Solitary (unicameral) bone cyst

The fallen fragment sign revisited

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Abstract. The fallen fragment sign is a prominent radiologic feature in a minority of cases of unicameral bone cyst (20% in this series). This sign is always associated with pathologic fracture. Intramedullary fracture fragments may be single or multiple and may or may not be entirely dislodged from overlying periosteum. The finding appears limited to unicameral bone cysts in patients with open physes. When present, the fallen fragment is a pathognomonic finding as it defines the interior of the cyst as nonsolid. This is particularly helpful in cases where absence of the cyst wall secondary to fracture can simulate an intramedullary malignancy with cortical erosion.

Key words: Unicameral bone cyst – Solitary bone cyst – Fallen fragment sign – Pathologic fracture

The unicameral bone cyst is a relatively common benign non-neoplastic lesion of bone. It appears classically as a radiolucent defect surrounded by an attenuated cortex in the proximal portion of a long tubular bone. The lesion is solitary and is usually located within the metaphysis adjacent to the physis [11]. Roentgenographic differentiation from benign fibrous lesions of bone is important in planning treatment and predicting clinical outcome. Jaffe and Lichtenstein in 1942 reported the first significant clinical series of unicameral bone cysts and described many of the classic clinical and radiographic features [4]. Since then, numerous articles have appeared further defining the radiographic appearance of the lesion [1–3, 5, 8, 10].

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A pathognomonic sign of a unicameral bone cyst, the "fallen fragment sign", was first described by Reynolds in 1969 [12] and again by McGlynn in 1981 [6]. The fallen fragment is caused by fracture of the cyst wall and dislodgement of fragments into the cyst cavity. Its presence establishes that the cystic contents are not solid, thus distinguishing the unicameral bone cyst from fibrous lesions of bone which have a solid or fleshy center.

In this study, we present a striking example of this radiologic finding. Multiple fallen fragments and a large cortical defect were seen on both plain radiographs and computed tomography scans. We also discuss the radiographic findings in our other cases which demonstrate the fallen fragment sign.

Case report

KD, a 14-year-old boy, fell on his outstretched left arm and developed severe pain and swelling of the left shoulder. The patient was otherwise healthy, and the shoulder was previously asymptomatic. Plain radiographs revealed a pathologic fracture through a lytic lesion of the proximal humeral metaphysis (Fig. 1A). Computed tomography (CT) studies were performed to evaluate the possibility of a destructive malignant lesion which was suggested on an oblique plain radiograph (Fig. 1B). A CT examination revealed cortical fragments within the cystic lesion, a finding that confirmed the diagnosis of solitary bone cyst (Fig. 1C–E). Plain radiographs were taken with the arm in both full abduction and adduction to demonstrate free movement of bone fragments within the cyst, but no movement was demonstrated. This was believed to be secondary to hematoma formation from the fracture. The patient was treated conservatively. At 6 weeks, fracture healing was evident, and the cyst cavity was beginning to fill with callus.

Materials and methods

Fifty-one cases of unicameral bone cyst were reviewed from the files, collected by the three senior authors over a 25-year period. There were 39 cases in patients 17 years or younger and 12 cases in patients older than 17. The anatomic distribu-
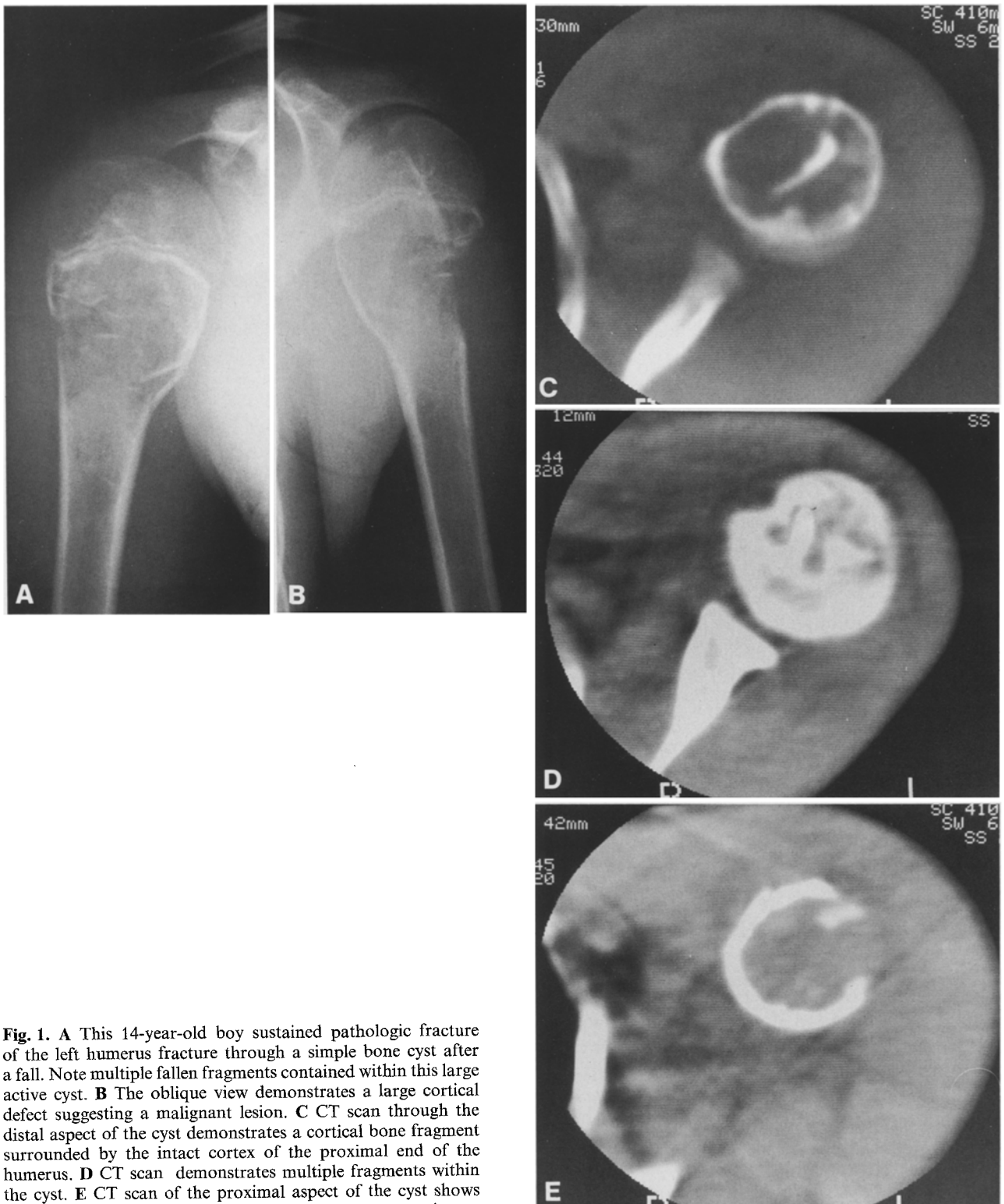
Fig. 1. A This 14-year-old boy sustained pathologic fracture of the left humerus fracture through a simple bone cyst after a fall. Note multiple fallen fragments contained within this large active cyst. B The oblique view demonstrates a large cortical defect suggesting a malignant lesion. C CT scan through the distal aspect of the cyst demonstrates a cortical bone fragment surrounded by the intact cortex of the proximal end of the humerus. D CT scan demonstrates multiple fragments within the cyst. E CT scan of the proximal aspect of the cyst shows loss of cortex and a cortical fragment within the cyst cavity

Table 1. See text

Case	Site	Physis	Stage	Fragments
1	proximal humerus	open	active	multiple
2	proximal humerus	open	active	multiple
3	proximal humerus	open	active	single
4	proximal humerus	open	latent	single
5	proximal humerus	open	active	multiple
6	proximal humerus	open	latent	single
7	proximal humerus	open	active	single
8	distal tibia	open	latent	multiple
9	distal fibula	open	active	single
10	proximal tibia	open	active	multiple

tion of lesions was similar to that of previously reported series, two-thirds of the lesions being located in the proximal end of the humerus or femur. Of these 51 cases, there were ten examples of the fallen fragment sign. These cases are summarized in Table 1.

Results

All cases were associated with pathologic fracture through the cyst. All cysts were seen in patients with open epiphyses. There were seven cases in the humerus and one each in the distal tibia, distal fibula, and proximal tibia. In five cases, the fragments were completely dislodged and were noted to be lying free in the dependent portion of the cyst (Fig. 2). In three cases the fragment was hinged into the cyst cavity but was not completely dislodged from the periosteum (Fig. 3). In two cases, a combination of hinged and displaced fragments was noted (Fig. 4). In five cases, multiple fragments were seen (Fig. 5). Seven of the cases were seen in "active" cysts (those which were adjacent to the physis), and three were in "latent" cysts.

Discussion

The fallen fragment sign occurs as a result of pathologic fracture through the thin cortical wall of the unicameral bone cyst. It is postulated by Reynolds that when the trauma which produces this pathologic fracture is minor, the surrounding periosteal sleeve remains intact, preventing the usual outward displacement of fracture fragments
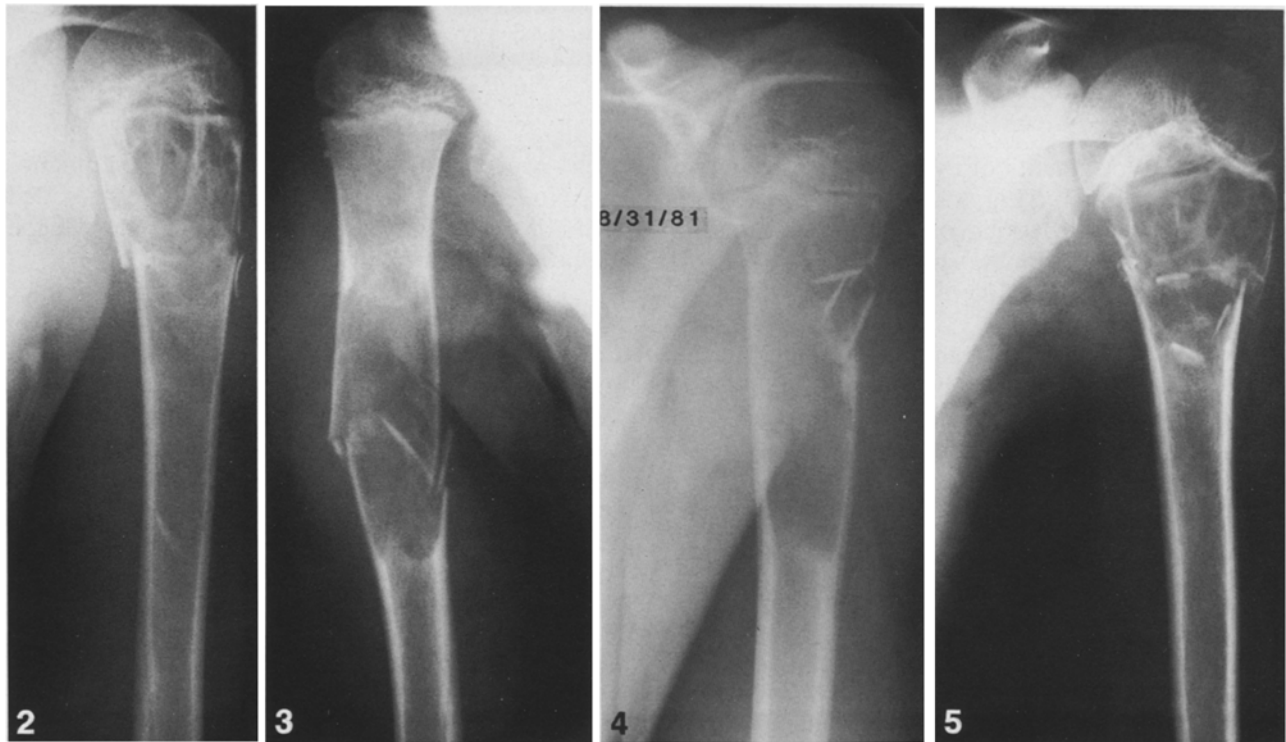

Fig. 2. An 11-year-old boy with a pathologic fracture through large solitary bone cyst. Distal displacement of a fallen fragment establishes that the lesion extends well into the diaphysis

Fig. 3. A 12-year-old boy with a pathologic fracture of a latent solitary bone cyst. Note the single fragment hinged into the center of the medullary canal. This is a forme fruste variant of the fallen fragment sign

Fig. 4. A 14-year-old boy with a solitary bone cyst demonstrated multiple cortical bone fragments. Examples of both completely displaced and hinged fragments are noted in the same cyst

Fig. 5. A 12-year-old girl with a pathologic feature through a large solitary bone cyst. Multiple cortical bone fragments were displaced into the dependent portion of the cyst
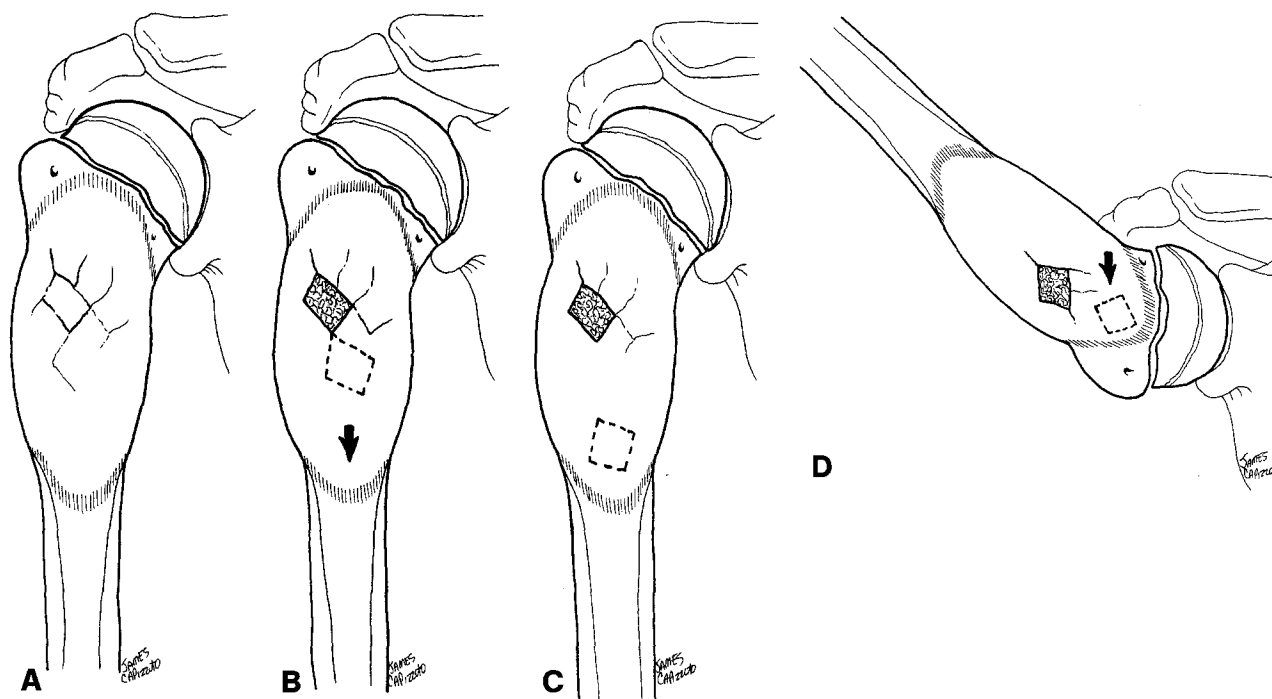
Fig. 6A–D. Pathogenesis of the fallen fragment sign. A Solitary bone cyst in the proximal end of the humerus. Thinned cortex shows "eggshell" cracks following minimal trauma. Periosteum remains intact around the comminuted undisplaced fragments. B A large cortical fragment attached to the underlying periosteum only at the bone has become a hinge-like flap (also referred to as a "trap door" fragment). C Cortical fragment has now become completely displaced and lies at the bottom of the cyst cavity creating the "fallen fragment sign". D Lying free in the cyst cavity the fragment is able to gravitate toward the top of the cavity on change in position of the arm

and causing fragments of cyst wall to fall inward into the cyst cavity (Fig. 6A–D). In other benign lesions of bone with solid or fleshy interiors such as fibrous dysplasia, aneurysmal bone cyst, or nonossifying fibroma, the fallen fragment does not occur [12].

Although pathologic fracture is present in 65% of all cases of unicameral bone cyst [1, 3, 9], the true incidence of the fallen fragment sign has not, to date, been reported. In our series, 10 of 51 patients (20%) had the fallen fragment sign. This figure may not represent the true incidence, however, as the cases in this study were collected largely from consultation files, many of which were considered diagnostic problem cases. It is noteworthy that all ten cases occurred in patients with open physes. This finding is consistent with previous reports in which all examples of the fallen fragment sign were seen in patients with open physes. When considering only juvenile unicameral bone cysts, the incidence of the fallen fragment sign was 26%.

All of our cases were also noted to be in long tubular bones which is consistent with previously published cases. Wilner [14] notes, however, that when the fallen fragment sign occurs in cysts in unusual locations such as the pelvis, calcaneus, cla-

vicle, or rib, it is helpful in confirming the diagnosis. No radiographs of these cases were provided in his text nor encountered in our review of the literature [6, 7, 11, 12].

Some variation in the radiographic appearance of the fallen fragment sign was noted in our cases. Several examples of both single and multiple fragments were noted. In addition, fragments were noted to be either completely displaced and lying in the dependent portion of the cyst or hinged into the center of the cyst. The latter example was referred to by Reynolds [12] as a "forme fruste" of the fallen fragment. It is caused by inward displacement of a cyst wall fragment that is still connected to overlying periosteum. In some cases, both varieties of fallen fragment were seen in the same cyst. In terms of both anatomic location and physeal proximity (active or latent), cases which demonstrated the fallen fragment sign followed a pattern typical for unicameral bone cysts.

While the presence of the fallen fragment establishes the hollow nature of the cyst, it can also be of value in estimating size. In cases where the cyst extends well into the diaphysis, it may be difficult to identify the distal extent of the lesion. However, when the fallen fragment is free and lies in

the most distal portion of the cyst, the true size of the cyst may be appreciated. This concept is dramatically illustrated in one case (Fig. 2) in which the intramedullary cortical fragment is seen far down the diaphysis.

Since the fallen fragment sign serves as a pathognomic indicator of solitary bone cyst, accurate identification is important. Multiple views may be necessary to establish the intramedullary location of cortical bone fragments. Pathologic fracture through a solid fibrous lesion can produce fragments that appear intramedullary, but are in fact in the extraosseous soft tissue. When in doubt, other imaging techniques may be helpful. Plain radiographs taken in both the erect and recumbent positions may demonstrate free movement of fragments within the cyst [7]. Fracture hematoma may, however, prevent free movement of bone fragments within the cavity as was likely in our case. Alternatively, CT scanning is extremely helpful in establishing the intramedullary location of bone fragments (Fig. 1 C, D, E). In addition, CT also allows more detailed evaluation of soft tissue which may confirm the benign radiographic appearance, as it did in our case presentation.

Cortical fracture fragments, it should be noted, are not the only source of radio-opaque strutures within solitary bone cysts. Sanerkin, in reporting on the content of 85 solitary bone cysts, found a variety of solid material within the cyst cavity in one-third of the cases [13]. This amorphous tissue which often lay free in the cyst cavity showed a variable degree of ossification and calcification. It appeared to have originated from old floccules of fibrin coagula which were not subject to early organization by granulation tissue because they were formed in an avascular inert chamber. This tissue underwent senescence and calcific encrustation. Eventually, Sanerkin stated, the calcified coagulum was vascularized from the cyst lining and became ossified in a manner analogous to that oc-

curring in enchondral ossification. While this is a unique and pathognomonic histologic finding for simple bone cysts, it is quite rare for such tissue to be seen on routine radiographs. When it is sufficiently calcified and large enough to be visualized on radiographs, differentiation from fallen fragments is not difficult. It appears as a patchy density with indistinct edges or as a rounded incompletely calcified mass. This is quite different from the sharp edges of cortical bone fracture fragments.

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