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Outcome of aneurysmal bone cysts treated by extended curettage, cryosurgery and bone grafting

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Abstract

Aneurysmal bone cyst has a variable radiological appearance and should be considered in the differential diagnosis of any uni-focal or multi-focal radiolucent lesion. No role for conservative management as it is a surgical problem. ABC in the extremities can be managed by curettage and different kind of bone grafts. Recurrence rate is high especially in a young age with open growth plates. The use of liquid nitrogen as an adjuvant measure after extended curettage decrease tumor recurrence rate.

Patients and Methods

A case series of 25 patients with aneurysmal bone cyst involving different anatomic locations in the skeleton who were evaluated and staged according to Enneking *et al.* system as 20 active benign lesions and 5 aggressive benign lesions. Extended curettage was achieved in all the patients followed by application of liquid nitrogen for 2 cycles and lastly reconstruction of the cavity by bone graft. The mean age at surgery was 14. 7 years at operation (3–35 years). The average follow-up was 48 months (range, 24-72 months).

Results

The Musculoskeletal tumor Society (MTS) score described by Enneking *et al.* was used to assess functional outcome. At least follow-up the functional score ranges from 70% to 94%, with an average of 86%. One case developed local recurrence and managed by second operation. 2 cases developed superficial post-operative wound infection and treated conservatively.

Conclusion

Extended curettage of aneurysmal bone cyst with adjuvant cryotherapy had similar results to those of marginal resection and that no major bony reconstruction was required. We recommend the use of cryotherapy as an adjuvant to the surgical treatment of aneurysmal bone cysts. It provides local tumor control. Combination with bone grafting achieved consolidation of the lesion in all our patients with no major complications.

Keywords: Aneurysmal Bone, Extended Curettage, Cryosurgery, Bone Grafting

Introduction

Aneurysmal bone cyst of bone is a benign tumor like lesion. Although benign, the ABC can be a rapidly growing and destructive bone lesion. Malignant transformation does not occur [1-9].

The ABC may occur in any bone in the body and at any age and the ratio of female to male is 2:1.

The primary lesion is found mostly in the second decade of life (85% presented before 20 years of age) [7].

Recurrence rate is variable (5–24%): depending on the morphologic types, with the central types had a recurrence rate of 24%, whereas the peripheral forms the recurrence rate was only 5% [7, 8, 10].

The treatment of choice is curettage and autogenous bone grafting. Other lines of treatment are radiotherapy, embolization, marginal or even wide resections [7, 10, 11, 12]. Adjuvant therapy has been described with phenol liquid nitrogen and bone cement.

The technique entails curettage and burr drilling of the tumor cavity, instillation of liquid nitrogen and reconstruction of the cavity using bone grafting. Cryosurgery has achieved good local control of aggressive benign tumors.

All patients with contained cavity who met the criteria for cryosurgical ablation were treated in the same way. All patients in the current study were subsequently followed for more than two years (24 to 72 months; median, 48 months). The surgical concept and technique of liquid nitrogen Cryo ablation, together with all related complications and the rate of local tumor control all are presented.

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Patients and Methods

Twenty five patients with aneurismal bone cyst at different skeletal locations were operated and followed up between January 2008 and January 2015 at our institution. There were 16 females (64%) and 9 males (36%) with a mean age of 14. 7 years at operation (7 – 35 years). The right side was affected in 13 patients (52%) and the left side in 12 patients (48%). Pain and swelling were the presenting symptoms in all the cases. All the patients were presented with no previous surgical intervention. Except two cases no. (1, 3) The average follow-up was 48 months (range, 24-72 months).

All the lesions were staged by standard plain radiographs, computed tomography and/ or magnetic resonance imaging.

No pre-operative biopsy was done as the radiological diagnosis was evident; we confirm the diagnosis after the curettage of the lesions through histopathological examination of the curetted tissues in all the cases.

According to the staging system of Enneking *et al* [1-5]. The lesions were 20 active benign (80%) (Grade 2) and 5 aggressive benign (20%) (Grade 3).

Neither chemotherapy nor local radiation was given to any patient either pre-operatively or postoperatively.

Operative technique

The surgical technique consists of three separate stages:

Stage 1 (tumor excision)

After exposure of the involved bone and soft tissues, a cortical window the size of the longest longitudinal dimension of the tumor is made, ensuring that it is large enough to expose the entire tumor. It has to be elliptical with its axis parallel to the long axis of the bone to reduce the stress riser effect.

All gross tumors were removed with hand curettes (Fig. 1 B). High- speed burr drilling (Resectional curettage) was used for the removal of the inner reactive shell of the tumor cavity (Fig. 1 C).

Stage 2 (Liquid nitrogen application)

For cryotherapy before introduction of liquid nitrogen, bone perforations were identified and sealed using gel foam. The surrounding skin, soft tissues and neurovascular bundles were protected and shielded using gel foam and gauze soaked with saline to prevent extravasations of liquid nitrogen and thus protect the adjacent neurovascular structures and the skin. Large skin flaps were retracted to protect them from any possible spillage of the liquid nitrogen (Fig, 1 D, E and F). The direct pour technique as described by Marcove *et al.* was performed in all the cases. Liquid nitrogen (-196 °C) was poured through a stainless steel funnel into the tumor cavity (Fig, 1 D, E, F and G). The surrounding soft tissues were irrigated with warm normal saline to avoid thermal injury. Two freeze and thaw cycles were administered, each cycle lasting 1 to 2 min and spontaneous thaw was allowed to occur for 3 to 5 min. after evaporation, the cavity was irrigated with normal saline and good hemostasis was ensured.

Stage 3 (Reconstruction and application of the graft)

Reconstruction of the skeletal defect after tumor curettage and liquid nitrogen application by harvesting of the cancellous bone graft from iliac crest in 25 cases to fill the cavity. Autogenous fibular strut graft was applied to add structural support in 6 cases and combined cancellous iliac bone graft and fibular strut graft in 6 cases. Added internal plate fixation was applied in the 22 cases at proximal femur (Fig, 2 A, B and C).

A protective brace and/ or cast were used till graft incorporation and cyst ossification in 3 cases.

All patients had a clinical and radiological evaluation at monthly interval for the first six months then every two months for two years (Fig, 2 A, B and C). The aim of the early follow-up is to detect local recurrence. After that the patients were followed every 6 months.

Table 1: demographic data of the 25 patients with aneurismal bone cyst

Case number	Sex	Age (years)	Location	Graft used	Follow up (months)	Union (months)	Complica-tions	Results
1	Male	10	Proximal femur	Cancellous iliac crest &Fibular strut graft	72	8	Recurrence &Re-grafting	70%
2	female	14	Distal radius	Cancellous iliac crest	27	6	-----	90%
3	female	12	Distal femur	Corticocancellous iliac crest	33	6	Recurrence &Re-grafting	70%
4	male	34	Distal femur	Corticocancellous iliac crest	32	5	-----	88%
5	female	10	Proximal femur	Corticocancellous iliac crest &Fibular strut graft	58	9	-----	80%
6	male	6	Proximal femur	Corticocancellous iliac crest &Fibular strut graft	55	10	-----	78%
7	male	16	Proximal tibia	Corticocancellous iliac crest	34	4	-----	70%
8	male	12	Proximal tibia	Corticocancellous iliac crest	28	14	-----	92%
9	female	19	Distal femur	Corticocancellous iliac crest	60	6	-----	86%
10	male	21	Proximal humerus	Corticocancellous iliac crest	36	6	-----	94%
11	female	12	Proximal humerus	Corticocancellous iliac crest	30	5	-----	88%
12	male	14	Proximal tibia	Corticocancellous iliac crest	48	5	Superficial infection	90%
13	female	12	Proximal femur	Cancellous iliac crest & Fibular strut graft	72	10	-----	92%
14	female	11	Distal radius	Cancellous iliac crest	39	6	-----	82%
15	female	9	Proximal	Corticocancellous iliac crest	37	5	-----	84%

			humerus					
16	female	18	Proximal femur	Cancellous iliac crest & fibular strut graft	24	8	-----	88%
17	female	22	Distal femur	Corticocancellous iliac crest	50	6	Superficial infection	86%
18	female	12	Proximal tibia	Corticocancellous iliac crest	34	6	-----	92%
19	female	14	Proximal ulna	Cancellous iliac crest & fibular strut graft	36	7	-----	84%
20	male	17	Distal femur	Corticocancellous iliac crest	26	6		90%
21	female	20	Distal femur	Corticocancellous iliac crest	48	5		76%
22	Male	12	Proximal humerus	Corticocancellous iliac crest	66	6		94%
23	female	10	Proximal femur	cancellous iliac crest & fibular strut graft	72	8		86%
24	female	14	Proximal tibia	Corticocancellous iliac crest	72	4		90%
25	female	15	Proximal tibia	Corticocancellous iliac crest	72	4		88%

Radiographic evaluation

A system developed by Heiple *et al* [13]. For bone graft incorporation is used for radiologic assessment depending on the parameters of graft union and appearance of the graft with assessments of any evidence of fibular graft/cancellous graft restoration whether partial or complete or the graft is largely intact or reorganized completely.

Functional evaluation

The patients were evaluated with regard to function by using the system proposed by Enneking *et al.* and modified by the Musculoskeletal Tumor society [14, 15].

Results

Radiologic results: Graft union and cyst healing was achieved in all the patients. Partial graft resorption happened in one patient at the proximal femur with cyst recurrence. Graft union and cyst healing was achieved at an average of 5.7 months (4-14 months).

Functional results: At the end of the follow-up period, all the patients were alive and free from the diseases. Using the standard system of the musculoskeletal tumor society, the functional score at the final follow-up ranges from 70% to 94%, with an average of 86% [14, 15].

Complications

Graft resorption and tumor recurrence at 8 months post-operatively in one case at the proximal femur with growth plate affection by a picture like Perth's disease and was managed by re-grafting after extended curettage and healing and remodeling occurred within 6 months later on. Two cases developed superficial post-operative wound infection and managed by daily dressing and antibiotics and resolved within days. One case developed nerve palsy and recovered after 5 months post-operatively (posterior interosseus nerve in the proximal radius lesion).



(a) After making the bony windows. The blood filled cavity.



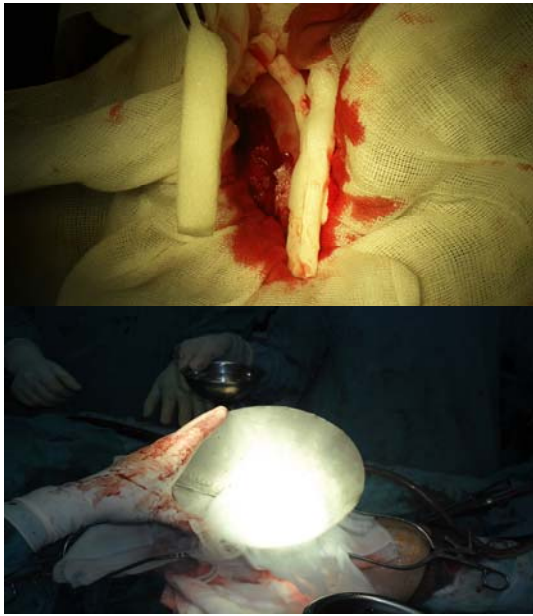
(b) Curettage of the cavity by standard curette.



(c) Extended curettage by burr.



(d) The clean cavity after curettage.



(e & f) Application of liquid nitrogen.

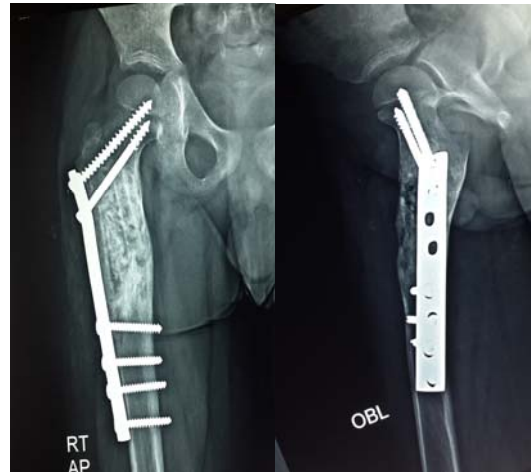


(G) Application of the fibular strut graft.

Fig. 1: Case (1): male patient 10 years old.



(a) Pre-operative x-rays antero-posterior-lateral.



(b) x-rays antero-posterior- lateral Three months post-operative



(c) x-rays antero-posterior- lateral Three years post-operative with complete graft incorporation and remodeling.



(d) X-rays antero-posterior- lateral Three years post-operative with complete graft incorporation and Remodeling after removal of plate and screws.

Fig 2: X-rays of the above case.

Discussion

In our prospective clinical study, 25 patients with aneurismal bone cyst of bone were treated using extended curettage, cryotherapy and bone graft with or without internal fixation. At a mean follow-up of 48 months, all the patients were alive and free from any signs of local or systemic recurrence, with good functional outcome. Our results are comparable with the published results of these techniques.

Recurrence rate after curettage of aneurismal bone cyst is variable and reconstruction of the bone defect created after curettage is a major problem.

The ideal reconstruction should have biological affinity, resistance to infection, sufficient biological strength and durability and solve the patient problems permanently without the necessity of revision surgery later on^[6-12].

The resulting defect may be reconstructed by a variety of methods and the choice depends upon the surgeon personal experience and the availability of the various reconstructive techniques including allograft^[16].

Allograft is an alternative method to the autogenous bone grafting for reconstruction of the curetted cyst cavity and to restore structural continuity in a skeletal defect after a wide resection of an aggressive lesions. But with much higher incidence of complications such as graft fracture and resorption, non-union and infection that make the outcome unpredictable plus the limited supply of allograft in our country^[11, 16].

The vascularized fibular bone transfer is a more favorable method for reconstruction after tumor excision with also more favorable results than allograft and the non-vascularized autogenous bone graft with a short time of graft union and incorporation and less incidence of graft fracture and infection. But is technically demanding and needs a specialized team and is a lengthy procedure and is restricted for a major skeletal defect created after resection of a malignant bone tumors when a large structural graft is needed and not for a simple benign aneurismal bone cyst lesions even if recurrent^[17, 18].

Autogenous bone grafting from the proximal 2/3 of the fibula and/or the iliac crest is the most accepted method for reconstruction and filling of a skeletal defect after curettage of a bone cyst cavity^[5, 6, 7, 10].

Phenol is a non-selective cytotoxic agent and when applied directly to the surface of curetted tumors, it kills remaining residual tumor and normal cells. Used as an adjuvant after curettage of an aneurismal bone cyst, the reported recurrence rate is 12.5% to 20%^[19, 20].

The rationale of the use of PMMA cement as adjuvant treatment is based on its heating effect^[21]. For local control of giant-cell tumors good results with recurrence rates of 5% have been reported^[21, 22], but no results are available for the use of PMMA cement as adjuvant treatment of aneurismal bone cysts.

Cryotherapy using liquid nitrogen has been used for the treatment of benign stage-2 (active), stage-3 (aggressive) and low-grade malignant stage IA skeletal tumors^[23, 24, 25]. It is often advocated to avoid extensive surgical destruction of tissue. Intralesional resection is performed by curettage. Adjuvant treatment is currently given by spraying liquid nitrogen into the bony cavity. This method must be considered marginal by oncologic principles^[3]. The advantage, compared with local resection, is that the supportive function of bone is preserved and reconstructive surgery can be limited.

In 238 patients with ABC studied in the Mayo Clinic files, more than 80% of the lesions were in long bones, flat bones, or the spinal column. In the 153 patients treated, 19% had recurrence after curettage (intralesional excision). Recurrence was most common during the first 2 post-operative years and no adjuvant measures were used^[26].

In another study done by Gibbs *et al.*^[27], they reviewed 40 patients who had been managed by the same surgeon for an aneurismal bone cyst; the median duration of follow-up was 87 months (15 to 267 months). Of the 40 patients, 34 had curettage with use of a high-speed burr. Of these 34.22 had

filling of the defect with a cancellous autogenous graft; 4 with a cancellous allograft; and 3 with polymethyl methacrylate. In 5 patients, no material was put into the defect. The remaining 6 patients had resection through the margin of the lesion. Four (12%) of the 34 patients who had curettage had a local recurrence. No patient who had an excision through the margin of the lesion had a local recurrence all local recurrences were in skeletally immature girls who were three, four, ten and eleven years old. Local recurrence was associated only with a young age and open growth plates. All local recurrences occurred within two years post-operatively and all were treated successfully with a second operation. Their final conclusion was that: rates of local control of almost 90% can be achieved with thorough curettage with use of a mechanical burr and without use other adjuvant in patients who have an aneurismal bone cyst of an extremity. A young age and open growth plates are associated with an increased risk of local recurrence. The only case with local recurrence in our study was a six year old girl with the aneurismal bone cyst in the neck of femur in whom the cryotherapy was applied for one cycle only.

Mankin *et al.*^[28], reported that their recurrence rate was 20%, after intralesional excision, which should be considered high compared with other series. And he recommends that Lesions occurred in the proximal femur should perhaps be treated more aggressively, partly because of the high rate of local recurrence and the risk of fracture. The most appropriate techniques for some of these tumors are primary resective surgery and allograft implantation. The rate of local recurrence in our study was 5% (one case) after adding cryosurgery to the technique and it was managed with second operation.

Schreuder *et al.*^[29]. Treated 26 patients with 27 aneurysmal bone cysts by curettage and cryotherapy and evaluated local tumor control, complications and functional outcome. The mean follow-up time was 47 months (19 to 154). There was local recurrence in one patient. Two patients developed deep wound infections and one had a postoperative fracture. They compared their results with previous reports in which several different methods of treatment had been used and concluded that curettage with adjuvant cryotherapy had similar results to those of marginal resection and that no major bony reconstruction was required.

They recommend the use of cryotherapy as an adjuvant to the surgical treatment of aneurismal bone cysts. It provides local tumor control. Combination with bone grafting achieved consolidation of the lesion in all of their patients.

In our study, union and cyst ossification was achieved in all the cases at an average of 5.7 months. As regard the complication was in one case of partial graft resorption at the proximal femur with cyst recurrence and managed by reoperation. 2 cases developed superficial postoperative wound infection and treated conservatively and one case developed nerve palsy and recovered after 5 months post-operatively. The average follow-up was 48 months (range, 24-72 months). The functional score, at the final follow-up, ranges from 70% to 94% with an average of 86%.

The only recurrence was probably due to the technique as only one cycle of cryotherapy had been used. Marcove *et al.*^[30]. Reported a recurrence rate of 18% which after additional treatments with the same technique decreased to 4%. Marginal and wide resection of aneurismal bone cysts lead to comparable local control, but more extensive reconstructive surgery is then needed, with associated morbidity. In general, we recommend the use of cryotherapy as an adjuvant to curettage for the treatment of aneurismal bone cysts. The use

of a bone graft allowed for consolidation in all cases with short time for cyst healing^[31].

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