

**LETTER TO THE EDITOR****TO THE EDITOR****Spontaneous Atraumatic Bilateral Pedicle Insufficiency Fractures**

**Keywords:** Bilateral pedicle fracture, Bisphosphonate, Spondylolisthesis, Osteoporosis, Lumbosacral spine

Isolated bilateral pedicle fractures in the spine are rare, and an extremely uncommon etiology for spondylolisthesis.<sup>1–3</sup> We report a case of bilateral pedicle stress fractures accompanying spondylolisthesis in a patient taking Risedronate for 10 years. This is the first reported case of spondylolisthesis accompanying spontaneous bilateral pedicle stress fractures involving one vertebra in a patient on bisphosphonate therapy for osteoporosis. Informed consent to write up this case report was obtained from the patient.

A 79-year-old woman presented with progressively worsening low back pain (LBP) for 6 weeks without any traumatic episodes or previous spinal surgery. Her LBP was mechanical in nature; worse with activities but relieved by sitting or lying down. Past medical history showed she suffered from severe osteoporosis and had been taking a bisphosphonate drug (Risedronate; 35 mg once per week) for 10 years. She also had chondrosarcoma of the chest wall with required resection in 2013 and colon cancer with bowel resection in 2009, both with free margins and no signs or symptoms of local or distant recurrence.

A CT scan 6 months before presentation, to follow-up on benign cystic lesions in her liver, demonstrated no significant vertebrae abnormalities (Figure 1). A further CT scan to investigate a 6-week history of atraumatic mechanical back and flank pain demonstrated new bilateral L5 pedicle fractures (Figure 2), which prompted her referral for consultation with a spine surgeon.

On physical examination, she was able to stand and walk without assistance, although her LBP increased gradually in the standing position. She was neurologically intact. At presentation, she rated her pain as 8/10 on a numerical pain rating scale and scored 43/50 on the Oswestry Disability Index (ODI).

Standard X-ray imaging of the lumbar spine (i.e., lateral, anterior–posterior, and flexion/extension views) showed stable grade 1 spondylolisthesis at L5-S1 and bilateral L5 pedicle fractures. There was mild degenerative scoliosis but we do not think this was relevant because of the bilateral nature of the injury. Subsequent MRI (Figure 3) confirmed these X-ray imaging findings and, together with a technetium-99m methyl diphosphonate bone scan, found no local or distant findings that would suggest tumor involvement. Dual-energy X-ray absorptiometry T-scores for bone mineral density of the femoral neck and lumbar spine were  $-2.5$  and  $-2.7$ , respectively.

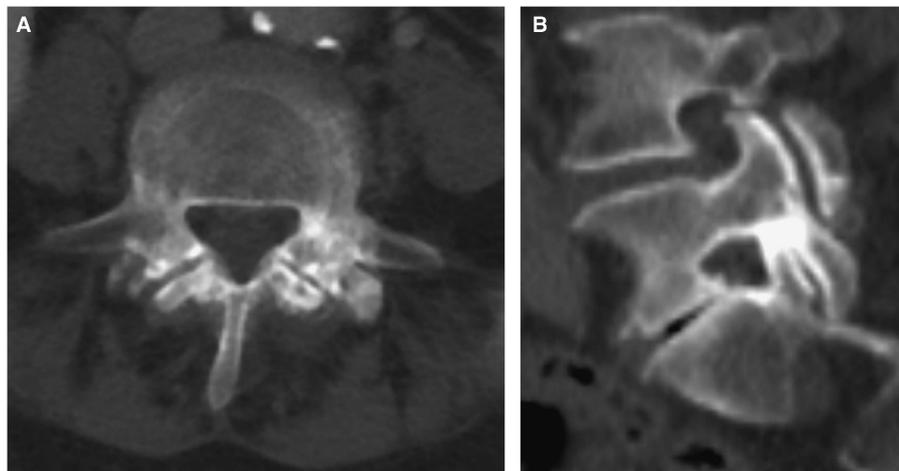
Options for treatment including surgery versus brace versus observation were discussed with the patient. After hearing the costs, risks, and benefits of each option, she chose conservative therapy.

She was initially managed with advice to remain active within pain tolerance, reassurance, and over-the-counter analgesia. She was referred to an endocrinologist with specialization in osteoporosis. Her Risedronate was stopped and she was started on Denosumab as well as calcium and vitamin D supplementation.

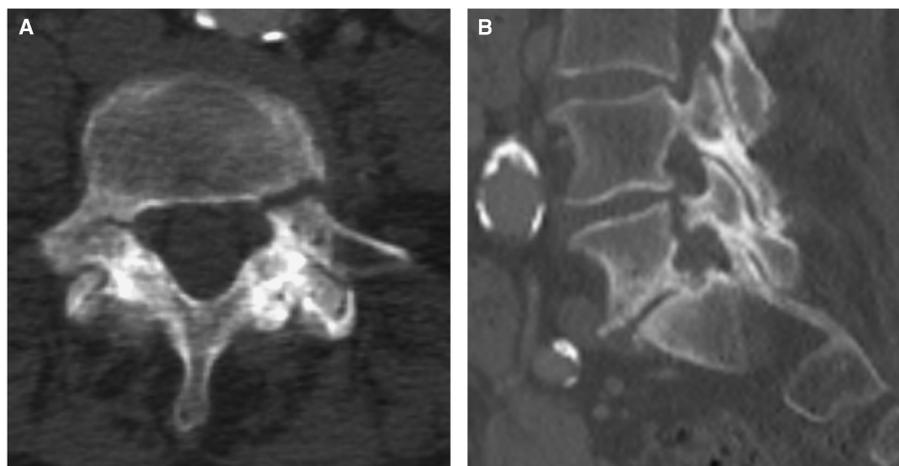
Thereafter, she was followed with serial X-rays which demonstrated stability of the listhesis, and she was prescribed physiotherapy. At 1-year follow-up, her pain score was lowered to 5/10 and her ODI score was improved to 31/50. X-rays demonstrated no progression of the listhesis (Figure 4). A CT scan performed 2 years after initial presentation demonstrated healing of the bilateral pedicle fractures (Figure 5).

There are two types of stress fractures: (1) fatigue fractures which occur in normal bone that is subjected to repetitive abnormal stress and (2) insufficiency fractures which are due to normal stress on a structurally defected bone. Pediculolysis is a fatigue-type stress fracture.<sup>4</sup> An *in vitro* study applying mechanical stress on vertebrae showed that pedicle is the second weakest part of the vertebra after pars interarticularis.<sup>5</sup>

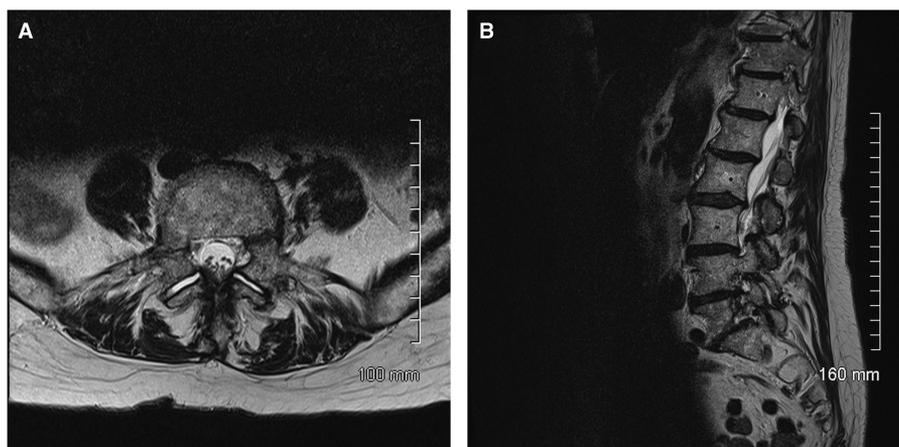
Defects within the pedicle of the lumbar spine may be congenital, traumatic, iatrogenic, or pathologic.<sup>6</sup> They can present as a unilateral pedicle fracture with contralateral pars lysis,<sup>4,7</sup> it can



**Figure 1:** (A,B) Axial and sagittal CT scan of the abdomen performed 6 months before presentation. No fractures or spondylolisthesis noted.



**Figure 2:** (A,B) Axial and sagittal CT scan of the abdomen performed before presentation. Demonstrating bilateral L5 pedicle fractures.



**Figure 3:** (A,B) Axial and sagittal T2 weight MRI scan of the lumbar spine performed at presentation. Demonstrating bilateral L5 pedicle fractures with grade 1 spondylolisthesis at L5-S1, and no findings suggestive of tumor involvement.

happen in one or more levels in the lumbar spine,<sup>8,9</sup> or they may be associated with spondylolisthesis.<sup>2,3</sup> In younger adults the diagnosis is extremely rare and is nearly always associated with sports in which hyperextension is common (e.g., ballet), whereas in older adults it is usually due to non-diagnosis of a defect from adolescence presenting later with associated LBP.<sup>10,11</sup>

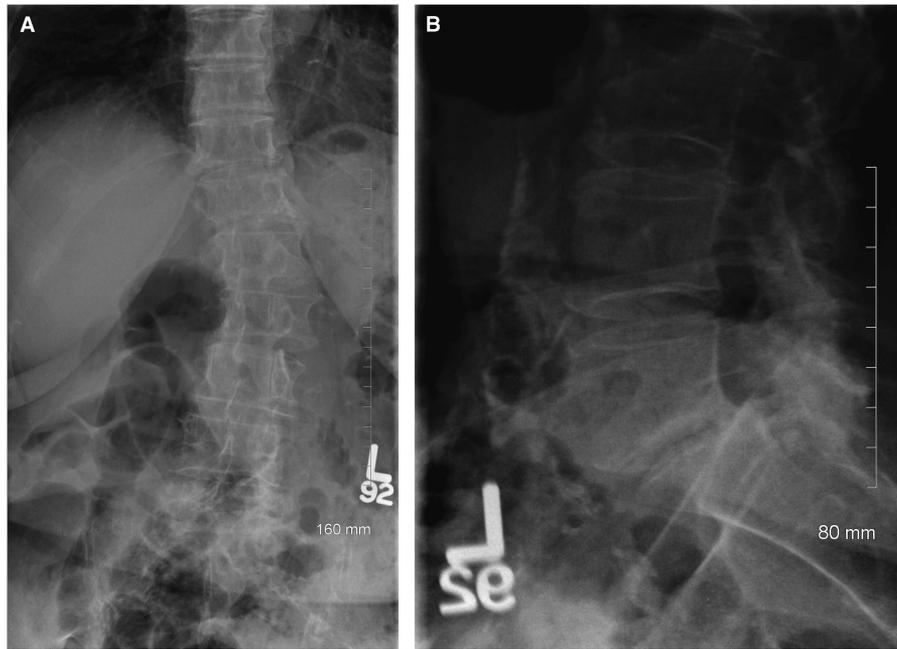
Pedicle stress fractures may present as an acute traumatic fracture or have signs of established pseudoarthrosis.<sup>12</sup> Gunzburg and Fraser<sup>7</sup> introduced the term pediculolysis in the 1990s and presented a case of a unilateral stress fracture of the pedicle associated with a contralateral pre-existing spondylolysis. In contrast, we are presenting a case of isolated bilateral pedicle stress fractures that was not present on CT scan 6 months before presentation. Previous reports have presented similar cases, yet they have been in association with previous spine surgery,<sup>13</sup> or in the setting of previous osteoporotic vertebral body fractures.<sup>14</sup>

Although prolonged bisphosphonate therapy is generally considered safe, it does carry the potential risk of over-suppressing bone turnover, which may compromise the biomechanical bone properties in high stress areas. It also impairs stress fracture healing by blocking the osteoclastic remodeling of

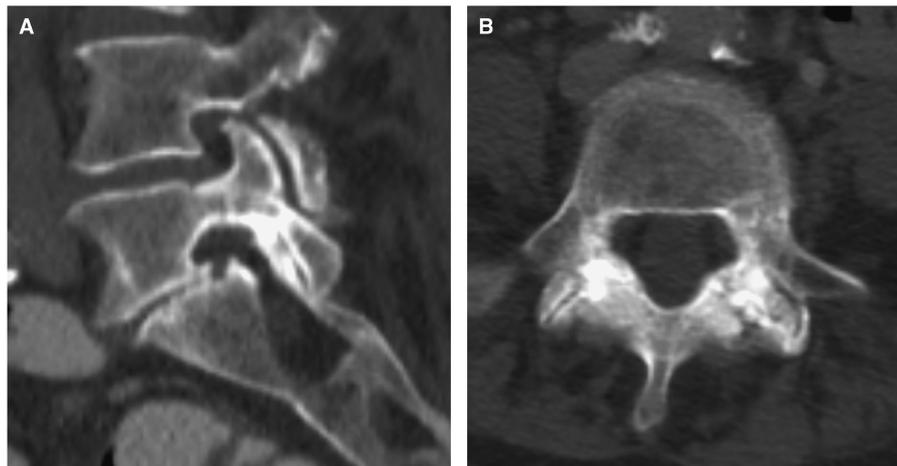
callus from woven to lamellar bone.<sup>15,16</sup> This has led to, and has been reported to relate with, the so-called “atypical fractures.” Such atypical fractures associated with bisphosphonate use have been reported previously in the proximal femur<sup>17</sup> and other skeletal sites.<sup>18,19</sup>

El Rachkidi et al<sup>20</sup> suggested that the pedicles may be another site of atypical fracture not previously reported with long-term Risedronate therapy. Karabay et al<sup>9</sup> reported a case of multi-level bilateral pedicle fractures with no other suggestive predisposing factor in which the patient was on a bisphosphonate. Like defects of the pars interarticularis, pedicle stress fractures may heal with bony sclerosis or progress to pseudoarthrosis.<sup>12,13</sup> To date, there is no confirmed method of treatment because of its rarity.<sup>2</sup>

This report represents one of the first cases of “*de novo*” atraumatic pedicle insufficiency bilateral fractures without any pre-existing biomechanical risk factors. We believe this type of fracture is easily overlooked on plain radiographs if undisplaced,<sup>21</sup> thus justifying advanced modalities if suspected. It may be associated with subsequent development of lytic spondylolisthesis, though the strength of such an association is not clear. In our case, the patient had a known history of osteoporosis



**Figure 4:** (A,B) Anterior–posterior and lateral X-rays of the lumbar spine performed 1 year after presentation. Demonstrating no progression of listhesis at L5-S1.



**Figure 5:** (A,B) Axial and sagittal CT scan of the spine performed 2 years after presentation. Demonstrating healed L5 pedicle fractures and residual grade 1 spondylolisthesis at L5-S1.

and prolonged Risedronate use. Thus, future surveillance for this diagnosis may demonstrate an association, especially as the incidence of bisphosphate therapy increases with the burgeoning older patient population.

#### DISCLOSURES

Saad Surur, Amy Zeglinski-Spinney, Mohamed M. El Koussy, Darren M. Roffey, and Eugene K. Wai have nothing to disclose.

#### STATEMENT OF AUTHORSHIP

EKW conceived of the report and contributed to the interpretation of the case, and wrote the first draft. SS, AZS, MME, and DMR contributed further to the interpretation of the case and the writing of subsequent drafts. EKW obtained the necessary

diagnostic images. All authors provided critical revisions of the manuscript and approved the final version.

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