ORIGINAL ARTICLE

Acta Orthop Traumatol Turc 2014;48(5):495-499
doi: 10.3944/AOTT.2014.14.0039

Iliac bone cysts adjacent to the sacroiliac joint: an unusual cause of sacroiliac pain

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Objective: The aim of this study was to describe cystic bone lesions involving the sacroiliac region of the iliac bone as a rare cause of sacroiliac joint-related pain.

Methods: The study included 9 patients with benign cystic bone lesion in Zone 1 according to Enneking and Dunham with a minimum of 2 years follow-up. Detailed radiological examination was performed using magnetic resonance imaging or computed tomography. Extended curettage, adjuvant treatment with phenol and thermal cauterization and bone grafting with auto/allografts were performed. Patients were analyzed for age of onset, side of involvement, clinical and radiological findings, tumor stage, complications and clinical/radiological findings at the final follow-up.

Results: Female to male ratio was 4 to 5. Average age at the time of diagnosis was 40.3 (range: 27 to 54) years. Average diameter of the lesion was 6.6 (range: 4 to 11) cm. Superficial infection was detected in one patient and hypoesthesia around the site of incision in two. Clinical improvement in pain and limp was reported in all patients. No radiological recurrence was detected after an average follow-up time of 30.7 (range: 21 to 40) months.

Conclusion: Benign tumor-like cystic lesions should be kept in mind in the differential diagnosis of sacroiliac joint-related pain. Such lesions can be safely and effectively managed with intralesional curettage, local adjuvant methods and bone grafting.

Key words: Aneurysmal bone cyst; pelvis; sacroiliac pain; simple bone cyst.

The sacroiliac joint is the largest axial joint in the human body and transmits the load from the spine to the lower extremities. Sacroiliac joint-related pain is a very common clinical symptom and is responsible for 13 to 30% of all lower back pain cases.[1] As 70% of the population suffer from lower back pain at least once in their lifetime, the abundance of sacroiliac joint-related pain in the population can be better understood.[2] Recent studies demonstrated that the reliability of physical examination in the diagnosis of sacroiliac joint pathologies is low and positive findings can be detected even in asymptomatic patients.[3,4] In addition, conventional radiography is not helpful in diagnosis because of the complex anatomy of the pelvis. Therefore, diagnosis of sacroiliac joint pathologies can be difficult and delays are very common. Discogenic pain, trauma, rheumatic diseases, malignity, pregnancy and sports-related pain exist in the differential diagnosis (Table 1).[5,6] On the other hand, the posterior

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Submitted: January 23, 2014 **Accepted:** July 09, 2014

Available online at
www.aott.org.tr
doi: 10.3944/AOTT.2014.14.0039
QR (Quick Response) Code

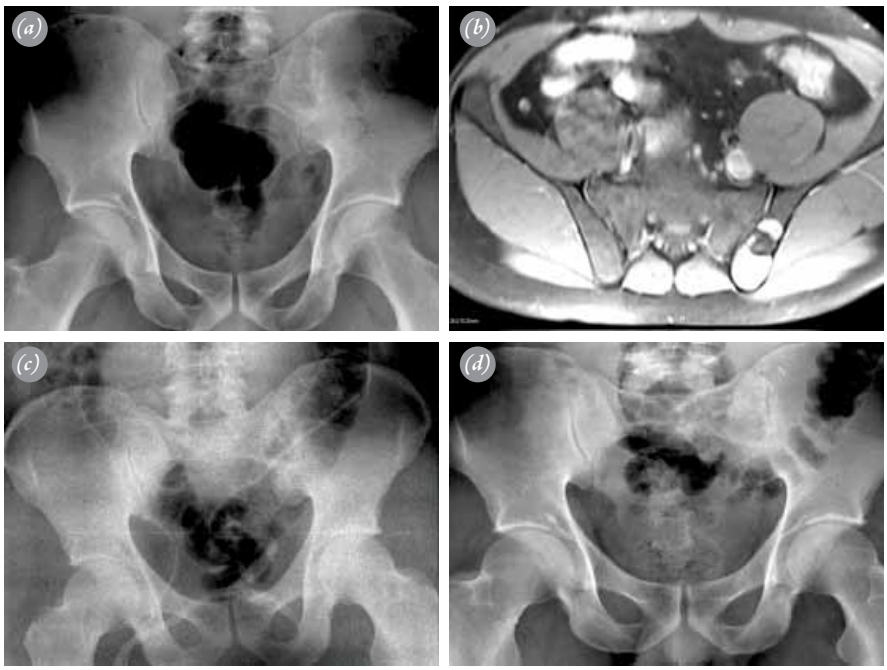

Fig. 1. Thirty-five years old male admitted with pain in the left sacroiliac region. **(a)** Anteroposterior radiograph demonstrates a cystic bone lesion in the left iliac bone adjacent to the sacroiliac joint. **(b)** Axial magnetic resonance image of the lesion. **(c)** Intralesional curettage and bone grafting was performed. **(d)** After 31 months of follow-up, there was no radiological sign of recurrence on the anteroposterior radiograph of the pelvis.

region of the iliac bone, adjacent to the sacroiliac joint, is an uncommon and unexpected site for benign cystic tumor or tumor-like lesions and is not described in the differential diagnosis of sacroiliac joint-related pain.

The aim of this study was to describe the benign cystic bone lesions in pelvic Zone 1 in patients with complaints of sacroiliac joint pain and to present the surgical outcomes.

Patients and methods

The study included 9 patients with cystic bone lesion located in Zone 1, according to Enneking and Dunham,[7] identified after clinical and radiographic evaluation of patients with lower back pain at a single institution between 2007 and 2010. Informed consent was received from all patients. Detailed radiological examination was performed with either computed tomography (CT) or magnetic resonance imaging (MRI).

Surgery was performed in all cases because symptoms were not relieved with rest and conservative methods. Extended curettage, adjuvant phenol and thermal cauterization and bone grafting with auto/allografts were performed. Patients were analyzed for age, gender, site, clinical and radiological findings at onset, tumor stage, surgical outcomes, complications and clinical/radiological findings at the final follow-up.

Results

Female to male ratio was 4 to 5. Average age at onset was 40.3 (range: 27 to 54) years. The average follow-up was 30.7 (range: 21 to 40) months. The main symptom was monolateral sacroiliac pain ongoing for a minimum of 3 months, worsening with activity and unresponsive to analgesics. All patients had a history of unsuccessful conservative treatment for lower back pain. Antalgic gait was detected in 3 patients. In physical examination, tenderness was detected over the sacroiliac region and the posterior superior iliac spine. Increase in pain was reported on the ipsilateral flexion-abduction-external rotation test in all patients. There was no sacral nerve root deficit.

Patients underwent routine radiographic evaluation due to their history of pain unresponsive to rest and bone tenderness in palpation. Benign cystic lesion was detected in the posterior part of the iliac bone adjacent to the sacroiliac joint in anteroposterior pelvic and sacroiliac radiographs. There were no pathological fractures. Detailed radiological evaluation with CT or MRI was performed to detect the localization, size and soft tissue involvement of the lesions (Figs. 1 and 2). The largest diameter of the lesions was an average of 6.6 (range: 4 to 11) cm.

Surgical procedures were performed in the prone

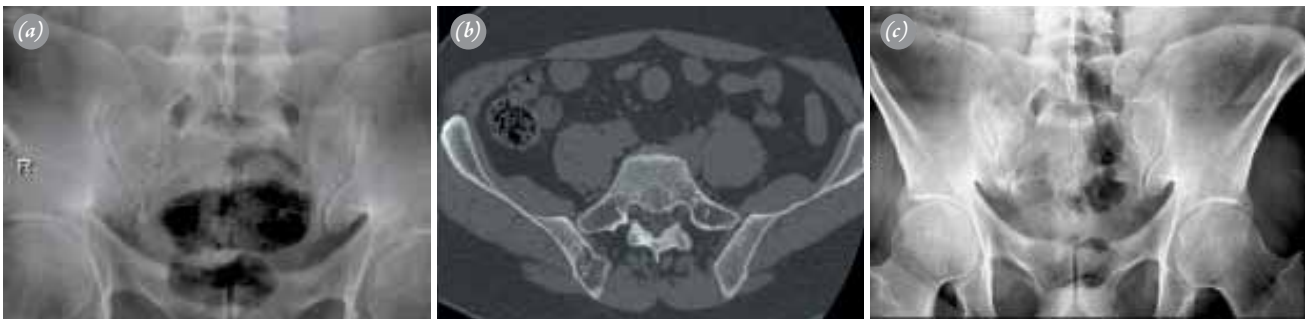

Fig. 2. Forty-four years old male admitted with pain in the right sacroiliac region. **(a)** Anteroposterior radiograph demonstrates a cystic bone lesion in the right iliac bone adjacent to the sacroiliac joint. **(b)** Computed tomography image of the lesion. **(c)** After intralesional curettage and grafting, there was no radiological sign of recurrence on the anteroposterior radiograph of the pelvis taken at the 29th month follow-up.

position under general anesthesia. After the lesion was detected with fluoroscopy, a cortical window was opened over the posterior iliac crest. Intralesional curettage was performed using a high-speed burr. Following adjuvant treatment with phenol and thermal cauterization, the bone defect was filled with autogenic or allogeneic bone grafts. Average intraoperative blood loss was 180 cc. Patients were allowed to mobilize at the 2nd postoperative day with partial weight-bearing as tolerated. Follow-up visits were performed periodically at 6 to 12 month intervals. Histological examination of the curettage materials revealed benign cystic bone lesion in all patients.

There was no pathological fracture or intraoperative major neurovascular damage. Early superficial wound infection was treated with debridement and parenteral antibiotics in one patient. Deep wound infection was not seen in any patients. Two patients reported permanent hypoesthesia around the incision through the gluteal region.

Full weight-bearing was allowed for patients at the 6th week of surgery. All patients reported relief in pain and limp. After an average follow-up of 30.7 months, there was no clinical worsening or radiological recurrence in any patient (Table 2).

Table 1. Differential diagnosis of sacroiliac joint-related pain.

- Trauma
- Piriformis syndrome
- Hip joint pathology (fracture, avascular necrosis, degenerative arthritis)
- Discogenic pain
- Zygapophyseal joint pain
- Rheumatoid arthritis
- Ankylosing spondylitis
- Pregnancy
- Myofascial pain
- Trochanteric bursitis
- Reflecting visceral pain
- Malignancy
- Radiculopathy
- Sports-related pain

Discussion

While many clinical conditions have been described in the differential diagnosis of sacroiliac joint-related pain, benign cystic bone lesions have not been included (Table 1). Generally, pelvic bone tumors are malignant.[8] In contrast, benign cystic tumor-like lesions of the bone mostly involve the long bones of the upper and lower extremities. Zehetgruber et al. reported a prevalence of 0.32/100000 for aneurysmal bone cysts and 0.30/100000 for simple bone cysts.[9] The pelvic bone is a very rare anatomic location for these infrequent lesions. Unni reported iliac bone involvement in 8% of 289 patients with aneurysmal bone cyst.[10] Again, Mankin et al. reported 13 patients with pelvic lesion among 150 patients with aneurysmal bone cysts.[11] Pelvic bone involvement of approximately 2% percent has been reported among simple bone cysts.[12,13] The posterior portion of the iliac bone adjacent to the sacroiliac joint is a very rare anatomic localization for cystic bone lesions. Papagelopoulos et al. stated that only 5 of 40 patients with aneurysmal bone cysts involved the pelvic Zone 1 according to Enneking and Dunham.[14] In another study, the lesion was localized in the posterior part of the iliac bone in 5 of 16 patients with simple bone cysts.[15]

Due to the insidious onset and clinical symptoms of mild pain, there is a high possibility of diagnostic delays with pelvic bone tumors.[8] Sim et al.[16] stated that clinical symptoms of primary bone lesions of the pelvis can be easily confounded with lumber disc hernia. In this study, symptoms lasted for a minimum of three months and all patients were prediagnosed with lumber disc hernia and were treated with conservative methods.

Table 2. Summary of the patients included to the study.

Patient no.	Age (years)	Gender	Side	Lesion width (cm)	Imaging technique	Graft type	Follow-up time (months)	Complications
1	40	M	Left	6	MRI	Allograft	37	Hypoesthesia
2	44	M	Right	5	CT	Autograft	29	None
3	52	F	Left	8	MRI	Allograft	25	None
4	43	M	Right	4	MRI	Allograft	40	None
5	54	M	Left	7	MRI	Allograft	34	None
6	39	F	Left	8	MRI	Allograft	33	Superficial infection
7	27	F	Right	5	CT	Autograft	26	None
8	35	M	Left	11	MRI	Autograft	31	Hypoesthesia
9	29	F	Left	5	MRI	Allograft	21	None

CT: Computed tomography; MRI: Magnetic resonance imaging.

The three-dimensional complex pelvic anatomy, overlapping of anatomical structures in the two-dimensional radiographic methods and artifacts of bowel gas can complicate diagnosis and lead to the bone lesions being overlooked. Therefore, CT and MRI are more effective for the evaluation of the localization, soft tissue components, content and joint extension of the lesion.[17,18] Furthermore, MRI plays an important role in the differential diagnosis of cystic tumor-like lesions of the bone.[19,20] In this study, CT and MRI were used for detailed radiological examination.

Treatment of simple bone cysts is controversial. Several treatment methods have been described, including conservative methods, bone marrow injections, curettage + grafting and internal fixation.[21-23] Although these treatment methods have similar success rates, curettage and grafting is accepted as the definitive treatment method for simple bone cysts staged as active lesions.[24,25]

Aneurysmal bone cysts are staged as benign, local aggressive lesions. Current treatment method is extended curettage, local adjuvant treatments and bone grafting, however, local recurrence rates of up to 20% have been reported.[10,26] Local recurrence risk is higher in the younger population.[27] Benign cystic tumor-like lesions are more common in the first two decades of life and generally resorb or became inactive following skeletal maturity. Because all 9 patients included in this study were in their third decade or older, the cystic bone lesions can be staged as latent lesions according to Enneking.[28] On the other hand, because symptoms were present for at least three months and did not respond to conservative methods and lesions were close to the weight-bearing joint patients were treated with surgical intervention. Extended intralesional curettage, local adjuvant treatment with phenol/thermal cauterization and bone grafting were performed. Minor complications included superficial joint infection in one patient and hypoesthesia around the incision and gluteal lesion in two. There were no major neurovascular or septic complications. After an average of 30.7 months of follow-up, a significant relief in symptoms was reported in all patients and there were no radiological signs of recurrence.

In conclusion, benign cystic bone tumors and tumor-like lesions should be kept in mind in the differential diagnosis of monolateral sacroiliac joint-related pain. Conventional radiography can be inadequate in diagnosis due to the complex anatomy of the pelvis. Suspicious lesions in this area must be investigated with detailed radiological methods such as CT or MRI. In case of failure in conservative methods, such lesions can be safely and effectively treated with extended intralesional curettage, local adjuvant treatments and bone grafting.

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