Sacral fatigue fractures in children with sacral spina bifida occulta

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In this report, we present two cases of 9-year-old children with spina bifida occulta (SBO) of the sacrum, who were diagnosed with sacral fatigue fractures. In both patients, MRI showed a linear signal void and high signal in sacral ala on the short tau inversion recovery sequence. Sacral SBO at the same level of the sacral fracture was observed in each patient on computed tomography images. These lesions healed with rest. This is the first literature reporting cases with sacral stress fractures who had SBO at the same level of fracture. *J Pediatr Orthop B* 25:278–282

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Introduction

Nontraumatic sacral fractures can be characterized as insufficiency fractures in elderly populations and fatigue fractures in younger populations and athletes [1–3]. Insufficiency fractures occur in osteoporotic patients or in patients who have undergone radiation therapy for pelvic malignancy. Fatigue fractures occur in physically active individuals, such as long-distance runners, in whom repetitive stress loads occur [4]. Several reports have described their occurrence in athletes who engage in vigorous exercise. However, sacral fatigue fractures in children are very uncommon [4–6], especially in those younger than 10 years of age.

Here, we present two cases of 9-year-old children diagnosed with sacral fatigue fractures.

Case reports

The authors obtained informed written consent from the patients' parents to publish this case report.

Case 1

A 9-year-old baseball player presented with a 1-month history of right low back and buttock pain during baseball. He had just returned to baseball after cessation of sports activity because of osteochondritis dissecans of the left knee. MRI showed signal changes in the right sacral ala. He was referred to our department for further workup of his low back and buttock pain.

The patient had no paresthesia, weakness, fever, bowel or bladder dysfunction, or weight loss. He had no history of previous pelvic disease, and family history was nonspecific. No history of trauma was reported. Physical examination indicated local tenderness over the right iliac crest and slight limitation in the range of motion in the right hip; however, no areas of decreased sensation or leg-length discrepancy were noted. Pain was exacerbated by activity and relieved on rest.

Plain radiographs of the pelvis showed no abnormal findings such as osteolytic or sclerotic changes (Fig. 1). MRI showed a linear signal void in the right S1 ala on both T1-weighted and short tau inversion recovery (STIR) coronal images. Further, there were low signal intensity changes on T1-weighted images and high signal changes on STIR surrounding the linear signal void (Fig. 2a and b). A linear sclerotic change was observed at the same area on computed tomography, and spina bifida

Fig. 1

Unremarkable initial anterior plain radiograph of the pelvis.

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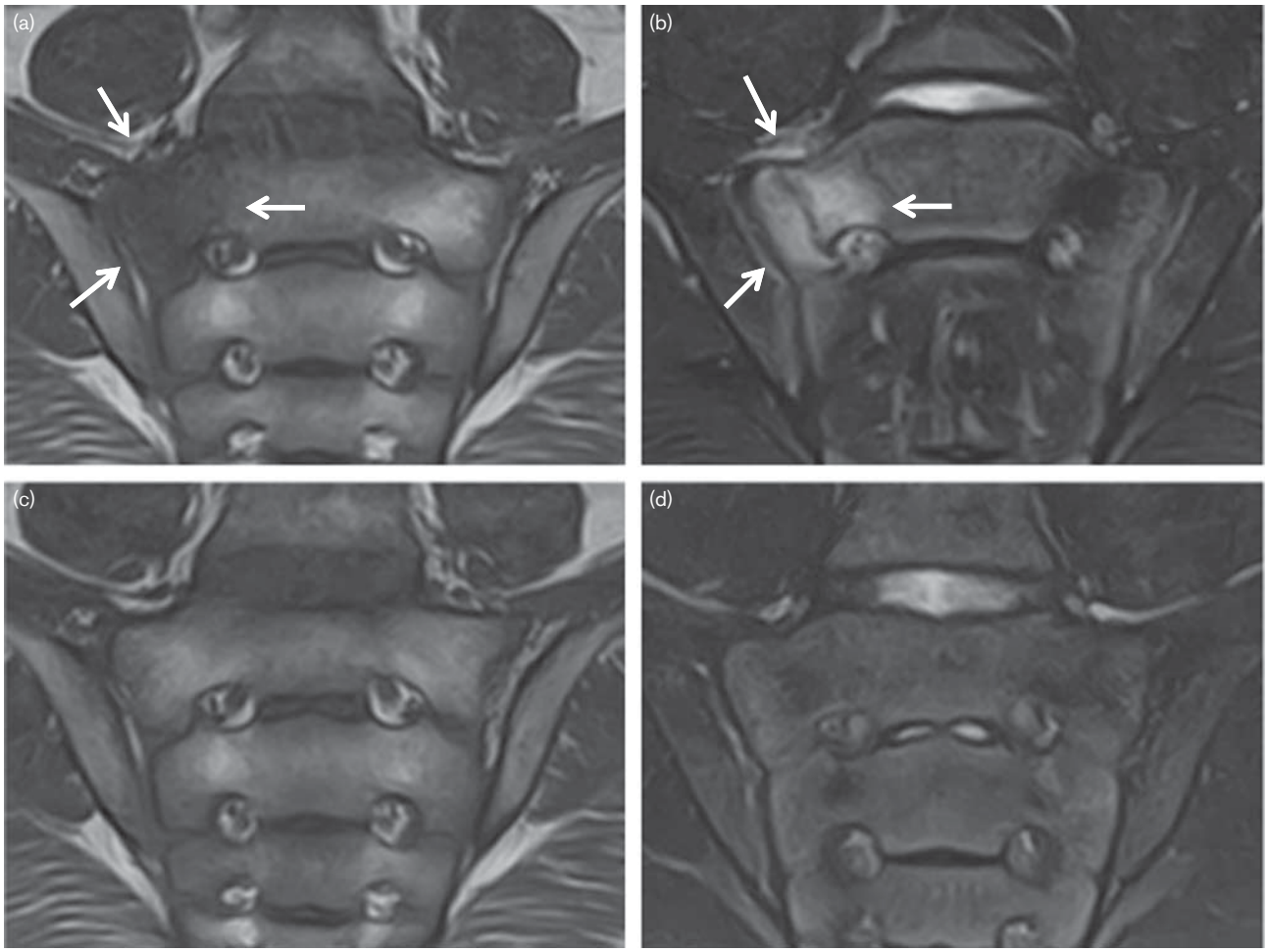
Fig. 2



Coronal MR images in case 1. (a, b) Initial examination. (c, d) Three months later. Coronal images showing (a) diffuse low marrow signals with a central linear signal void (arrows) in the T1-weighted image and (b) diffuse increased signals in the STIR image at the right S1 ala (arrows). (c, d) The low signals in the T1-weighted image, high signals in the STIR image, and the linear signal void are no longer evident 3 months later. STIR, short tau inversion recovery.

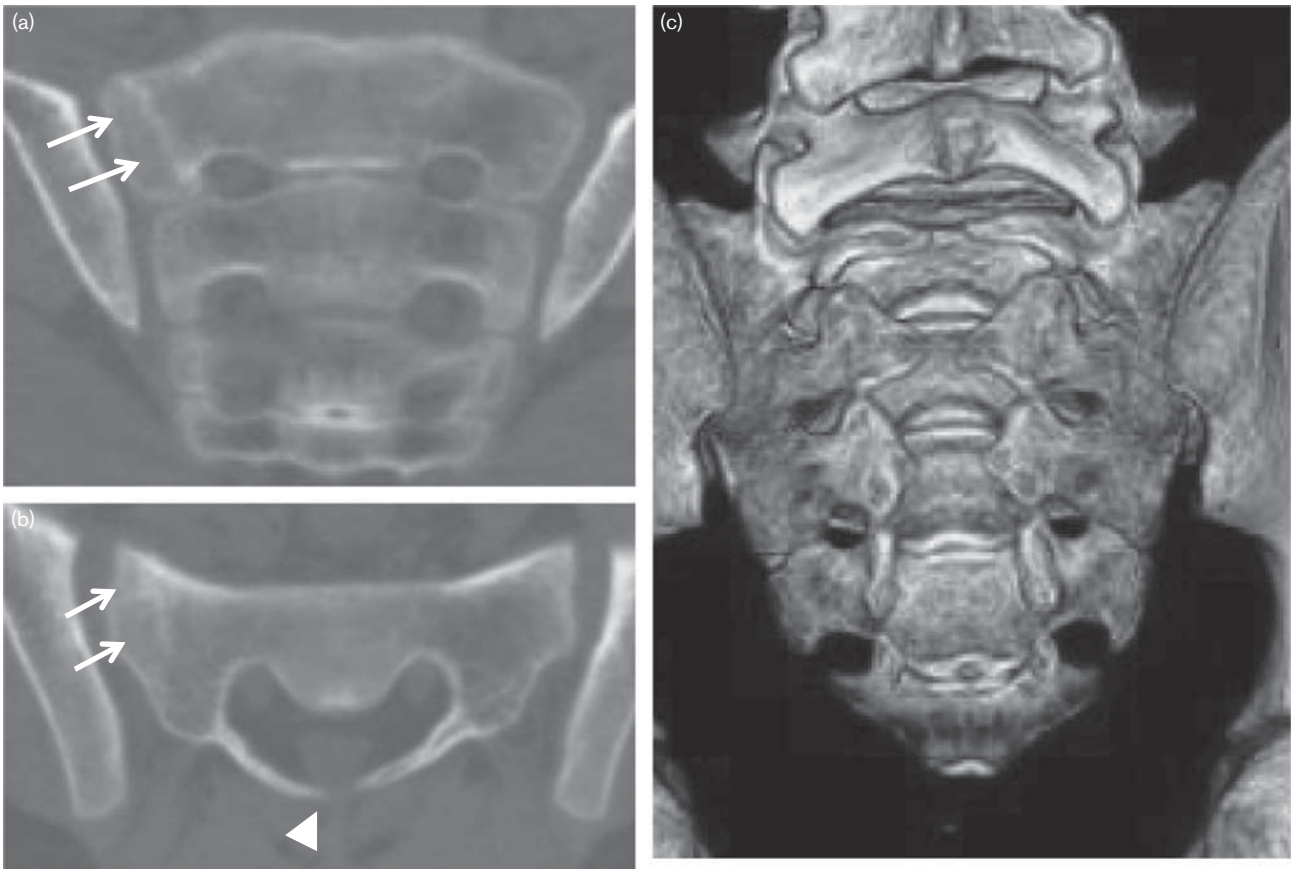
occulta (SBO) was noted from S1 to S5 (Fig. 3). A diagnosis of sacral fatigue fracture was made on the basis of both clinical and imaging data. We advised the patient to withdraw from all sports activities for 6 weeks. Three months after symptom onset, pain was improved and signal changes in the right S1 ala had disappeared on MRI (Fig. 2c and d).

Case 2

A 9-year-old male baseball player experienced pain in the left hip after playing baseball. He stopped playing baseball and rested following onset. No history of paresthesia, weakness, fever, bowel or bladder dysfunction, or weight loss was noted. His previous medical history and family history were nonspecific, and there was no history of trauma. MRI showed signal intensity changes in the left sacral ala. Three weeks after symptom onset, he was referred to our department for further workup of the persistent left hip pain. However, the pain had already improved upon presentation to us. Physical examination indicated no tenderness over the paraspinal region, buttocks, low back, or femoral head. Both his lower extremities showed full range of motion and full motor strength, and there were no areas of decreased sensation or leg-length discrepancy. The FABER-Patrick test was negative on both sides. His reflexes were symmetric, bilateral, and normal. He could stand on one leg without pain.

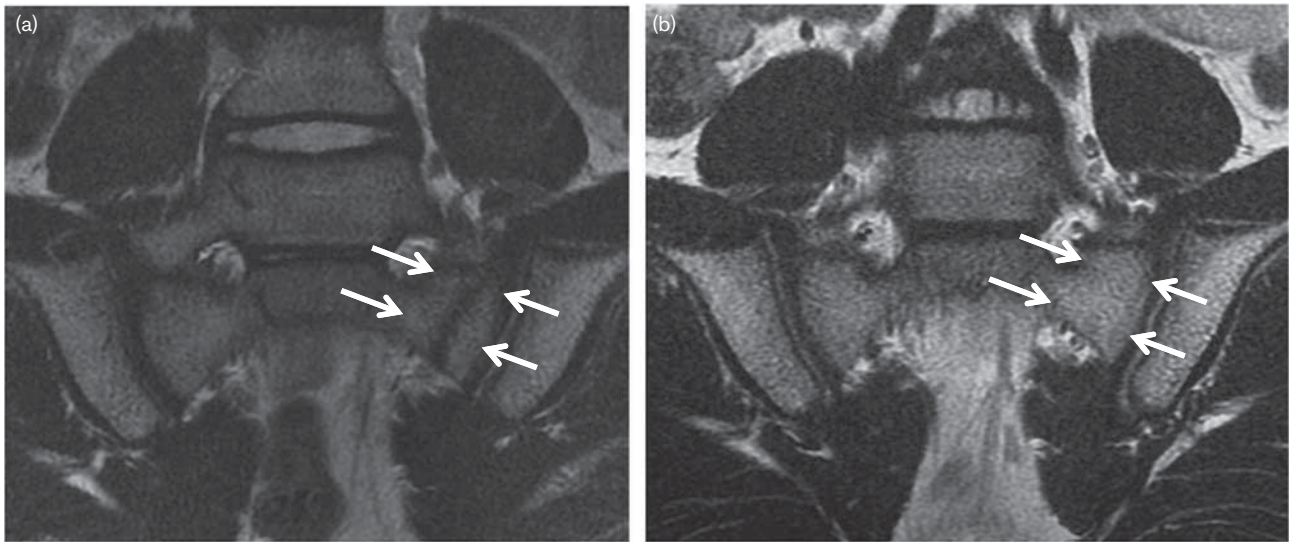
Plain radiographs of the pelvis were normal. MRI showed a linear signal void on the T2-weighted coronal image in the left S2 sacral ala (Fig. 4a). Computed tomography indicated a linear band of medullary sclerosis at the same site and SBO was noted at S2 (Fig. 5). This patient had a transitional vertebra in the lumbosacral region (Fig. 5c). The diagnosis of sacral fatigue fracture was made.

Fig. 3



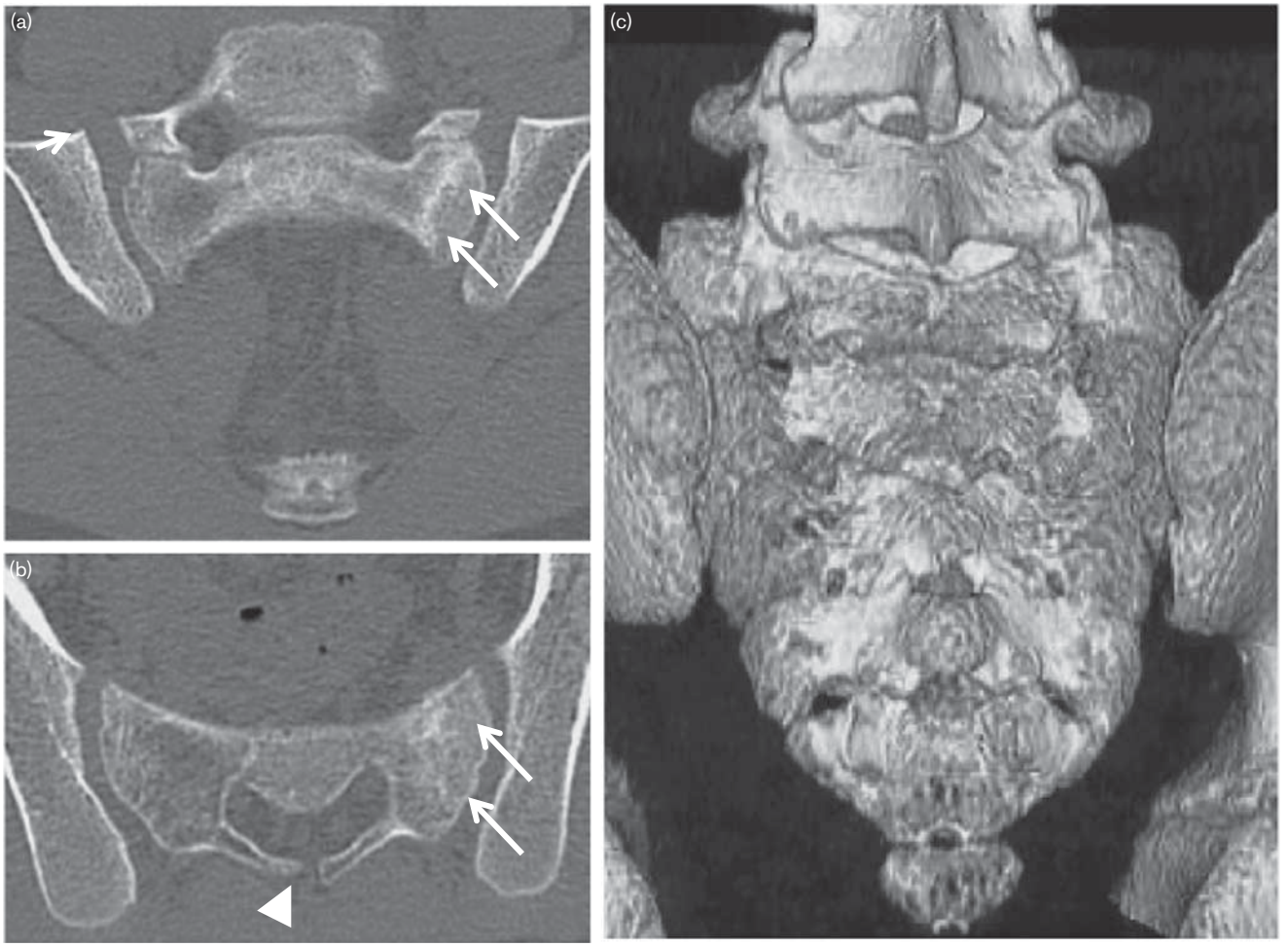
(a) Coronal and (b) axial CT images showing linear medullar sclerosis in the right S1 ala (arrows) and SBO (arrowhead). (c) Three-dimensional-CT image showing SBO from S1 to S5. CT, computed tomography; SBO, spina bifida occulta.

Fig. 4



T2-weighted coronal MR image in case 2. (a) Initial examination. (b) Two months later. Coronal MR images showing the linear signal void (arrows) in the T2-weighted image (a). The linear signal void in the T2-weighted image is no longer evident 2 months later (b).

Fig. 5



(a) Coronal and (b) axial CT images showing linear medullar sclerosis in the left S2 ala (arrows) and SBO (arrowhead). (c) Three-dimensional-CT image showing SBO at S2. CT, computed tomography; SBO, spina bifida occulta.

Because his pain had already improved, he was allowed to resume sports activities as long as pain was tolerable. Two months later, T2-weighted coronal image showed the linear signal void at the fracture to be unclear and diffuse (Fig. 4b). The signal intensity of the surrounding medulla was essentially normal. The patient had no recurrent pain or complaints and returned to sports activities.

Discussion

Fatigue fractures in children usually occur in the lower extremities, most commonly in the tibia, fibula, or metatarsals [7,8]. Fatigue fractures of the sacrum are less commonly reported, but may be encountered as the result of serious athletic training for long-distance running [9]. To our knowledge, there are only five case reports of fatigue fractures of the sacrum in children aged 12 years or younger in the literature. Sacral fatigue fractures are very uncommon in children and all published reports describe excellent clinical outcomes with conservative treatment (Table 1) [5,7,10–12].

Symptoms of sacral fatigue fractures are low back pain of gradual onset, occasionally radiating to the hip or the groin. Most patients have normal results on neurological examination, and most show full range of motion of the back and lower extremities. With respect to radiological findings, plain radiographs are usually normal as they are not sensitive enough to show sacral fatigue fractures, but may be useful in revealing other causes of back pain. Bone scintigraphy and MRI are effective for diagnosis [1, 2,4–7,9–12]. Bone scintigraphy shows increased activity in the lateral part of the sacrum and MRI shows bone marrow edema as an early sign of a stress fracture. T1-weighted and T2-weighted and STIR images show a linear signal void with surrounding diffuse decreased marrow signals on the T1-weighted images and surrounding high-intensity signals on the T2-weighted and

Table 1 Previous studies on sacral fatigue fracture in children aged 12 years or younger

References	Age (years)	Sex	Activity	Symptom(s) (duration)	Treatment (outcome)	SBO
Grier *et al.* [7]	9	Male	Not mentioned	Pain in left superior sacral, groin, and thigh (4 weeks)	Conservative (full recovery)	Not mentioned
Martin *et al.* [10]	9	Female	Ballet	Left buttock pain (2 months)	Conservative (resolved after 8 weeks)	Not mentioned
Rajah *et al.* [11]	11	Female	Aerobic exercise	Right groin pain (3 weeks)	Not mentioned (not mentioned)	Not mentioned
Lam and Moulton [12]	10	Male	Physical education	Low back pain and right buttock pain (2 weeks)	Bed rest for 1 week (returned to normal over 4 weeks)	Not mentioned
Mangla *et al.* [5]	9	Female	Soccer	Left buttock pain (3 weeks)	Cessation of athletic activities for 1 month (resumed activities after 4 weeks)	Not mentioned

SBO, spina bifida occulta.

STIR images. The radiological findings in our cases were consistent with these features.

In both our patients, SBO was found at each affected level of the sacral fatigue fractures. However, we could find no reports describing the presence of SBO in pediatric patients with sacral fatigue fractures (Table 1) [5,7,10–12]. In addition, athletes typically incur fatigue fractures, but neither patient was a highly enthusiastic athlete. Sakai *et al.* [13] reported a significantly higher incidence of lumbar spondylolysis among patients with SBO than in those without SBO (16.2 vs. 5.0%, respectively. Several studies in the literature have described a positive association between SBO and spondylolysis [14–16]. A recent finite element study showed a bifid arch in SBO and following laminectomy increases load across the isthmus, predisposing toward early fatigue fractures of the isthmus [17]. Similarly, SBO in the sacrum might affect the stress concentration on the sacral alae, predisposing toward stress fractures of the sacrum without repetitive stress.

Conclusion

We have presented two cases of sacral fatigue fractures in 9-year-old patients with SBO at each affected level. Sacral fatigue fracture is an important consideration in the differential diagnosis of lower back and pelvic pain in children and should be considered whenever a healthy, active child presents with unexplained persistent low back and buttock pain.

Acknowledgements

Conflicts of interest

There are no conflicts of interest.

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