SPONTANEOUS HEALING OF ANEURYSMAL BONE CYSTS

A REPORT OF THREE CASES

J. MALGHEM, B. MALDAGUE, W. ESSELINCKX, H. NOEL, P. DE NAYER, A. VINCENT

From St-Luc University Hospital, Brussels

We report three cases of spontaneous healing of aneurysmal bone cysts (ABC). In one case histological material was obtained after resection of the already ossified expansile mass discovered as a lytic lesion seven months previously. In the two other patients, spontaneous ossification of a radiologically presumed ABC in the lytic and expansile phase was observed after nine and seven months respectively. The healed lesions have remained stable at 12, 32, and 36 months respectively. These findings suggest that when the diagnosis can be made with confidence, and the lesion is in a location and at a stage that does not entail any risk of fracture or compression, expectant management should be considered.

Our three patients were aged 22, 19 and 18 years, older than usual for developing ABC. This is also true for many of the few other reported cases of spontaneous or almost spontaneous healing and suggests that ABC has a greater tendency to stabilise in older patients.

Since the report by Jaffe and Lichtenstein in 1942, aneurysmal bone cyst (ABC) has been recognised as a specific entity. ABC is an uncommon though not exceptional bone lesion representing 1% of all bone tumours for which a biopsy is carried out (Dahlin et al 1955; Mirra 1980). Despite its name, ABC is probably not an aneurysm, nor a cyst, nor a neoplasm (Bonakdarpour, Levy and Aegerter 1978), but the result of bone changes caused by a circulatory disturbance (Lichtenstein 1950; Mirra 1980; Wilner 1982). This may possibly be induced by trauma (Barnes 1956; Ginsburg 1974; Kushner, Vance and Kirkpatrick 1979; Mirra 1980; Edeiken 1981; Dabezies et al 1982) or be secondary to a benign or malignant primary disorder (Jaffe 1950; Biesecker et al 1970; Bonakdarpour et al 1978; Mirra 1980). Although ABC secondary to another disorder is fairly commonly reported in histological studies (Biesecker et al 1970; Bonakdarpour et al 1978), the radiological appearance of these cases is most often that of the underlying lesion (Bonakdarpour et al 1978). In clinical practice, ABC displays the radiological and evolutionary pattern of a distinct entity (Jaffe 1950; Campanacci, Capanana and Picci 1986).

ABC is an expansile lesion consisting of anastomosing cavernous spaces filled with unclotted blood (not stagnant but in movement) and lined with fibrous walls of varying thickness which usually contain osteoid tissue or osseous components, without elastic laminae or muscle layers (Jaffe 1950; Lichtenstein 1950; Dahlin et al 1955; Bonakdarpour et al 1978; Mirra 1980). Progression may be very rapid and the cyst may reach a considerable size. Some lesions show a sponge-like appearance while others are more fibrous with fewer cavities, looking rather like a ‘Swiss cheese’ (Dabska and Buraczewski 1969). In certain atypical tumours the solid component may predominate (Buirski and Watt 1984).

There is some controversy concerning the most appropriate method of treatment. Tumour recurrence is very frequent after bone curettage (Biesecker et al 1970; Mirra 1980; Campanacci et al 1986; Arlet et al 1987). Radiotherapy may be effective but sarcomatous change has been observed after this in some cases (Tillman et al 1968; Mirra 1980; Wilner 1982). On the other hand, there have been reports of spontaneous healing (Sherman and Soong 1957; Godfrey and Gresham 1959; Dabska and Buraczewski 1969; Rigault, Beneux and Desvygne 1972; Wilner 1982), regression after simple biopsy (Murray and Jacobson 1971; Campanacci et al 1976; Capanana et al 1985; McQueen, Chalmers and Smith 1985; Campanacci et al 1986; Scott, Connell and Duncan...
1986), and after biopsy and therapeutic embolisation (Murphy, Streeker and Schoenecker 1982).

We report three cases of lytic ABC which healed spontaneously without treatment. In the first case, histological material was obtained when resection of a completely ossified mass was eventually performed. In the remaining cases no surgical procedure was performed; the diagnosis was established on the typical radiological pattern and evolution.

CASE REPORTS

Case 1. An asymptomatic 22-year-old woman was referred for investigation of a painless mass within the pelvis, fortuitously discovered on a routine gynaecological examination. Radiological investigation showed a large endopelvic mass with slightly calcified walls connected to a moderately expanded, well-defined lytic lesion in the left iliopubic ramus (Fig. 1). The margins of the bone defect strongly suggested a benign process. The 'blow-out' appearance of the extra-osseous component pointed to the diagnosis of ABC. As the patient was entirely asymptomatic, resection of the mass was postponed for her personal convenience. Radiographs taken four months later revealed early re-ossification of the pubic ramus and development of a calcified shell around the

<table>
<thead>
<tr>
<th>Table I. Spontaneous and almost spontaneous healing of aneurysmal bone cysts</th>
</tr>
</thead>
<tbody>
<tr>
<td>Author</td>
</tr>
<tr>
<td>----------------------</td>
</tr>
<tr>
<td>Spontaneous healing</td>
</tr>
<tr>
<td>Rigault et al 1972</td>
</tr>
<tr>
<td>Wilner 1982</td>
</tr>
<tr>
<td>Malghem et al 1989</td>
</tr>
<tr>
<td></td>
</tr>
<tr>
<td></td>
</tr>
<tr>
<td>Healing after biopsy</td>
</tr>
<tr>
<td>Campanacci et al 1976</td>
</tr>
<tr>
<td>Campanacci et al 1985</td>
</tr>
<tr>
<td>McQueen et al 1985</td>
</tr>
<tr>
<td></td>
</tr>
<tr>
<td>Campanacci et al 1986</td>
</tr>
<tr>
<td>Scott et al 1986</td>
</tr>
<tr>
<td>Detection at healing stage</td>
</tr>
<tr>
<td></td>
</tr>
<tr>
<td>Dabska and Buraczewski 1969</td>
</tr>
</tbody>
</table>

Case 1. Radiographs showing a poorly calcified pelvic mass (arrows) adjacent to the left superior pubic ramus, in which there is moderately expanding lytic lesion. This lesion is sharply-defined and lined with a thin layer of sclerotic bone (arrowheads).

Case 1. Seven months later, healing is complete with no treatment. The endopelvic mass is almost completely calcified and the cystic lesion in the pubic ramus has re-ossified. A radiograph of the resected pelvic mass shows a finely ossified structure with a relatively narrow communication with the pubic ramus (arrows).
pelvic mass. Three months later (Fig. 2a), further ossification of the pubic ramus and of the endopelvic mass had taken place.

Resection of the endopelvic mass was then performed. The lesions in the pubic ramus and within the pelvis communicated through a narrow ‘diabolo-like’ connection (Fig. 2b), suggesting that the extruded material from the pubic ramus had initially been fluid. Histologically, the pelvic mass showed a honeycomb pattern of blood-filled spaces of various sizes separated by strands of fibrous tissue and abundant mature new bone with fatty marrow (Fig. 3). This appearance may represent the ultimate phase of maturation of a healing ABC. Radiographs one year later displayed a practically normal pelvis (Fig. 4).

Case 2. A 19-year-old woman presented with a three-month history of moderate pain in the left groin, which developed after a sports injury. Radiographs showed a markedly eccentric, expansile, lytic lesion in the medial segment of the upper femoral shaft, with a 5 × 5 × 6 cm extra-osseous mass lined by an extremely thin bony shell (Fig. 5a). The pattern was typical of the ‘blow-out’ appearance of ABC. CT scans with and without intravenous injection of iodine contrast medium, showed a thin-walled, finely lobulated mass with sharp borders and no internal calcification. Communication between the extra-osseous mass and the lytic bone lesion was through a relatively narrow interruption in the cortex (Fig. 5b). Inside the mass, there were several fluid levels and some streaks of vascularised tissue but there was no peripheral, hypervascularised area. In addition, no abnormal tissue was visible outside the thin wall (Fig. 5c). The diagnosis of ABC was made upon this appearance.

Case 2. Figure 5a – The initial radiograph shows an extra-osseous mass with a poorly calcified wall. Figure 5b – CT scan through the upper part of the lesion shows the geographic contour of the bone defect and the relatively narrow foramen between the intra- and extra-osseous components (arrowheads). There is no extension of the lesion into the surrounding soft tissues, despite some minor defects in the bony shell (arrow). Figure 5c – The lower scan shows the subperiosteal part of the lesion after intravenous injection of contrast medium. The expanding lesion shows wide areas of relatively low density with some fluid levels (arrowhead), separated by thin streaks of vascularised tissue. Note the absence of hypervascularised tissue at the periphery of the expansile mass.
Since pain was slight, bone weakening seemed moderate, and surgery would have been extensive, we decided, with the informed consent of the patient, to observe the lesion by repeated examination, in the hope of spontaneous healing. Figure 6 shows this radiological follow-up. After four-and-a-half months, expansion had progressed but the wall appeared slightly more calcified, suggesting some early maturation. At nine months, expansion seemed to have stopped and bone formation had started within the lesion. At 2 years 8 months, the intra-osseous defect was no longer visible and the extra-osseous mass was completely ossified. The overall size of the lesion had decreased. By that time, the patient had become entirely asymptomatic and healing was considered complete.

Case 3. An 18-year-old man gave a two-week history of low back pain which had started after playing basketball. Radiographs of the lumbar spine showed marked expansion of the left pedicle of L5 vertebra (Fig. 7a). A CT scan (Fig. 7b) showed a lytic, expansile lesion extending into the pars interarticularis and transverse process. The cortices were thinned and exhibited minute interruptions and fissures probably responsible for the painful symptoms. The characteristics of the lesion (spinal location, marked expansile tendency, thinning and remodelling of the adjacent cortices, and absence of internal calcification) suggested the diagnosis of ABC in an early phase of stabilisation.

With a reasonable hope of spontaneous healing without major complication, surgery was delayed indefinitely. The patient and his parents were informed of the absolute necessity of careful monitoring. Follow-up examinations confirmed further stabilisation of the expansile process, progressive thickening of the bony shell and some internal re-ossification (Figs 7c and 7d). At three years, radiological follow-up showed complete stabilisation, and healing was considered complete.

DISCUSSION

The radiological features of ABC vary according to the phase of development (Fig. 8). Four main stages are distinguished (Dabska and Buraczewski 1969; Wilner 1982):

1) in the initial lytic phase, a well-defined area of bone resorption with no distinctive features is observed;
2) in the phase of active development, there is the typical subperiosteal, 'blow out' expansile appearance;
3) in the stabilisation phase, there is a distinct peripheral bony shell with internal septa and trabeculations, resulting in the so-called 'soap bubble' appearance;
4) finally, the healing phase is characterised by progressive ossification of the cyst resulting in a dense bony mass of irregular structure. At this stage, recurrence is not seen (Wilner 1982).

Aneurysmal bone cyst is generally detected at the expanding or stabilisation phase (stage 2 or 3). The healing phase is usually observed after treatment, but the possibility of spontaneous healing has been considered by several authors (Barnes 1956; Sherman and Soong 1957; Godfrey and Gresham 1959; Campanacci et al 1976), but has only exceptionally been documented.

Sherman and Soong (1957) mention three cases of spontaneous healing in a series of 17 patients followed up for 3 months to 3 years before treatment. Rigault et al (1972) report a cervical spine lesion ossifying in 19

Fig. 6

Case 2. Radiographic follow-up with no treatment. From left to right: at 4.5 months, expansion has progressed, but the shell is slightly more calcified; at 9 months, expansion has stopped and the bony shell has thickened; at 1 year 9 months, the mass is almost totally ossified; and at 2 years 8 months, its size has slightly decreased.
Case 3. Figures 7a and b – The initial radiograph shows cystic expansion of the left pedicle and transverse process of the fifth lumbar vertebra and the CT scan shows the geographic contour of the area of bone destruction (black arrowheads), and minute cracks in the bony shell (arrow). There is no significant expansion into adjacent soft tissues, despite some defects in the bone shell (white arrowheads).

Figures 7c and d – After 22 months, bone expansion has stabilised and there is almost complete re-ossification.

It requires an unusual combination of circumstances to allow a presumed ABC to follow its spontaneous course towards healing without either treatment or histological proof. It is by chance that our first patient was initially untreated: she wished to postpone resection of the accidentally discovered pelvic mass. Careful radiological observation was proposed to the second and third patients to spare them from an operation, but only because several important conditions were fulfilled. The patients had trivial symptoms, and bone integrity was only moderately compromised. The patients agreed to a very strict radiological follow-up and also accepted reduced physical activity for more than one year to avoid the risk of fracture. Above all, of course, the radiological diagnosis had a very high level of probability.

In many cases, the radiological diagnosis of ABC may prove very difficult, particularly in the early expansive phase. Visualisation of fluid levels on CT, as in our second patient (Fig. 5c) is a very useful sign and has been seen not only on CT but also on MRI (Hertzanu, Mendelsohn and Gottschalk 1984; Hudson 1984; Hudson, Hamlin and Fitzsimmons 1985; Beltran et al 1986). However, fluid levels are sometimes seen in other benign lesions (Kahmann et al 1985; Resnik, Steffe and Wang 1986) as well as in malignant telangiectatic osteosarcomas (Hudson 1984), which occur in the same age group as ABC and sometimes may have a very similar appearance (Dabska and Buraczewski 1969; Matsuno et al 1976; Kaufman and Towbin 1981; Vanel et al 1987). In our first patient, histological analysis of the surgical specimen obtained after spontaneous healing revealed benign fibrous tissue and mature new bone around cavernous blood-filled spaces. In the second case, the diagnosis of telangiectatic sarcoma could practically be ruled out, taking into account the truly subperiosteal extension of the lesion in contrast to the typically central location of telangiectatic sarcomas (Matsuno et al 1976; Vanel et al 1987) and the poorly vascularised appearance on CT.
scans with intravenous contrast. This poor vascularisation of ABC appears also on angiograms (Lindbom et al 1961; Gunterberg, Kindblom and Laurin 1977) and contrasts with the appearance of malignant tumours. In addition, we noted the absence of peripheral vascularised tissue in contrast to the shell of malignant cells lining telangiectatic sarcomas (Matsuno et al 1976).

In our first two patients, the narrow communication between the extra-osseous expansile part and the intra-osseous lytic part seemed an additional sign in favour of ABC. This pattern suggests the expansion of fluid material through a narrow foramen. In the third patient, with a spinal lesion, this feature was not observed, but the risk of telangiectatic sarcoma in this location was negligible, since this tumour, like other osteosarcomas (Shives et al 1986), is very unusual in the spine (Matsuno et al 1976; Vanel et al 1987), while over 10% of ABC involve this region (Wilner 1982; Capanna et al 1985).

The spinal location in Case 3 might suggest other benign lesions, particularly benign osteoblastoma (Bonakdarpour et al 1978), but this usually contains internal calcifications and shows peripheral hyperostosis, both of which were absent in our case.

Conclusions. Aneurysmal bone cyst may heal spontaneously, as shown in our three patients, and in a few other reported cases. It is important, however, to note that such observations are rare. In the vast majority of cases, the uncertainty of the diagnosis and the risk of fracture or dramatic expansion of the lesion, require a surgical approach. Nevertheless, the possibility of spontaneous healing provides a better understanding of the natural history of the lesion and encourages consideration of a conservative therapeutic approach in selected cases.

No benefits in any form have been received or will be received from a commercial party related directly or indirectly to the subject of this article.

REFERENCES


