

Vacuum phenomena in insufficiency fractures of the sacrum

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Abstract. *Objective.* Insufficiency fractures of the sacrum are found in women who have undergone radiation therapy to the lower abdomen as well as those suffering from osteoporosis of postmenopausal, steroid-induced, or primary biliary cirrhosis-related origin. Increased uptake in bone scintigraphy and osteolytic changes in these fractures can be misinterpreted as bone metastases, leading to unnecessary biopsies and other procedures in the ensuing search for non-existent primary tumor. *Patients.* In eight female patients averaging 69.4 years of age, insufficiency fracture of the sacrum was diagnosed by computed tomography (CT) and bone scintigraphy. Three underwent a total of five MRI examinations. Malignancy was excluded by histology in two patients and follow-up of at least 6 months in the remainder. Retrospective analysis of CT scans of 13 patients with metastases in the sacrum revealed no vacuum phenomena. *Results.* In seven of eight patients with insufficiency fracture of the sacrum, vacuum phenomena were shown on CT examination. The gas was localized centrally within the ventral part of the fracture in three patients; gas was located in ten adjacent sacro-iliac joints of six patients. *Conclusions.* The vacuum phenomenon may be an incidental finding in osteoarthritis of the sacro-iliac joint, but it has not been previously recognized in IFS. The presence of intra-articular vacuum phenomena in the sacro-iliac joints in combination with a sacral fracture and vacuum phenomena located within the sacral fracture supports a diagnosis of insufficiency fracture or may indeed be the clue by which this diagnosis is established.

Key words: Computed tomography – Insufficiency fractures – Osteoporosis – Sacro-iliac joint – Radiation, complications – Sacral bone

Insufficiency fractures of the sacrum are a well-known complication in women who have undergone radiation therapy of the pelvis or are suffering from postmenopausal, steroid-induced, or primary biliary cirrhosis-related osteoporosis [1–11]. These fractures are characterized by increased activity on radionuclide bone scans. The lateral masses of the sacrum contain large amounts of hematopoietic bone marrow and are therefore often the site of bone metastasis. Increased uptake in bone scintigraphy in lateral masses of the sacrum and osteolytic destructions on radiographs and computed tomography (CT), especially in patients with a history of a tumor elsewhere, are suggestive of skeletal metastasis, prompting biopsies and additional diagnostic procedures to find a primary tumor [2]. We present seven out of eight patients with insufficiency fractures of the sacrum, in whom we observed vacuum phenomena within the fracture or in the adjacent sacroiliac joints, indicating the benignity of the lesion. This phenomenon has not been recognized previously.

Patients and methods

Eight patients with insufficiency fractures of the sacrum were treated in our clinic within the last 3 years. All were women; their average age was 69.4 years (range 55–84 years). Two of the patients had a neoplastic history and had received radiation therapy to the pelvis, two suffered from primary biliary cirrhosis-induced osteoporosis, two had postmenopausal osteoporosis, and one patient had had a total hip replacement 4 months earlier. One patient had been suffering from rheumatoid arthritis for 35 years with subsequent steroid-induced osteoporosis; in one woman osteoporosis was not recognized until the sacral fractures appeared.

All patients were examined by repeated radiographs, bone scintigraphy, and CT. For three women, follow-up CT-studies were available. Three of the patients also underwent a total of five magnetic resonance imaging (MRI) examinations. Biopsy was performed in two cases. All patients were followed from 7 months to 2 years. Every patient underwent thorough tumor screening, but neither primary tumors nor metastatic lesions were found.

All of our patients with insufficiency fractures of the sacrum suffered from local pain, which was most severe when pressure was applied to the sacrum. Two patients presented with saddle an-

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esthesia, numbness of both legs, urinary and bowel incontinence, but no motor neurological deficits. All patients had increased serum alkaline phosphatase levels of 200–500 U/l.

CT examinations of 13 patients with proven metastasis to the

sacrum were analyzed retrospectively. We reviewed patients who received radiation therapy to the sacrum and had CT examinations prior to radiation therapy. No gas enclosures in the lytic areas or sacro-iliac joints were found in any of these 13 patients.

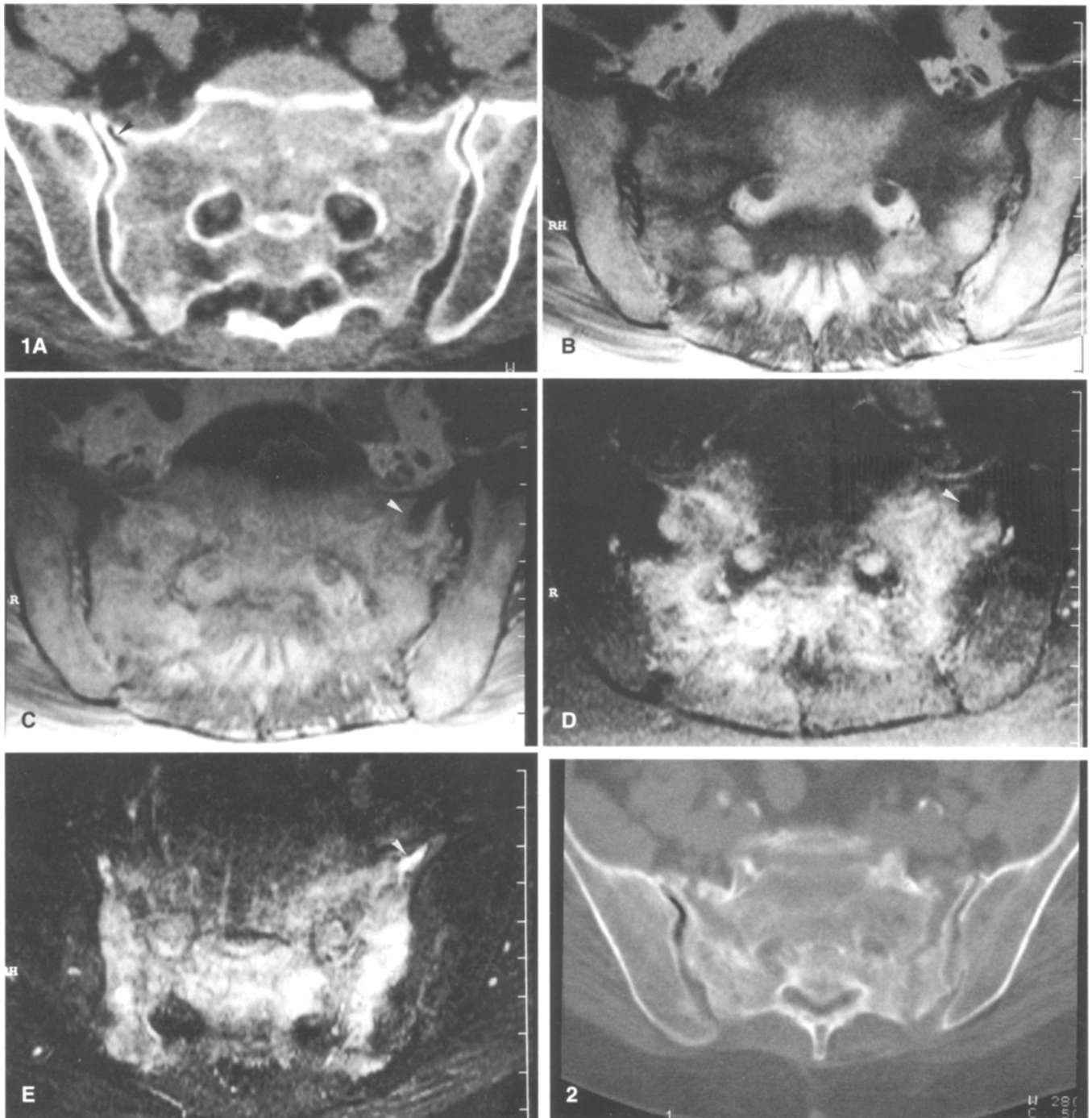


Fig. 1. A Computed tomography (CT) scan shows vacuum phenomenon in the ventral part of the right sacral bone with irregular sclerosis of the lateral masses. B T1-weighted magnetic resonance (MR) image (570/15 ms) exhibits low-intensity areas in both lateral masses of the sacrum and the central portion between the neural foramina. C After gadopentetate administration enhancement of these areas is shown. D Enhancement is demonstrated to good advantage by the frequency-selective fat-suppressed T1-weighted image (602/15 ms). High-signal areas

represent contrast material uptake. Free water in the developing ventral fracture gaps shows no gadopentetate uptake. E These areas but display high signal on the STIR image (150/3800/50 ms)

Fig. 2. CT scan of a patient with bilateral sacral fractures. The fracture lines extend back from the ventral portion of the sacrum close to the sacro-iliac joint posteriorly. Vacuum phenomenon in the right sacro-iliac joint

Results

On CT, seven of our eight patients showed bilateral fractures of the lateral masses of the sacrum. In addition, in one patient a horizontal fracture was found which extended between the two vertical fracture lines. Four of these fractures were detected by bone scintigraphy. All patients showed increased uptake at the fracture site; in six patients with bilateral fractures, bone scans demonstrated a characteristic H-like pattern independent of the presence of a horizontal fracture line. In one patient no evidence of a fracture was present initially on CT, in spite of increased uptake in bone scintigraphy as characteristic of a fracture. The fractures in the remaining seven patients were visible on CT; in one of these patients fracture lines only were present at the most ventral portion of the sacrum on both sides on CT and gadopentate-enhanced MRI (Fig. 1B–E). In this patient the whole sacrum was irregularly structured with osteosclerotic areas on CT (Fig. 1A) and severe bone marrow edema on MRI (Fig. 1B–E). In four patients the fractures were initially diagnosed by CT. On conventional radiographs, the fractures of two patients could not be diagnosed earlier than 3 months after positive bone scans were found.

As visualized by CT, the fracture lines extended from the ventral portion of the sacrum close to the sacroiliac joint posteriorly and nearly parallel to the sacroiliac joint. In 11/15 fractures, the posterior course of the fracture line extended close to the S2 neural foramina. The fracture lines of the patients with neurological symptoms extended into the S2 neural foramina. In one patient, a horizontal fracture connected the bilateral sagittal fractures. In three patients, gas was found on CT within the anterior region of the fracture, (Figs. 1A, 3A, 4). In six patients, including two with gas inside the fracture, we found vacuum phenomena in sacroiliac joints adjacent to the fracture lines. The gas collections were bilateral in four of these patients and unilateral in two (Fig. 2). These gas accumulations were not visible on conventional films or MRI.

MRI was performed in three patients. In two patients, the appearances were initially interpreted as compatible with metastasis (Figs. 1, 3). These two patients underwent biopsy. However, in view of the final diagnosis of insufficiency fracture, we conclude that the signal alterations represent prominent bone marrow edema.

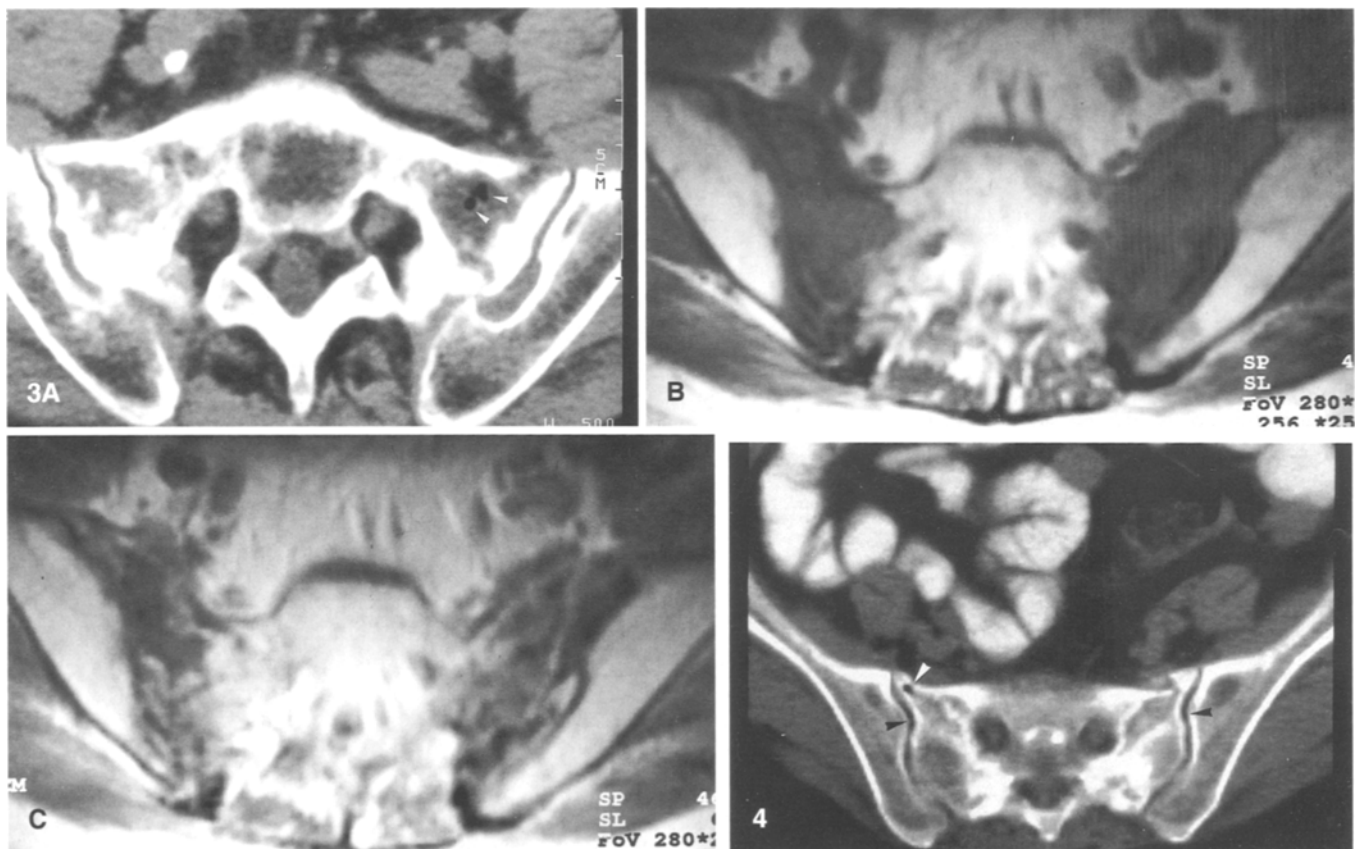


Fig. 3. A On the CT scan of this patient the fractures appear as large destructive areas. Gas is present in the fracture on the left side (*arrowheads*). T1-weighted (TR/TE=500/15 ms) MR images before (B) and after (C) intravenous administration of gadopentetate 0.1 mmol/kg b.w. Low-signal areas compatible with bone marrow edema extend far beyond the fracture lines. Moderate inhomogenous contrast enhancement can be seen

Fig. 4. Bilateral insufficiency fractures of the sacrum. CT scan with soft tissue window setting, exhibiting gas in the anterior part of the right insufficiency fracture (*white arrowhead*) and in both sacro-iliac joints (*black arrowheads*)

Discussion

Postmenopausal osteoporosis, steroid osteoporosis, and radiation therapy to tumors of the pelvis are the most important causes of insufficiency fractures of the sacrum [1–11]. As yet, this type of insufficiency fracture is found only in female patients. Stress fractures of the sacrum, on the other hand, are found in patients with normal bone structure and are dominated by reparative processes and sclerosis. They are reported in long distance runners and army recruits; six cases have been published up to the present [12–14].

Insufficiency fractures of the sacrum may pose problems in differential diagnosis. Increase in tracer uptake is regularly found in bone scintigraphy [15, 16]. All our patients had increased alkaline phosphatase serum levels as well. If increased uptake in the region of the lateral mass of the sacrum is found in follow-up of patients with malignant tumors, metastasis has to be ruled out. Insufficiency fractures of the sacrum are characterized radiographically by bony sclerosis and inhomogeneities of the lateral mass, and the fracture lines may not be readily identified.

In three of our patients with insufficiency fractures of the sacrum, gas inclusions were found unilaterally in the ventral portion of the fracture line within the lateral mass of the sacrum (Figs. 1A, 3A, 4). The stresses that cause the fractures in the first place prevent healing, promoting separation with release of gas at the fracture site. All insufficiency fractures have a tendency to non-union with formation of pseudoarthroses [17]. Malignant infiltration of the marrow with bone destruction does not produce a vacuum phenomenon. In our control group no pathological fracture of the lateral mass of the sacrum was found in spite of the presence of metastatic lesions, and no gas collection was found.

In six of our patients, vacuum phenomena were also found in the sacro-iliac (SI) joints, two unilaterally and four bilaterally. The increased mobility in the posterior ring of the pelvis in the presence of insufficiency fractures of the sacrum favors gas formation in the sacro-iliac joint. In no case of sacral metastasis was a sacro-iliac vacuum phenomenon detected. However, vacuum phenomena within the sacro-iliac joint can also be found in degenerative disorders of the sacro-iliac joint. Even if vacuum phenomena cannot prove the benignity of lytic areas within the sacrum, gas within the fracture gap or the SI joint is suggestive of insufficiency fracture of the sacrum.

Spinal vacuum phenomena are related to accumulation of nitrogen within crevices in the intervertebral disk or vertebra. Vacuum phenomena also are found occasionally in osteoarthritic joints. The presence of gas in a disk is probably related to a degenerative or osteonecrotic process [18–20]. Occasionally, in patients with vertebral compression fracture secondary to tumor, vacuum phenomena will appear within adjacent disks [21] or within the affected bone [17, 18, 22]. In fact, these are rare cases and the presence of gas in a vertebra is generally, related to a non-neoplastic etiology. No case has been reported with gas in a tumor-related sacral fracture.

In reviewing original articles on insufficiency fractures of the sacrum we found CT scans of 17 patients reproduced [1–3, 5, 11, 16, 23, 25]. Only six images failed to show any vacuum phenomenon. Gas enclosures were visible in ten sacro-iliac joints and questionable in another three. Vacuum phenomenon in the ventral portion of a sacral insufficiency fracture is evident in one patient from the series from Rafii et al. [23] and probably in patients reported by Blomlie et al. [2], De Smet and Neff [5], and Rafii et al. [23]. As only one CT section from each every patient is reproduced, some vacuum phenomena have probably been missed or are not shown due to window level settings or quality of reproduction.

The single most important etiological factor for insufficiency fractures of the sacrum is previous radiation therapy, which makes the bone more fragile [1, 2, 6, 8, 23]. Characteristically, these fractures occur bilaterally, but not necessarily at the same time. In one our patients, bilateral fractures were recognized within a period of 3 months (Fig. 1). Insufficiency fractures of the sacrum can lead to neurological symptoms [7, 24]. Two of our patients developed severe neurological symptoms over a period of 2 and 5 weeks respectively. Symptoms included paresthesia of both legs and loss of sensitivity of the bladder and rectum with urinary and bowel incontinence; eventually, ambulatory capacity was impaired due to severe back pain and weakness of both legs. An increase in erythrocyte sedimentation rate and cerebrospinal fluid protein suggested an inflammatory or post-inflammatory process, but was presumably caused by mechanical effects on the nerve roots. Similar cases with neurological deficits as prominent complaints have been published [7, 24].

Other suggested causes of insufficiency fractures of the sacrum, include Tarlov cysts [25]. We found Tarlov cysts in two of our patients bilaterally in the region of S1 and S2. A decrease of the mass of the trabecular bone and the eburnated and rigid border of the cysts may decrease the elasticity and contribute to insufficiency fractures of the sacrum. The precise mechanism and relevance of Tarlov cysts in the etiology of insufficiency fractures of the sacrum, however, has not yet been elucidated and the fracture line does not pass through the cysts but through trabecular bone adjacent to them. Both of the patients with Tarlov cysts had chronic liver disease with an increased risk of developing osteoporosis [26]. The incidence of spontaneous fractures following orthotopic liver transplantation has been estimated to be 40% in primary biliary cirrhosis [27], and one of the three patients with sacral insufficiency fracture after liver transplantation in the series of Peris et al. [28] suffered from this disease. Patients with primary biliary cirrhosis and sacral fractures before liver transplantation have not been previously reported. One of our patients underwent liver transplantation and the bilateral sacral fractures healed within a period of 4 months.

Damage to the bone by previous radiation therapy or in osteopenia results in an atrophic type of reaction to repetitive microtrauma, and large osteolytic areas can develop (Fig. 3). MRI may be misleading, because the fracture lines are not clearly demarcated and adjacent

edema with signal alterations and contrast enhancement simulate neoplasm [11, 29]. In two of the three patients who underwent MRI, biopsy was performed. On histologic examination, bone marrow edema and reduction of hematopoietic bone marrow were found, which was totally absent in the patient with previous irradiation.

Conclusions

Frequently, insufficiency fractures of the sacrum present problems in differential diagnosis, particularly when the lesions are found in tumor patients and are associated with prominent bone destruction. Presence of gas in the sacro-iliac joints in combination with a sacral fracture supports a diagnosis of a benign lesion, but it may be an incidental finding in unassociated osteoarthritis of the sacro-iliac joint, thus not proving conclusively the benignity of the lesion. A vacuum phenomenon located within the sacral fracture supports the diagnosis of an insufficiency fracture or may be the clue by which this diagnosis can be established.

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