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Possible Precursor of Unicameral Bone Cysts

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Although most aspects of solitary bone cyst have been clearly documented^{1,2,3}, the pathogenesis remains a mystery. The following two unique cases were inadvertently discovered while under development and provide additional evidence for consideration in theories of pathogenesis.

Case Reports

CASE 1. E. B., a ten-year-old white girl was treated in September 1960 for epiphyseolysis of the right femoral head. A lesion in the subtrochanteric region of the left femur was incidentally noted (Figs. 1-A and 1-B), measuring one and one-half by one and one-quarter inches. There was a rim of increased bone density near the periphery, possibly separated from the normal bone by a thin radiolucent zone. Irregular circular small radiopacities were seen more centrally. The parents were warned about slipped epiphysis on the opposite side and were advised to bring the child back immediately if symptoms occurred in the left hip.

The right hip healed and became asymptomatic. Two years later the girl complained of pain and limp referable to the left hip. Roentgenograms were again made (Figs. 1-C and 1-D). Whereas slipping of the contralateral epiphysis was suspected, a fully developed unicameral bone cyst was found in the subtrochanteric region at the site of the previous irregular increase in density instead.



FIG. 1-A

Anteroposterior roentgenogram of the pelvis and hips, September 1960.

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FIG. 1-B

Lateral roentgenogram of the pelvis and hips, September 1960.



FIG. 1-C

Anteroposterior roentgenogram of the pelvis and hips, 1962.

At operation, the cyst, measuring approximately four by one and three-quarter inches, was extirpated. It contained approximately ten milliliters of clear yellow fluid and a fibrous-tissue lining. After the curettage, the cavity was treated with phenol and alcohol and then packed with autogenous iliac-bone chips. It healed (Figs. 1-E and 1-F). There was no recurrence. The patient remained asymptomatic.

Microscopic examination of the cyst lining was reported by the pathology department of Chester County Hospital—"benign and typical of unicameral bone cyst."

CASE 2. M. A., a two-month-old white female infant was seen in February 1962 for congenital dislocation of the right hip. Treatment in an abduction splint was unsuccessful. Six months later



FIG. 1-D



FIG. 1-E

Fig. 1-D: Lateral roentgenogram of the left femur, 1962.

Fig. 1-E: Anteroposterior roentgenogram of the left femur, 1963.

Fig. 1-F: Lateral roentgenogram of the left femur, 1963.



FIG. 1-F

closed reduction was gently accomplished without anesthesia and the frog-leg position was maintained in a plaster cast for six weeks. The patient was then treated for two months in bilateral toe-to-groin casts held in abduction and internal rotation by a long cross bar. Long abduction braces were then used full time for six months and as a night splint for over three years. The clinical examination was normal by September 1964. Roentgenograms made in 1964 and February 1965 (Fig. 2-A) demonstrated a suspicious irregular increase in density in the intramedullary portion of the subtrochanteric area of the right femur. In January 1966 (Fig. 2-B), the same area was occupied by a small centrally located cystic lesion. In July 1967 (Fig. 2-C), because of enlargement of this lesion, surgery was done. The cyst cavity measured two and one-half by three and one-quarter inches and contained about two milliliters of clear yellow fluid. The lining was thin and a few osseous ridges extended across its length. One dense osseous ridge almost occluded the medullary canal below. The canal superiorly was easily entered with a curette. The cavity was filled with autogenous iliac chips and strips after reaming with a high-speed air-driven burr.

The specimen and the roentgenograms were forwarded to E. E. Aegerter, M.D. for comment. He wrote "The unusual position and circumstances of this lesion make it rather unique. I agree with you that it is almost certainly a solitary cyst. Recently I have collected a number of these cysts which seem to be secondary to hemorrhage in the cancellous tissue, some of them post traumatic. It is interesting to speculate here whether previous surgical manipulation may have played a role in the genesis of this lesion."

Discussion

A search of the English-language literature on this subject revealed no observation of a cyst under development. All reported cysts, and those personally observed,



FIG. 2-A



FIG. 2-B



FIG. 2-C

Fig. 2-A: Anteroposterior roentgenogram of the right hip, February 1965.

Fig. 2-B: Anteroposterior roentgenogram of the right hip, January 1966.

Fig. 2-C: Anteroposterior roentgenogram of the right hip, July 1967.

with the exception of the present cases, have been well formed when recognized. Other than the classification of these lesions into active (those extending to the vicinity of, or abutting on, the epiphyseal line) and latent (those that occupy an area away from the epiphyseal line), there is no known recording of the rate of development of a cyst. Many well developed lesions, under observation, have migrated away from the epiphyseal plate and even enlarged somewhat as growth and maturation occurred.

The cyst in Case 1 developed within a two-year period at the site of an eccentric benign lesion with the roentgenographic appearance of a non-ossifying fibroma or calcified cartilage rest.

The cyst in Case 2 developed over a five-year span at the site of a poorly defined lesion, more centrally placed than

that in Case 1. The initial lesion here also had the characteristics of a non-ossifying fibroma or a calcified cartilage rest. Both lesions became oval, concentric, and centrally located in the subtrochanteric area of the femur. Neither abutted on the epiphyseal line.

The present cases suggest two possibilities and may lend support to Cohen's hypothesis of etiology which postulates the following stages of development:

1. Formation of a focus of fibrous tissue in an area of rapid resorption of bone;
2. Blockage of sinusoidal vessels in an area such as this, leading to accumulation of interstitial fluid in the fibrous tissue;

3. Subsequent equilibration of this fluid with that in unblocked vessels, giving the characteristics of cyst fluid similar to serum plasma. If the present cases indeed did develop within or adjacent to a non-ossifying fibroma, or calcified cartilage rest, then this would offer support for this hypothesis. However, it is possible that the cysts described developed independently and merely engulfed the original lesion seen in Figures 1-A, 1-B, and 2-A.

Neither of these cysts abutted on the epiphyseal line at any time in their development. This would be a point against etiologies based on disturbances of endochondral ossification unless there is more than one sequence of development. Garceau and Gregory discussed the various etiologic theories and noted only that the lesions occur during growth periods and in locations where extensive remodeling is taking place. They speak of remodeling out of control.

Jaffe mentioned the Mönckeberg theory of etiology, favored by Konjetzny as well as Geschickter and Copeland, that unicameral cysts represent a healing form of a giant-cell tumour or of osteitis fibrosa. He stated that solitary bone cyst "is in no way related to any of these conditions. Nor does it develop through cystic softening of a fibro-osseous disease focus." He also discards the Pommer theory, of formation in an area of intramedullary hematoma due to unrecognized trauma, as unsound, and without clinical, roentgenographic, or anatomical proof.

Luck reviewed various etiologic beliefs including those of Pommer, Geschickter, Jaffe, and Lichtenstein and finally, Phemister and Gordon (solitary cysts represent the healed end stage of a localized, low-grade pyogenic osteomyelitis) only to conclude that "Thus far the various etiologic theories have been of little practical value in our understanding and treatment of the solitary cyst."

Conclusions

The known facts concerning unicameral bone cyst, such as the anatomical distribution, age incidence, histology, characteristics of the contained fluid, and the development in areas of rapid bone remodeling, lend credence to the theory of pathogenesis which postulates a sequence of events initiated by vascular obstruction in a focus of fibrous tissue located in an area of rapid remodeling of bone. The present two cases, which are the first on record which were observed during the early stage of development, may lend support to this theory since they were preceded by lesions which may very well have represented foci of fibrous tissue in non-ossifying fibromas. Since non-ossifying fibroma is a very common lesion, however, it is strange that this sequence of events has never been observed and reported previously.

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