Primary Treatment of Aneurysmal Bone Cyst of the Pelvis by Transcatheter Arterial Embolization

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Abstract

Background: Aneurysmal bone cysts (ABCs) could be treated by curettage, curettage and filler or even by resection. When located in the pelvis, surgery is always associated with excessive bleeding. Selective arterial embolization can be performed in pelvic ABCs as a preoperative procedure to minimize blood loss or could be used as a primary treatment option.

Purpose: The purpose of this study is to evaluate the clinical efficiency of transcatheter arterial embolization as a definitive treatment for ABCs of the pelvis.

Patients and Methods: This is a single centre prospective study of patients from July 2004 to April 2012. Twelve consecutive patients (7 males and 5 females) between the ages of 7 and 36 years (mean age 15 years) with pelvic ABCs underwent superselective transcatheter arterial embolization using poly vinyl alcohol (PVA) microparticles.

Results: Technical success of transcatheter arterial embolization was achieved in all cases (100%). There were no post procedure complications. At a mean follow-up of 36 months (range: 6 to 62 months), none of these patients required further surgical procedures. Follow-up radiographs showed bone formation and sclerosis of the lesion as early as 6 weeks.

Conclusion: Selective transcatheter arterial embolization is a safe and effective primary treatment method for pelvic ABCs.

Key Words: Pelvic aneurysmal bone cyst—Arterial embolization — Poly vinyl alcohol (PVA) particles.

Introduction

ANEURYSMAL bone cysts (ABCs) are benign, non neoplastic hypervascular expanding lesions that can be small and non aggressive or usually large and aggressive with major bone destruction. They represent 1% of all primary bone tumours with 70% occurrence in the long bones and spine; and 8 to 12% in pelvis which is a less common location.

ABCs are either primary bone lesions (70%) or secondary on a pre-existing pathology such as giant cell tumour, osteoblastoma, chondroblastoma and less commonly fibrous dysplasia and non ossifying fibroma [1,2].

The surgical treatment options for the accessible tubular extremity bones include curettage, curettage and bone grafting and en bloc excision. Other treatment modalities, such as cryotherapy, percutaneous intraluminal injection of fibrosing agents and transcatheter arterial embolization have been used [1,3]. Surgical treatment of ABCs of the pelvis is a challenging therapeutic problem due to many factors such as relative inaccessibility of the lesions, associated intra-operative bleeding, proximity to the adjacent neurovascular structures and the vulnerability of the acetabulum and the sacroiliac joint [2,4].

The method of surgical treatment of the pelvic ABCs must be individualized according to each case and it depends upon the location, extent and aggressiveness of the lesion. Lesions >5cm with major bone destruction or cortical thinning, endangering the acetabulum or sacroiliac joints require more aggressive approach with the use of excision-curettage technique; unfortunately, complete resection in this location is often impossible [1,2].

Several authors reported the use of selective transcatheter arterial embolization as a preoperative adjunct to reduce blood loss in relatively inaccessible lesions in the pelvis with significant soft tissue expansion, those in proximity of neurovas-
cular structures and in large aggressive lesions to minimize intra-operative haemorrhage resulting in better visibility and allowing a complete excision [1,2,5,6]. However, there are few reports on using this technique as a definitive method of treatment in the long bones and spine [1,7,8]. The purpose of this study is to evaluate the clinical efficiency of transcatheter arterial embolization as a primary definitive treatment for ABCs of the pelvis.

**Patients and Methods**

From July 2004 to April 2012, we had 12 consecutive patients with pathologically proven pelvic ABC, treated by transcatheter arterial embolization as a definitive primary treatment. They were 7 males and 5 females between the ages of 7 and 36 years (mean age 15 years).

In all the 12 cases, plain radiographs, computed tomography (CT) and magnetic resonance imaging (MRI) were diagnostic for ABC (Figs. 1,4,8a). All patients underwent preprocedure biopsy and histopathological examination confirmed the diagnosis of ABC.

In all the patients, acetabulum was involved by the lesion (the quadrilateral plate, both anterior and posterior columns were involved in 7 patients, the anterior column with part of the quadrilateral plate were involved in 3 patients and posterior column was involved in the last 2 patients) threatening its integrity (Figs. 1,4,8).

In 5 patients, there were large expanding lesions with major bone destruction and intrapelvic extension displacing the adjacent iliac vessels, urinary bladder and rectum to the contralateral side (Figs. 1 c,1 d,8a).

All of the patients experienced dull pain in the involved side, 6 had decreased range of motion of the affected hip and 3 had flexion deformity of the hip. All underwent selective and superselective transcatheter arterial embolization of the feeding arteries originating from the internal iliac, the external iliac and sometimes the femoral arteries.

The common femoral artery in the contralateral side of the pelvic ABC was punctured and catheterized with placement of 5F introducer sheath, and then selective internal and external iliac diagnostic angiography were performed with 5F Cobra catheter (Terumo Corporation, Tokyo, Japan) to define the feeding arteries and show the tumour blush (Figs. 2a,2b,5a,5b,7a).

Superselective catheterization of the arterial feeders was performed either by the mother cobra catheter (Figs. 2a,2b,7a) or by coaxial advancement of micro catheter (Renegade HI-FLO microcatheter, Boston Scientific, USA) through the Cobra catheter (Fig. 5b). In each case, we superselectively catheterized all of the individual arterial feeders.

Embolization was performed with 355 to 500 or 500 to 710 gm poly vinyl alcohol (PVA) particles (Contour-PVA embolization particles Target Therapeutics, Boston Scientific, USA). In all cases, administration of the particles was continued till stasis within the feeder vessel was encountered, obliteration of the lesion perfusion and vanishing of the tumour blush in the control angiograms (Figs. 2c,2d,5c,5d,7b).

Clinical and pelvic X-rays follow-up were performed after 6 weeks, 3 and 6 months and then every 6 months to monitor symptoms and to assess bone formation and ossification. Patients had an average follow-up period of 36 months (6 to 62 months).

**Results**

The sizes of the lesions ranged from 5x3 to 11x5cm in maximum cross section dimension which is large in such location with 5 patients that had intrapelvic extension, displacing the adjacent iliac vessels, urinary bladder and rectum to the contralateral side.

Technical success of the embolization procedure was achieved in all patients. All of our patients were discharged 24h after the procedure. We did not have any morbid post procedure complications.

During the early follow-up period, all the patients with hip pain showed marked improvement of pain within 4 weeks. The ones with decreased range of motion regained full range of movement after three months. The flexion deformity in 3 patients was totally resolved in 3 months. We noticed the starting of bone formation early in the 6th week of follow-up radiographs (Fig. 6a) which became obvious with notable ossification in the 3 months films. The later films show mature woven bone (Figs. 3,6b,8b,c).

No recurrence was found during the follow-up period. None of the 12 patients needed surgery or second intervention.
Fig. (1-A): AP Plain radiography of the pelvis showing expanding osteolytic lesion (ABC) involving the right iliopubic ramus and extending to the acetabulum and ischium.

Fig. (1-C): Axial CT soft tissue window showing the fluid level with high CT attenuation blood density.

Fig. (1-B): Axial CT bone window showing the involvement of the quadrilateral plate, anterior and posterior columns of the acetabulum.

Fig. (1-D): Axial T2W MR images showing the lesion cystic appearance with multiple fluid-fluid levels and the intra pelvic extension displacing the urinary bladder, acetabular & ischial involvement.

Fig. (2): (Same patient in Fig. 1): A-B: Superselective catheterization of the right obturator artery showing arterial perfusion in the early phase with blush of the lesion which is evident in the late phase.

C-D: Control angiogram after embolization by PVA particles showing occlusion of the vascular bed and vanishing of the blush in the early and late arterial phases.
Fig. (3): (Same patient in Figs. 1,2) the 9 months follow-up radiograph showing progressive ossification and bone formation.

Fig. (4-A): AP radiograph of the pelvis showing ABC involving the roof of the right acetabulum, superior pubic ramus, ischial bone and ramus.

Fig. (4-B,C): Axial MR T2W images showing multilocular cystic lesion with multiple fluid-fluid levels expanding the quadrilateral plate and acetabular columns.

Fig. (5): Same patient in (Fig. 4 A,B) Selective right external iliac [using cobra catheter] (A) & Superselective angiograms with the microcatheter in the pubic branch of the inferior epigastric artery-out of the external iliac (B) showing arterial perfusion and dense blush of the lesion with (C,D) Control angiogram after embolization by PVA particles showing occlusion of the vascular bed and almost complete eradication of the blush.
Fig. (6): (Same patient in Figs. 4,5): (A) The 6 weeks follow-up radiograph showing starting ossification of the lesion (B) The 9 months follow-up showing further bone formation.

Fig. (7-A): Selective left internal iliac angiogram showing arterial perfusion and dense blush of the lesion (B) Post embolization angiogram showing arterial vanishing of the blush of the lesion.

Fig. (8): (Same patient in Fig.7) (A) Initial CT scan of the pelvis showing the lesion eroding the acetabulum (B-C) Follow-up CT after 7 months showing evident ossification of the lesion & bone formation.
Discussion

Surgical treatment of ABCs of the pelvis is a challenging therapeutic problem due to many factors, such as relative inaccessibility of the lesions, associated intra-operative bleeding, proximity to the adjacent neurovascular structures and the vulnerability of the acetabulum and the sacroiliac joint. The method of treatment should be tailored according to location, extent, aggressiveness of the lesion and adjacent structures at risk.

The aforementioned factors open the way to the non classical methods of treatment such as percutaneous intrallesional injection and transcatheter arterial embolization to play a role besides the classic curettage and excision.

Transcatheter arterial embolization of pelvic ABC had been used as a preoperative adjunct treatment to reduce blood loss in relatively inaccessible and aggressive lesions in selected case reports as in the work of Wathiong et al. [2] and Yildirim et al. [1]. Both reported good outcome with bloodless field and later post operative intrallesional bone formation in their follow-up. Brastianos et al. [6] also described that it contributed to excellent results in the 5 cases they used as preoperative procedure in the sacral lesions. Also, Yildirim et al. [1] advocated selective arterial embolization as definitive treatment of ABCs in cases that are not accessible for primary surgical treatment.

Selective arterial embolization for the arterial feeders of ABCs as a primary modality of treatment was described in some studies and case reports outside the pelvis (in long bones, thoracolumbar and cervical spine) by Konya et al. [7] and Mohit et al. [9], as well as in the proximal femur (one case reported by Rossi et al. [8]). There is a limited discussion of pelvic ABCs embolization. Branko et al. [10] described 4 cases, De Cristofaro et al. [5] described 9 cases in the sacrum and pelvis, collectively.

In our institution, we used to perform arterial embolization for pelvic ABCs as preoperative adjunct treatment with remarkable reduction of intraoperative blood loss. In one case, the patient skipped surgery in the early post procedure period after complete occlusion of the arterial feeders by selective embolization. During the preoperative preparation, a recent plain radiograph for the pelvis was obtained and it showed the starting of bone formation. The operation was cancelled and follow-up radiograph after 3 months from the embolization was done revealing evident ossification and bone formation.

In the following 11 cases with pelvic ABCs, we aimed to complete occlusion of all the arterial feeders of the lesions and to monitor the response of using selective arterial embolization as definitive treatment.

In all cases, we selectively and superselectively catheterized the arterial feeders of the lesions and occluded them by PVA particles till obliteration of the lesion perfusion and vanishing of the tumour blush in the control angiograms were seen. Determination of the particles size is according to the calibre of the feeders and at what level (proximal or distal) the occlusion is needed. We believe that the more we obliterate the tumour bed, the better the response will be.

Only one of our patients developed post embolization syndrome with low grade fever and pelvic pain 48 h after being discharged and she was controlled by antipyretics and analgesics for 3 days. We did not have any morbid post procedure complications. All patients experienced marked improvement in the intensity of pain during the early 4 weeks.

During the follow-up period, we encountered progressive ossification and bone formation within the lesions with no recurrence in any of the 12 cases, so none of them needed further surgery or second session of embolization.

Conclusion:

Conclusively, transcatheter arterial embolization is a safe and effective treatment option for pelvic ABCs. It can be used not only as preoperative adjunct but as a definitive primary treatment for many lesions, especially those with large size, associated major bone destruction, endangering the integrity of the acetabulum as well as lesions with intra-pelvic extension or adjacent to susceptible neurovascular structures. Probably, the success of this technique relies on the ability to occlude all feeders of the lesion.

References


