

# UNICAMERAL BONE CYSTS OF THE PELVIS: A STUDY OF 16 CASES

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## ABSTRACT

**Unicameral bone cysts of the pelvis are extremely rare. This study summarizes the clinical, radiologic and pathologic features of 16 cases. Patients ranged in age from nine to 69. Most lesions were in the anterior portion of the iliac wing; many appeared to be related to an open iliac crest apophysis. This suggests that the pathogenesis of unicameral bone cysts in this portion of the ilium is similar to that seen in the proximal humerus and the proximal femur. The correct diagnosis was made preoperatively in only five cases. This indicates that, although they are well documented, unicameral bone cysts of the pelvis remain a diagnostic problem. Patients received a spectrum of treatments from curettage to observation. There appeared to be no difference in the outcome after any form of treatment. Therefore, unicameral bone cysts of the pelvis can be managed conservatively. The choice to manage patients conservatively depends on making the correct diagnosis based on clinical history and imaging. The most effective imaging is a combination of plain radiographs, computed tomography (CT) and magnetic resonance imaging (MRI).**

## INTRODUCTION

Unicameral bone cysts, also known as simple bone cysts, are relatively common bone lesions that usually occur in the proximal femur or proximal humerus in the developing skeleton. Very rarely, a unicameral bone cyst may form in the pelvis. Because of their rarity, lesions in the pelvis are often misdiagnosed and over-treated. This paper summarizes the clinical, radiographic and pathologic features of sixteen cases of unicameral bone cysts of the pelvis, collected over a thirty-year period.

## METHODS

The 16 patients were either collected from the orthopaedic surgery files of the Johns Hopkins Hospital or culled from the personal consultation files of one of us (EFM) (Table 1). Plain radiographs of the pelvis were available on all 16 patients. Thirteen patients had either CT scans or MRI studies or both. Histological material from 12 patients was available for review.

## RESULTS

Patients ranged in age from nine to 69 years. The mean age was 30 years. Six patients were age 20 or younger. There were ten females and six males.

Fourteen lesions occurred in the ilium. One was in the pubic ramus, and one was in the ischium. Of the 14 lesions in the ilium, 11 were present in the iliac wing. Five patients had cysts in the posterior part of the ilium adjacent to the sacroiliac joint. These posterior lesions occurred in the older group of patients.

These patients' symptoms varied. Two patients had dull, aching pain probably directly related to their cysts. Two patients had pain after minor trauma to the ilium. One patient suffered a pathologic fracture of the lesion after a fall. In eight patients, lesions were discovered incidentally during evaluation for other symptoms such as back pain or hip pain. The clinical presentation was unclear in the remaining three patients.

Nine patients had curettage of their lesions. In one of these patients, the lesion was filled with bone graft. Three patients had only a biopsy. One patient had drill holes placed, and another had a needle aspiration. Two patients were observed only and had no surgical intervention.

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Each author certifies that his or her institution has approved the reporting of this case series, that all investigations were conducted in conformity with ethical principles of research, and that informed consent was obtained.

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**TABLE 1**  
**Simple Bone Cysts of the Pelvis**

Case	Age/ Sex	Location	Presentation	Preoperative Diagnosis	Treatment	Years of Follow Up
1	9F	Left ilium	Asymptomatic	Fibrous dysplasia	No treatment	1
2	14F	Left ilium	Vague discomfort after trauma	Simple cyst	Aspiration	8 weeks
3	15M	Left ilium	Pathologic fracture after fall	Aneurysmal bone cyst	Biopsy and curettage	2
4	16F	Right ilium	Incidental finding	Simple cyst	Biopsy and curettage	4
5	17M	Right ischium	Incidental finding	Fibrous dysplasia	No treatment	1
6	20M	Left ilium	Unclear	Aneurysmal bone cyst	Biopsy only	1
7	26F	Right ilium	Vague pain after trauma	Fibrous dysplasia	Curettage, Re-curettage after 21 years	25
8	27F	Right ilium; posterior	Incidental finding Back pain	Simple cyst	Drill holes	8 months
9	33F	Left ilium	Vague dull pain	Aneurysmal bone cyst	Biopsy curettage	3
10	36M	Right ilium; posterior	Vague dull pain	Simple cyst	Curettage	2
11	43F	Left ilium	Hip pain	Fibrous dysplasia	Biopsy partial excision	3
12	48F	Right ilium; posterior	Incidental finding	Neoplasm	Cyst removed	2
13	48F	Pubic ramus	Incidental finding	Simple cyst	Curettage	3
14	48M	Right ilium; posterior	Unclear	Aneurysmal bone cyst	Needle aspiration	2
15	52M	Right ilium	Unclear	Neoplasm	Curettage	1
16	69F	Right ilium; posterior	Asymptomatic	Neoplasm	Biopsy only	1

In the seven patients who had no treatment, a biopsy only, or drilling, the follow-up was one year or less. Eight of the patients who were treated with curettage were followed from one to four years. One patient had a second curettage 21 years after the first.

**Radiographic Findings**

On plain radiographs, lesions were well-circumscribed lytic zones ranging in size from 3 to 12 cm. One lesion occupied almost the entire iliac wing (Figure 1). Most lesions had a faint sclerotic rim (Figure 2). Some lesions had a soap-bubble type appearance (Figure 3). These inner markings corresponded to ridges on the inside of the cysts as seen best on the CT scan. These were seen more frequently in older patients, suggesting attempted spontaneous healing (Figure 4). On CT scans, lesions showed no or minimal expansion of the wall of the pelvis. CT scans demonstrated that lesions were either in the anterior iliac wing or posterior, adjacent to the sacroiliac joint. The posterior location was more prevalent in older patients (Figure 5A and B). Except for a few cases with inner trabeculations, all le-

sions were unilocular. On T2-weighted MRI studies, lesions showed a uniform high signal corresponding to fluid (Figure 6). There were no fluid-fluid lines typical of aneurysmal bone cyst.

Based on these radiographic features, the diagnosis of unicameral bone cyst was made in only five patients. The diagnosis of fibrous dysplasia was made in five cases, and aneurysmal bone cyst was suspected in three patients. In three patients, the lesion was diagnosed as a neoplasm. The correct diagnosis was made when the lesions were studied with a combination of plain radiographs, CT scans and MRI.

**Histopathologic Findings**

Tissue was available for study from 12 patients. Tissue was obtained at curettage or from an open biopsy. In all cases where surgery had been performed, the surgeon reported the lesion to be filled with clear sero-sanguineous fluid. The biopsy showed a fibrous membrane with varying amounts of reactive bone (Figure 7). In several cases, a fluffy material was present in the fibrous wall (Figure 8). This was identical to the char-

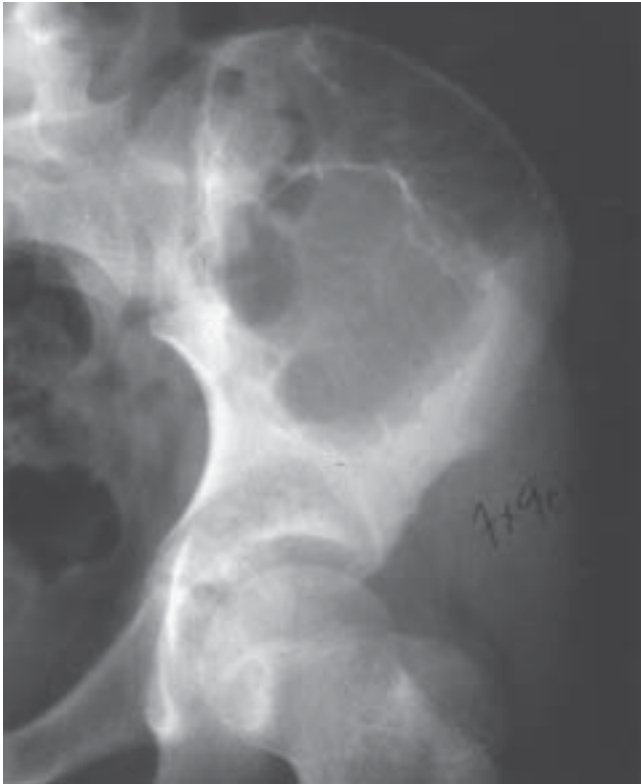


Figure 1. Plain radiograph of a unicameral bone cyst of the iliac wing from patient number one. This lesion occupies most of the iliac wing and was asymptomatic.



Figure 2. This unicameral bone cyst of the left ilium from patient number two has a faint sclerotic rim (arrows).

acteristic cementum-like material seen in the walls of typical unicameral bone cysts of the proximal humerus or proximal femur.<sup>1</sup>

### DISCUSSION

A unicameral bone cyst is a unilocular cavity in bone, lined by a fibrous membrane and filled with fluid. The lesion almost always arises in the metaphysis of a long bone adjacent to an epiphyseal plate. The development of the lesion is related to skeletal growth. For a period of time bone does not form in the metaphysis and the defect fills with fluid. Eventually, bone formation recommences and normal medullary bone forms between the physis and the cyst. This normal zone lengthens as skeletal growth continues. The cause of this process is not known.

The first description of a unicameral bone cyst was by Virchow in 1876.<sup>2</sup> Although Bloodgood in 1910<sup>3</sup> wrote a detailed early report, it was not until 1942 that Jaffe and Lichtenstein characterized fully the clinical, radio-

graphic and pathologic features of unicameral bone cysts.<sup>4</sup>

Although unicameral bone cysts may occur anywhere in the skeleton, two-thirds of cases occur either in the proximal humerus or proximal femur.<sup>5</sup> The pelvis is an uncommon site for the development of a unicameral cyst. This location accounts for only two percent of reported cases.<sup>6</sup> In 1975, Samuelson et al reported two cases, and summarized the 20 other cases documented at that time.<sup>7</sup> Since that summary, there have been isolated case reports.<sup>8,9,10</sup> In 1977, Norman and Schiffman reviewed 75 cases of simple bone cysts.<sup>11</sup> They noted that the nine cases which were in the pelvis occurred in older patients, and they suggested that the cysts developed later in life. More recently, however, Abdelwahab et al reported four cases of unicameral pelvic bone cysts in adolescents.<sup>6</sup> These authors concluded that in the pelvis, as in the long bones, cysts formed during skeletal development. Pelvic lesions may remain asymptomatic longer because they usually occur in non-weight-bearing portions of the ileum.



Figure 3. A unicameral bone cyst in the posterior portion of the ilium from patient number nine showing a multi-locular appearance. These internal markings are ridges on the inside of the cavity and suggest healing.



Figure 4. A unicameral bone cyst (arrow) from patient number seven in the right iliac wing showing changes suggestive of healing.

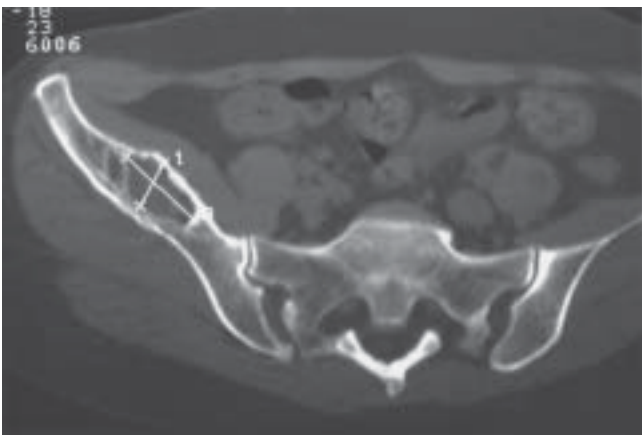


Figure 5A. CT scan of the left ilium of patient number six showing a lesion with a single intralesional ridge. There is minimal expansion of the bone.

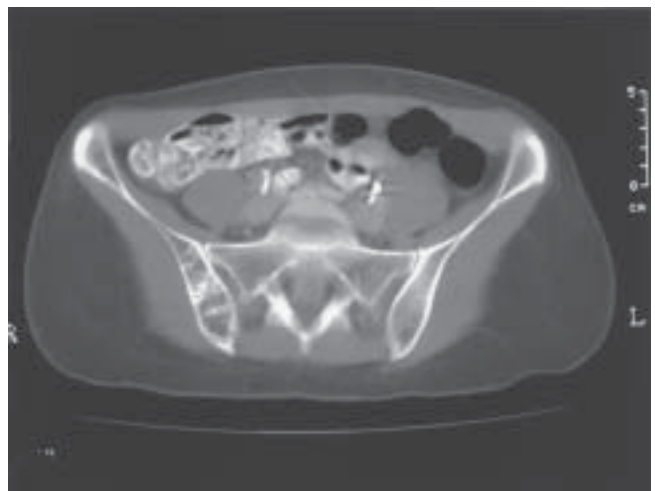
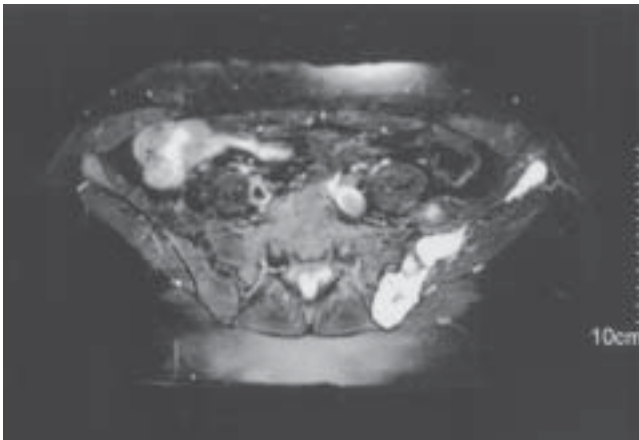
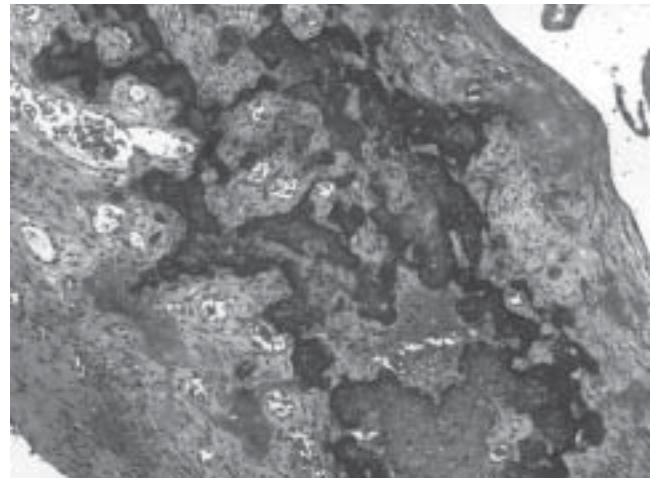


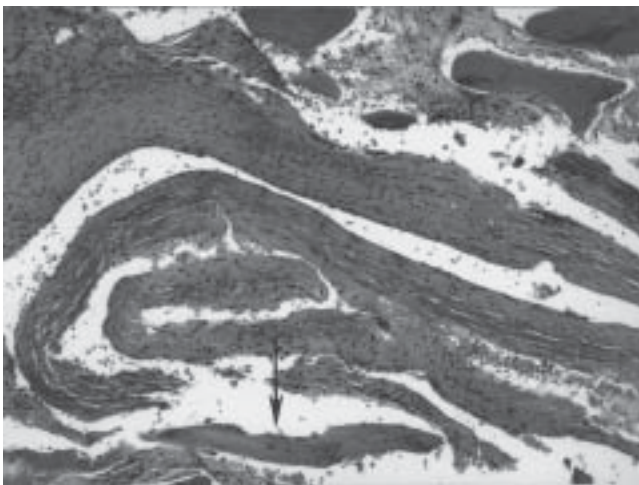
Figure 5B. CT scan of a unicameral bone cyst in the posterior portion of the ilium from patient number ten. Internal marking suggest healing.



**Figure 6.** T2-weighted MRI scan of the unicameral bone cyst of patient number 15. There is uniformly high signal throughout much of the iliac wing. This high signal represents fluid, and there is no cortical expansion or fluid-fluid line.



**Figure 8.** A high-powered photomicrograph (H&E, x40) of the fibrous membrane. There is fluffy, cementum-like material in the wall. This material is highly characteristic of unicameral bone cysts.



**Figure 7.** A low-powered photomicrograph (H&E, x20) of the fibrous membrane curetted from a pelvic unicameral bone cyst. Some reactive bone is present in the membrane (arrow).

In the present series of 16 cases, lesions were diagnosed in both young and older patients. In the adolescent group, more lesions occurred adjacent to an open iliac crest apophysis. This suggests that the pathogenesis of unicameral bone cysts in the ilium is similar to that of lesions in the proximal humerus and femur. Five of our cyst cases occurred in the posterior portion of the ilium adjacent to the sacroiliac joint in an older age group, and may represent subchondral cysts related to sacroiliac joint degeneration. Two of our cyst cases had atypical locations, one in the pubis and one in the ischium. Because they were unilocular and filled with fluid, we regarded them as unicameral bone cysts.

The study of these 16 cases emphasizes several points. First, despite previous documentation of unicameral cysts in the pelvis, this lesion remains a diagnostic problem. The correct diagnosis was made preoperatively in only five cases. The most frequent misdiagnoses were fibrous dysplasia and aneurysmal bone cyst. An MRI demonstrating intralesional fluid should rule out fibrous dysplasia. The absence of cortical expansion and multiple locules with fluid-fluid lines rules out aneurysmal bone cyst.

The second point is that the frequent misdiagnosis leads to overtreatment. Follow-up indicates that patient outcomes are the same whether lesions are curetted, drilled or observed. Whether treated or not, cystic cavities persist in the pelvis and remain asymptomatic. Because the pelvis is a non-weight-bearing bone, these lesions do not threaten the integrity of the skeleton. This suggests that unicameral bone cysts of the pelvis may require no surgical intervention. Symptomatic lesions may be treatable with drill holes similar to proximal humeral lesions. Lesions in older patients tended to have more intralesional trabeculations suggesting spontaneous healing. Therefore, the correct preoperative diagnosis is critical to avoid overtreatment.

These cases indicate that simple cysts of the pelvis are most accurately diagnosed with a combination of plain radiographs, CT scans and MRI. Once the diagnosis is established, lesions may be best managed conservatively.

#### REFERENCES

1. **Mirra JM, Bernard GW, Bullough PG, Johnston W, Mink G:** Cementum-like bone production in solitary bone cysts (so-called "cementoma" of long bones). Report of three cases. Electron microscopic observations supporting a synovial origin to the simple bone cyst. *Clin Orthop* 1978; 135:295-307.
2. **Virchow R:** Ueber die bildung von knochencysten. *Monatsber d Kgl Akad D Wissenschaften, Sitzung der Phisikalischen-mathemat Klasse vom 12 Juni, 1876.*
3. **Bloodgood JC:** Benign bone cysts, osteitis fibrosa, giant cell sarcoma and bone aneurysm of the long pipe bones. A clinical and pathological study, with the conclusion that conservative treatment is justifiable. *Ann Surg* 1910; 52:145.
4. **Jaffe H, Lichtenstein L:** Solitary unicameral bone cyst. *Arch. Surg* 1942; 44:1004.
5. **Boseker E, Bickel W, Dahlin D:** A Clinicopathologic Study of Simple Unicameral Bone Cysts. *Surg Gynec and Obstet* 1968; 127: 550-560.
6. **Abdelwahab IF, Hermann G, Norton KI, et al:** Simple bone cysts of the pelvis in adolescents. A report of four cases. *J Bone Joint Surg (Am)* 1991; 73(7):1090-1094.
7. **Samuelson KM, Momberger GL, Coleman SS:** Solitary bone cyst of the ilium. Report of two cases and a review of the literature. *Rocky Mt Med J* 1975; 72(10):443-445.
8. **Blumberg ML:** CT of iliac unicameral bone cysts. *AJR Am J Roentgenol* 1981; 136:1231-1232.
9. **Baker DM:** Benign unicameral bone cyst. A study of forty-five cases with long-term follow up. *Clin Orthop* 1970; 71:140-151.
10. **Wray CC:** Unicameral bone cyst of the ilium. *JR Coll Surg Edinb* 1986; 31(4):233-236.
11. **Norman A, Schiffman M:** Simple bone cysts: factors of age dependency. *Radiology* 1977; 124(3):779-782.