TRANSCATHETER EMBOLISATION THERAPY OF AN ISCHIAL ANEURYSMAL BONE CYST

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Aneurysmal bone cysts are uncommon, benign but locally aggressive bone tumours of the young. They are usually treated by resection, or by curettage followed by bone grafting, or by radiotherapy. We report the successful transcatheter devascularisation of an aneurysmal bone cyst of the ischium which obviated the need for surgery or radiotherapy. Transcatheter embolisation therapy has previously been used to control bleeding and in the preparation of malignant and benign tumours for operative removal.

Aneurysmal bone cysts are highly vascular skeletal tumours of children, adolescents and young adults that can occur in nearly any bone of the body (Gunterberg, Kindblom and Laurin 1977; Dahlin 1978). They are best treated by excision but if this is not possible, curettage and bone grafting may be successful (Koskinen et al. 1976). However, limited accessibility for some anatomical sites has led some surgeons to use radiotherapy for selected aneurysmal bone cysts (Marks et al. 1976; Dahlin 1978). Unfortunately radiotherapy has its own complications including injury to the growth plate, osteoradionecrosis and late sarcomatous transformation (Marks et al. 1976; Brady 1979). We therefore present a case report showing the advantages of using the new technique of embolisation.

CASE REPORT

A Caucasian boy aged eight years and nine months was admitted to St Louis Children's Hospital in September 1977 for evaluation of the right hip and thigh which had been painful for one month. The pain was non-radiating, exacerbated by exercise and present both day and night. The boy had no related medical or family history. Clinical examination revealed one inch of wasting of the right thigh. Although there was no tenderness to palpation, he experienced pain with medial and lateral rotation of the hip and on straight leg raising to 60 degrees. He walked with an antalgic gait and had a positive Trendelenburg sign. The right quadriceps and hamstring muscles were found to be weak. Otherwise, he was physically normal and clinical laboratory tests were all negative.

Radiographs showed a lesion of the ischium which was diagnosed as an aneurysmal bone cyst. This diagnosis was confirmed by biopsy. For the next two months he was treated conservatively and not allowed to bear weight on the right hip. However, when he resumed weight-bearing the pain returned. In December 1977 he was readmitted for further radiological examination including computerised tomography and angiography, and the adoption of a definitive plan of treatment.

The choices of treatment available were limited to radical surgery, radiotherapy, and percutaneous transcatheter embolisation of the tumour. The latter was chosen because limited surgery would not be definitive, radical surgery would not yield acceptable limb salvage, and radiotherapy might cause sterility, avascular necrosis of the femoral epiphysis, or late sarcomatous transformation. Embolisation of the tumour was performed in two stages in January 1978.

Radiological findings and embolisation therapy. In December 1977 standard radiographs showed an aneurysmal bone cyst of the right ischium extending from the ischiopubic synchondrosis to the triradiate cartilage (Fig. 1). The tumour had expanded into the internal pelvic musculature as well as the hip joint. It was primarily lytic with a faint calcified rim except along the medial border where mineralisation had not occurred. Computerised tomography confirmed these findings and convincingly demonstrated how, due to encroachment by the tumour, operative removal would place the hip joint in serious jeopardy (Fig. 2).

At this time, a diagnostic arteriogram was performed to show the vascular anatomy of the aneurysmal cyst, to determine the feeding vessels and their origins, and to see whether it was technically possible to position the catheter precisely in the distal vessels. This was achieved by using a percutaneous transfemoral...
approach from the contralateral common femoral artery to pass a catheter around the aortic bifurcation, through the left common and internal iliac arteries, and finally into the obturator and internal pudendal arteries. Although the tumour received its major vascular supply from these branches of the internal iliac artery (Fig. 3), it also received minor vessels from small branches of the inferior gluteal artery and several terminal branches of the medial and lateral femoral circumflex arteries. As has been previously observed to be characteristic of aneurysmal bone cysts (Gunterberg et al. 1977), the periphery of the aneurysmal cyst was highly vascular while the centre was avascular.

In mid January 1978 the first percutaneous transcatheter embolisation was performed. Again, a catheter was introduced via the contralateral femoral artery, and the tip positioned in the right obturator artery (Fig. 4). Because terminal vessels of the pubic branch supplied the genitalia, the obturator artery was occluded where it bifurcated into the acetabular and pubic branches rather than at a more distal site. This was accomplished with relatively large pieces of Gelfoam. Two weeks later, the boy was readmitted and a similar procedure performed to occlude the right internal pudendal artery (Fig. 5). On this occasion, relatively small fragments of Gelfoam were used in an attempt to block the smaller, more peripheral branches on the surface of the aneurysmal cyst. Embolisation and arteriography were performed with sedation and local anaesthesia, and the patient experienced no morbidity.

**Follow-up.** Over a three-year period radiographs were taken at six-month intervals or longer and two follow-up computerised tomographic studies at approximately 18-month intervals. Results showed remarkable healing.

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**Fig. 1**—Anteroposterior radiograph of the pelvis taken at presentation shows an aneurysmal bone cyst of the right ischium. The arrows indicate the extent of the tumour. **Fig. 2**—Computerised tomographic scan through both hips taken at presentation shows aneurysmal cyst encroachment on the right hip joint and expansion into the pelvis with loss of its medial calcified boundary.

**Fig. 3**—Anteroposterior radiograph of the mid-arterial phase after injecting the internal iliac artery showing the surface vascularity of the ischial aneurysmal bone cyst which is supplied by terminal branches of the obturator, internal pudendal and inferior gluteal arteries. **Fig. 4**—Anteroposterior radiograph of the mid-arterial phase after placement of the catheter in the distal obturator artery (arrowed). Injection of contrast shows pubic arterial supply to the genitalia and retrograde filling of tumour vessels arising from terminal branches of the internal pudendal artery which is indicated by white arrow heads. **Fig. 5**—Anteroposterior radiograph of the mid-arterial phase after placement of the catheter and contrast injection in the internal pudendal artery shows its contribution to the vascularity of the supply to the aneurysmal bone cyst, but no retrograde filling of terminal branches of obturator artery which had been occluded two weeks earlier.
of the aneurysmal bone cyst. It decreased in size, re-established its borders and remineralised both peripherally and centrally (Fig. 6). Most importantly, the configuration of the hip reconstituted in a very acceptable fashion (Fig. 7). The boy is free of pain and takes part in normal activities, including competitive sports. His legs are of equal length with a full range of movements and normal strength in his right leg.

**DISCUSSION**

Transcatheter embolisation therapy has several advantages over other forms of treatment. First, it is a non-surgical technique that may be effective as the primary treatment but, if it fails, the ability to intervene surgically has not been precluded. In fact, surgical intervention may have been aided since haemostasis (a potential problem with aneurysmal bone cysts) may be less difficult as the tumour has been devascularised. Second, a percutaneous catheter may be inserted as many times as necessary to accomplish devascularisation of a lesion. In the case of benign osseous lesions, it may be more prudent to plan embolisation as a staged procedure rather than to attempt complete devascularisation at one time. A third advantage is that catheters can be manipulated into deep branch vessels and emboli placed very selectively—a precision of tissue injury that cannot be accomplished by either surgical or radiotherapeutic techniques. Fourth, transcatheter embolisation does not have the attendant morbidity and risks associated with operation and radiotherapy. For example, there is often little or no pain after embolic devascularisation as compared with pain after any operation. Similarly, as far as we know, there is no risk of sarcomatous transformation following embolisation—a situation that compares very favourably with radiotherapy.

As demonstrated by the case presented here, transcatheter embolisation techniques may have an important role as the primary treatment for a select group of benign tumours of bone. The tumours for which embolisation seems the treatment of choice are those that are highly vascular and in anatomical locations where surgical techniques are not definitive. We feel that transcatheter tumour embolisation is valuable and deserves further clinical trials.

**REFERENCES**


