANAGEMENT OF A RECURRENT JUXTAPHYSEAL ANEURYSMAL BONE CYST USING SCLEROTHERAPY

Vidyadhar Telang[1], Praveen Kumar Sinha[1], Siddharth Mahesh Shah[2]
[1] Dhanvantari Hospital, Mumbai, Maharashtra, India
[2] Breach Candy Hospital and Research Centre, Mumbai, Maharashtra, India

Correspondence to: Praveen Kumar Sinha (drpraveenksinha@gmail.com)

DOI: 10.5455/ijmsph.2014.280320142 Received Date: 19.03.2014 Accepted Date: 28.05.2014

ABSTRACT
Aneurysmal bone cysts (ABC) are commonly managed by localized excision (curettage/ extended curettage) with or without bone grafting or sclerotherapy. ABCs involving the lower end of radius are relatively uncommon. Here we present a case of an aggressive, juxtaphyseal ABC involving the lower end of radius in a 6 year old boy that failed treatment with localized excision and bone grafting twice. Finally, the lesion was successfully managed with a single session of sclerotherapy using 3% polidocanol. There was evidence of progressive opacification of the cyst on follow- up x- rays with no evidence of recurrence. At present, the child is over 2 years' follow- up since sclerotherapy without recurrence. However, the growth of lower end of radius is stunted due to physeal damage either due to the aggressive nature of the lesion or previous surgeries. Due to continued growth of the lower end of ulna, the child has radial deviation of the wrist. The parents have been counselled about a likely need for corrective surgery subsequently to manage the deformity.
Key Words: Aneurysmal Bone Cyst; Distal Radius Tumour; Sclerotherapy

Introduction

Aneurysmal bone cyst (ABC) is a benign, locally aggressive lesion of the skeletal system. It is commonly seen in the first two decades of life and is metaphyseal in location. While it commonly occurs in the femur, tibia or humerus; radius is a relatively uncommon site. Various treatment modalities for ABC include intralesional curettage, extended curettage (with high speed burr), curettage plus autologous bone grafting/ allografting/ cement packing, sclerotherapy, curopsy (biopsy with intention to cure)[1] or denosumab therapy[2]. Intralesional curettage plus bone grafting is considered a standard method treatment for ABC with an associated risk of recurrence between 15% and 20%.[3,4] While primary ABCs are more common, they can also occur secondary to other lesions like osteoblastoma, giant cell tumour of the bone, chondroblastoma or fibrous dysplasia. We report an interesting case of primary ABC involving the lower end of radius in a 6 year old male child that recurred after multiple surgeries but healed after a single session of sclerotherapy.

Case Report

A 6 year old boy presented to another centre with a fracture of the lower end of right radius after trivial trauma. Initial x-rays did not raise any suspicion of a pathological fracture and it was managed with closed reduction and above elbow cast. Though the fracture healed at 1 month, the child had a progressively increasing swelling and deformity above the wrist. X-Rays at that time revealed an eccentric, expansile and lytic lesion involving the lower end of radius. An MR showed a well-defined expansile, multiseptate metaphyseal lesion in the lower end of radius with fluid-fluid levels suggestive of an ABC. With that information at hand, a 'J' needle biopsy was undertaken and the histopathological diagnosis of ABC was confirmed (3 months after the fracture). With the tissue diagnosis available, the child was posted for intralesional curettage plus allografting (4 months after the fracture). At 2 months follow up x-ray after the index surgery, there was evidence of recurrence in the form of increase in size of the lesion and new radiolucencies which were rapidly progressive (Figure 1). Clinically, the child presented with a progressively increasing swelling above the wrist and radial deviation of the hand. Recurrence of ABC was managed with extended curettage with a high speed burr. Following the curettage, only a thin shell of ballooned out bone remained and was structurally incompetent. Hence a segmental lower end radius allograft was placed within the shell (7 months following the fracture, Figure 2). The physis was left undisturbed. At 5 months post- operative follow- up, there was no radiological evidence of recurrence and the allograft showed signs of consolidation (Figure 3). Though clinically, there was no increase in size of the swelling,

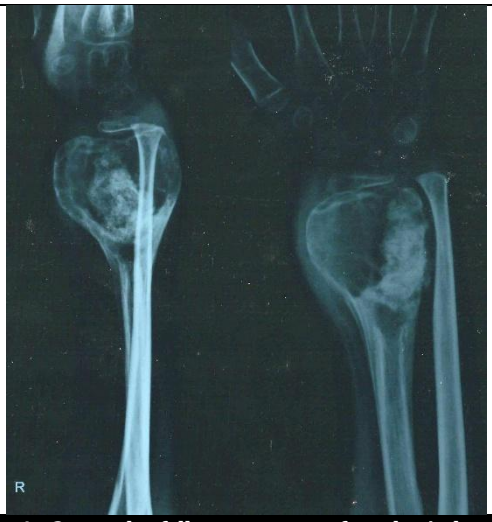

Figure-1: At 2 months follow up x-ray after the index surgery, there was evidence of recurrence in the form of increase in size of the lesion and new radiolucencies which were rapidly progressive

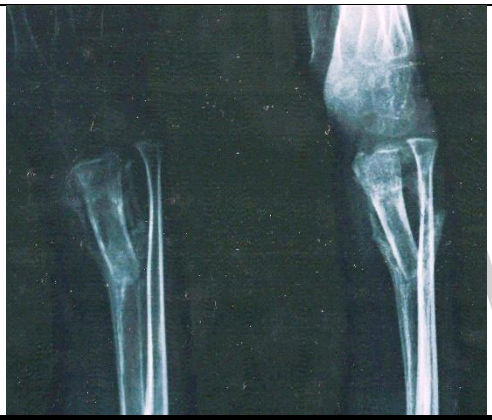

Figure-2: Segmental lower end radius allograft was placed within the shell (7 months following the fracture)

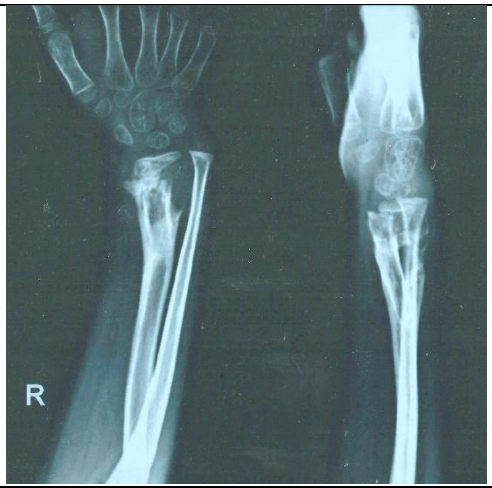

Figure-3: At 5 months post- operative follow- up: No evidence of recurrence and the allograft showed signs of consolidation

the radial deviation did not improve significantly. After about a year following the last surgery, the swelling started increasing in size. Radiologically there was increase in size of the lesion with newer radiolucencies and resorption of the bone graft (Figure 4).

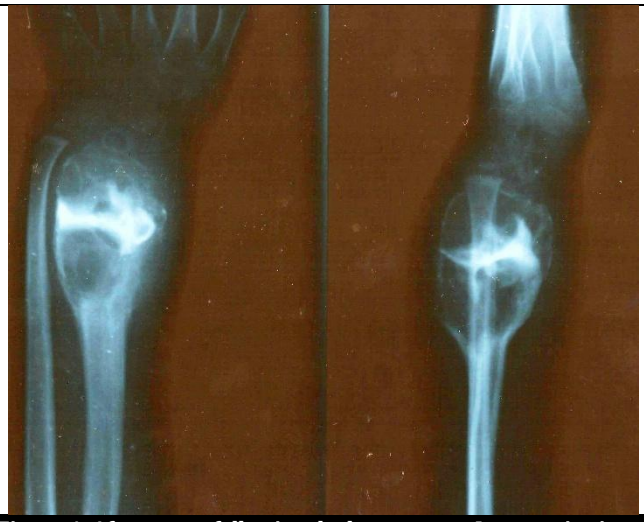

Figure-4: After a year following the last surgery: Increase in size of the lesion with newer radiolucencies and resorption of the bone graft

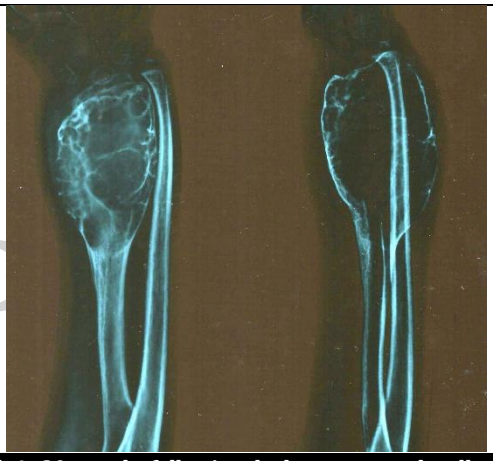

Figure-5: At 20 months following the last surgery, the allograft had completely resorbed

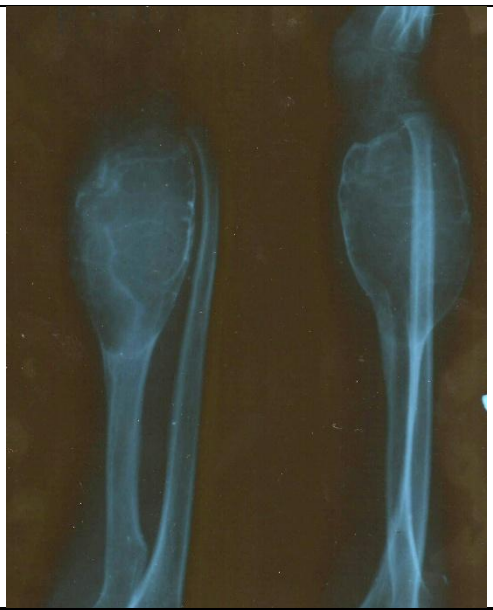

Figure-6: X- Ray just prior to injection of the sclerosant (24 months after the last surgery)

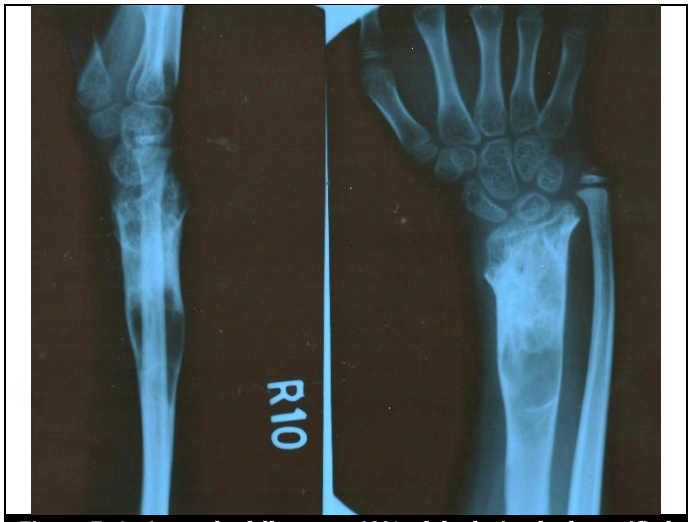

Figure-7: At 6 months follow- up, 60% of the lesion had opacified with no evidence of recurrence or increase in size of the lesion

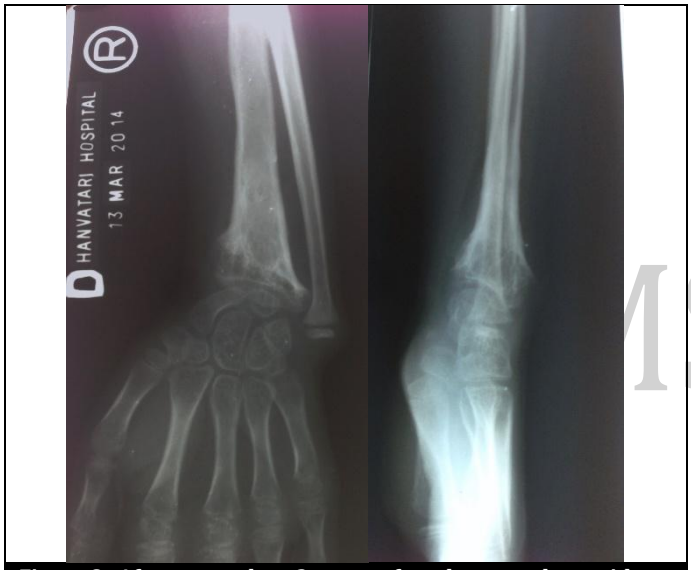

Figure-8: After more than 2 years after the procedure with no clinical signs or radiological evidence of recurrence

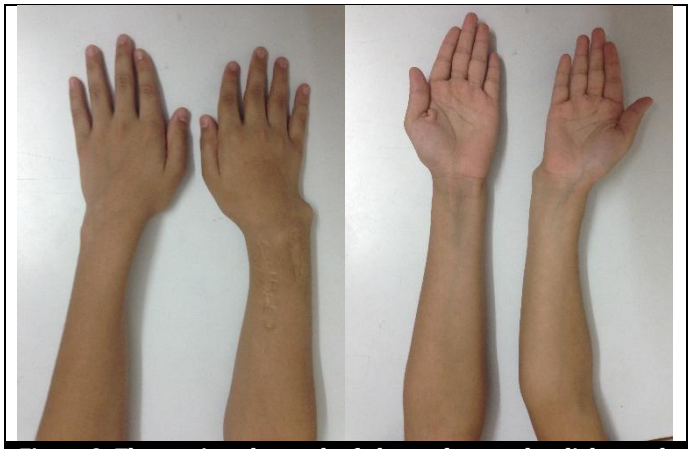

Figure-9: The continued growth of ulna and stunted radial growth has resulted in radial deviation of the wrist and the hand

Increase in the swelling and resorption of the bone graft progressed rapidly. At 20 months after the last surgery, the allograft had completely resorbed (Figure 5).

Figure-10: Full range of motion at wrist

It was evident that the ABC was aggressive in nature. It was decided to perform an en bloc resection with live vascularized fibular grafting (LVFG). At this point the patient had come to us for opinion. After reviewing the case details thoroughly, we were of the opinion that an alternative management must be offered. We felt that LVFG would be too morbid for the young child who was 8 years old by then. Moreover, the available literature on LVFG was too scarce to justify the treatment. Sclerotherapy was the other alternative that we could think of when curettage with bone grafting had failed. Varshney et al have reported lower recurrence rates, lower incidence of complications and better functional outcomes after sclerotherapy as compared to intralesional excision.[3] The use of an alcoholic solution of zein (EthiblocTM, Ethicon, Somerville, NJ) has been extensively reported for ABC. However, there were concerns regarding high failure and recurrence rates with this agent.[5]. Varshney et al.[3] and Rastogi et al.[6] have reported favourable results with 3% polidocanol (hydroxypolyethoxydodecan) for sclerotherapy in ABC.

Figure 6 shows the x- ray of the patient just prior to injection of the sclerosant (24 months after the last surgery). Though there is some evidence of presence of a physis in the x- ray, physeal damage or premature fusion could not be ruled out. After accounting for the magnification, the size of the lesion was determined to be

6.5 cm X 3.5 cm X 3.5 cm. If 1 mL of the sclerosant was to be injected for every 1mm3 volume of the lesion, the calculated volume of the sclerosant would way exceed the permissible limit (5 mL) of sclerosant injection. The procedure was carried out under GA and image intensifier (IITV) guidance. A 16 gauge needle was introduced into the ABC and its contents were aspirated. Subsequently, 3% polidocanol (Inj. Asklerol, Samarth Pharma, Mumbai, India) was injected slowly into the cyst until the maximum limit of 5 mL was reached. After injection of the sclerosant 1mL normal saline was injected and the needle was plugged to prevent backflow for one minute. Subsequently, the needle was removed. The child tolerated the procedure well. A splint was applied for 3 weeks following the procedure. No local adverse tissue reactions were observed in the post-operative period. The patient as followed- up at 1, 3, 6 and 12 months post- operatively and yearly thereafter. X-rays were obtained at every follow- up visit. There was evidence of progressive opacification of the lesion on follow- up x- rays. At 6 months follow- up, 60% of the lesion had opacified with no evidence of recurrence or increase in size of the lesion (Figure 7). Presently, the child is more than 2 years after the procedure with no clinical signs or radiological evidence of recurrence (Figure 8). However, there is growth arrest of the lower end of radius. It is likely that the physis at the lower end of radius was damaged due to the aggressive nature of the cyst or due to previous surgeries or has undergone premature fusion. The same was recognized prior to sclerotherapy and the prognosis regarding the same was conveyed to the parents. The continued growth of ulna and stunted radial growth has resulted in radial deviation of the wrist and the hand (Figures 9 & 10). The parents have been conveyed regarding a likely need for corrective surgery for the deformity at a later stage.

Discussion

The role of sclerotherapy in primary management of ABC is well established.[6-8] Authors have reported that sclerotherapy resulted in similar recurrence rates as compared to localized excision but with lesser morbidity.[3] We have presented a case of a highly aggressive ABC involving the lower end radius, an uncommon site for ABC that failed to resolve despite localized excisions twice but responded well to a single session of localized sclerotherapy. We felt that the exhaustion of surgical options involving localized excision was a real challenge in this case. The recurrence of ABC perhaps meant inadequate clearance of the lesion during previous surgeries. We felt that the best option available to us was injection of a sclerosant that would act by damaging the endothelium and initiating a coagulation cascade resulting in thrombosis.[7] However, use of sclerotherapy in management of recurrence of ABC has not been well reported. Hence, we have presented our experience with the same.

As mentioned previously, our choice of sclerosant was 3% polidocanol as compared to EthiblocTM due to lesser incidence of allergic, inflammatory and local reactions with the former.[9] Moreover, the reported success of polidocanol in patients with venous malformation was greater than EthiblocTM and ethanol (90% versus 65%- EthiblocTM and74%- ethanol).[10] In retrospect, we feel that it might have been better to offer sclerotherapy as the first option in this case as favourable outcomes have been reported with sclerotherapy in children. In a review of 29 children with ABC that were managed with sclerotherapy, Lambot- Juhan et al concluded that sclerotherapy was an effective treatment of ABC in them.[8] Moreover, it is known that juxtaphyseal ABCs have a high recurrence rate in skeletally immature patients.[11] Similarly, they also have a higher incidence of subsequent growth arrest and skeletal deformities.[12] Both these characteristics of recurrence as well as growth arrest were observed in our case.

Conclusion

We understand that it is difficult to draw conclusions on the basis of experience with a single case. But in light of the available literature and our own experience, we now offer sclerotherapy as the first treatment to paediatric patients with ABCs.

References

1. Reddy KI, Sinnaeve F, Gaston CL, Grimer RJ, Carter SR. Aneurysmal Bone Cysts: Do Simple Treatments Work? Clin Orthop Relat Res. 2014 Feb 15. [Epub ahead of print]
2. Pauli C, Fuchs B, Pfirrmann C, Bridge JA, Hofer S, Bode B. Response of an aggressive periosteal aneurysmal bone cyst (ABC) of the radius to denosumab therapy. World J Surg Oncol 2014;12:17.
3. Varshney MK, Rastogi S, Khan SA, Trikha V. Is sclerotherapy better than intralesional excision for treating aneurysmal bone cysts? Clin Orthop Relat Res 2010;468:1649-59.
4. Mankin HJ, Hornicek FJ, Ortiz-Cruz E, Villafuerte J, Gebhardt MC. Aneurysmal bone cyst: a review of 150 patients. J Clin Oncol 2005; 23: 6756–62.
5. Topouchian V, Mazda K, Hamze B, Laredo JD, Pennecot GF. Aneurysmal bone cysts in children: complications of fibrosing agent injection. Radiology 2004; 232: 522–6.
6. Rastogi S, Varshney MK, Trikha V, Khan SA, Choudhury B, Safaya R. Treatment of aneurysmal bone cyst with percutaneous sclerotherapy using polidocanol: a review of 72 cases with long term follow-up. J Bone Joint Surg Br 2006;88:1212–6.

7. Brosjo O, Pechon P, Hesla A, Tsagozis P, Bauer H. Sclerotherapy with polidocanol for treatment of aneurysmal bone cysts. Acta Orthop 2013;84:502-5.
8. Lambot-Juhan K, Pannier S, Grevent D, Pejin Z, Breton S, Berteloot L, et al. Primary aneurysmal bone cysts in children: percutaneous sclerotherapy with absolute alcohol and proposal of a vascular classification. Pediatr Radiol 2012;42:599-605.
9. Pollak J, White RI Jr. Peripheral vascular malformations. In: Kandarpa K (Ed). Peripheral vascular interventions. Philadelphia PA: Lippincott Williams & Wilkins; 2008: 497- 513.
10. van der Vleuten CJ, Kater A, Wijnen MH, Schultze Kool LJ, Rovers MM. Effectiveness of Sclerotherapy, Surgery, and Laser Therapy in Patients With Venous Malformations: A Systematic Review. Cardiovasc Intervent Radiol 2013 Nov 7. [Epub ahead of print]
11. Lin PP, Brown C, Raymond AK, Deavers MT, Yasko AW. Aneurysmal bone cysts recur at juxtaphyseal locations in skeletally immature patients. Clin Orthop Relat Res 2008;466:722-8.
12. Capanna R, Springfield DS, Biagini R, Ruggieri P, Giunti A. Juxtaepiphyseal aneurysmal bone cyst. Skeletal Radiol 1985;13:21-5.

Cite this article as: Telang V, Sinha PK, Shah SM. Successful management of a recurrent juxtaphyseal aneurysmal bone cyst using sclerotherapy. Int J Med Sci Public Health 2014;3:774-778.
Source of Support: Nil
Conflict of interest: None declared