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CASE REPORT

Intravertebral vacuum cleft sign: a cause of vertebral cold defect on bone scan

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Abstract A 67-year-old female presented with an acute compression fracture with an intravertebral vacuum cleft (IVC) sign of the T12 vertebra. Her bone scan demonstrated a cold defect of the fractured vertebra. Although the IVC sign is related to vertebral osteonecrosis, to the best of our knowledge, a cold defect on a bone scan has not been reported in an acute compression fracture with an IVC sign. In this case review, various imaging findings of osteonecrotic compression fractures are discussed along with a review of the current literature.

Keywords Compression fracture · Intravertebral vacuum · Bone scan · Osteonecrosis · Cold defect

Introduction

Vertebral osteonecrosis typically presents with an intravertebral vacuum cleft (IVC) sign. The IVC sign on plain radiography was first described in 1978 by Maldague et al. [1] as “a horizontal gas-density cleft” within the collapsed vertebral body. In their study, bone ischemia was histologically confirmed in one of the ten patients. Following the report by Maldague et al. [1], several additional studies have suggested that a compression fracture with an IVC sign has a benign etiology [2, 3]. From a histological perspective, a compression fracture with an IVC sign is highly related to vertebral osteonecrosis [4–6]. On MRI studies, the IVC gap can be replaced with fluid [6–8].

To the best of our knowledge, a vertebral cold defect on a bone scan in the context of an acute compression fracture with IVC has not been previously reported, despite the association of the IVC sign with vertebral osteonecrosis. On the bone scan, a compression fracture is generally known to increase uptake in the affected vertebra. Cold defects of the vertebra, especially in cancer patients, are usually considered a sign of osteolytic metastasis. Here, we present an acute compression fracture with IVC on the T12 vertebra that appeared as a cold defect on te bone scan. We suggest several plausible explanations for the presence of the unique cold defect in the study, which was not seen in previous reports [3, 9–11].

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Case report

A 67-year-old female underwent total gastrectomy for signet ring cell-type early gastric cancer at our hospital in July 2013. Preoperative F-18 fluorodeoxyglucose (FDG) PET/CT, abdominal CT, and chest X-ray revealed no metastatic lesions and no spinal compression fracture or intradiscal air (Fig. 1).

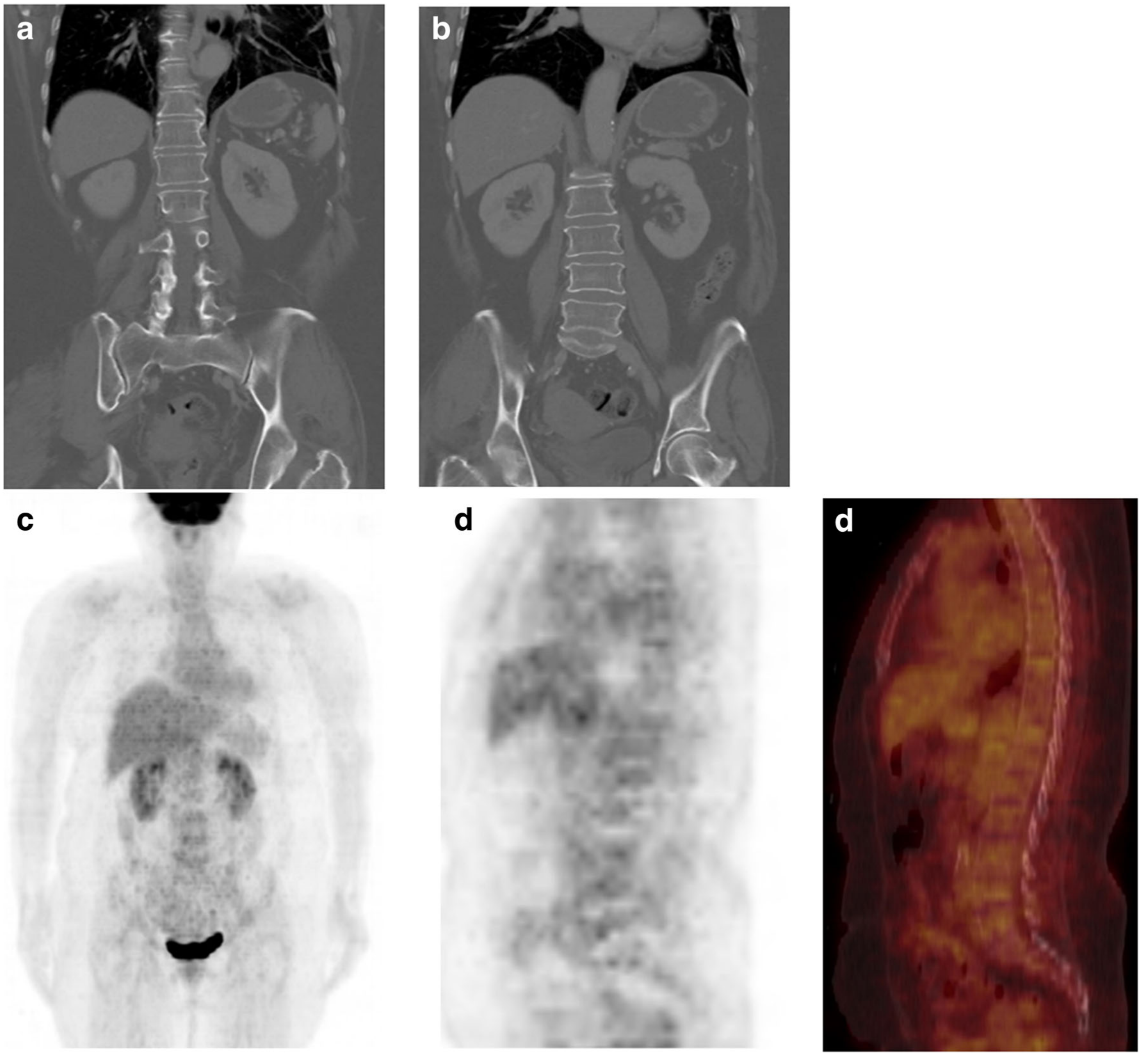

Fig. 1 CT and F-18 FDG PET/CT images used for stomach cancer staging. The bone window views of the coronal abdominal CT showed no metastatic lesions, compression fracture, or intradiscal air in the lower thoracic and lumbar vertebrae (**a** and **b**). There was no abnormal hypermetabolic lesion in the preoperative F-18 FDG PET/CT (**c**–**e**)

Two days after total gastrectomy (day 0), she fell on her buttocks (less than her own height) and subsequently complained of lower back pain. Radiographs of her spine on the day of her minor trauma (day 0) revealed a wedge-shaped compression fracture of the T12 vertebral body (Fig. 2). The next day (day 1), a CT imaging study of the spine was performed to further evaluate the patient's compression fracture. An IVC sign of the T12 vertebral body was noted on the CT scan, which was not visible on the initial spine radiograph (Fig. 3a, b). A bone scan using Tc-99m hydroxydiphosphonate (HDP) acquired the following day (day 2) showed a cold defect of the T12 vertebra without other significant abnormalities (Fig. 3c).

The patient's back pain worsened despite conservative analgesic treatment for 3 weeks, and the follow-up T and L spine radiographs were checked (day 24). Along with the previously noted compression fracture, the IVC, which was not observed in the initial radiograph, was demonstrated near the upper end plate of the T12 vertebral body (Fig. 4a). On the same day (day 24), a spine MRI was performed. Just below the end plate of the T12 vertebra, the MR images showed a sharply demarcated high signal intensity on the T2-weighted image (Fig. 2b) and low signal intensity on the T1-weighted image (Fig. 2c). These MR findings suggested that the IVC was replaced with fluid. Finally, approximately 1 week after the MRI (day 32), a bone biopsy at the T12 pedicle as well as percutaneous vertebroplasty was performed. The bone specimen of the T12 pedicle showed no evidence of neoplastic and/or infectious processes.

Discussion

The IVC sign appears as a transverse and linear air shadow on plain radiographs [12]. In addition, the IVC sign is more

Fig. 2 Plain radiograph of the L-spine. A wedge-shaped compression fracture of the T12 vertebral body (arrow) was noted on the plain radiograph on the day of the minor trauma. No significant IVC sign was seen in the fractured vertebral body

prominent in extension lateral views than in flexion views [13] and can be more easily detected by CT than by plain radiography [14]. When MRI is performed, the air-filled cleft may be replaced with a fluid signal [6, 8]. In the present case, the radiologic findings of the IVC sign were consistent with previous reports, except for the presence of a cold defect on the bone scan [6, 8, 14].

Compression fractures are a common radiologic finding. However, a compression fracture with an IVC sign can indicate another medical issue, that is, vertebral osteonecrosis. Maldague et al. [1] first reported the IVC sign in 1978, believing it could be associated with avascular necrosis. Since the report of Maldague et al. [1], histopathologic evidence of osteonecrosis in the vertebra with the IVC sign has been reported in several cases [15–18]. Libicher et al. [4] compared the histopathologic findings of vertebral compression fractures according to the presence of IVC on CT scans among a total of 180 studied patients. Of the 12 patients with IVC signs, 11 had osteonecrosis, and among the 13 patients who had osteonecrosis, 11 had IVC signs. Thus, the IVC sign can be considered a specific finding for vertebral osteonecrosis (sensitivity 85 %, specificity 99 %, and positive predictive value 91 %). In addition, medication with steroids carries similar risk factors for IVC and osteonecrosis of the femoral head [14].

Lin et al. [6] conducted a comparative study between the MR fluid sign of IVC and histopathologic findings in 20 consecutive patients. Among the specimens of the 20 studied patients, 14 had histopathologic findings of osteonecrosis. In the 14 patients with vertebral osteonecrosis, 86 % (12/14) had fluid signs on MRI, while 50 % (7/14) had IVC on plain radiographs. Further, the six patients without histopathologic findings of osteonecrosis showed no fluid sign. Thus, the authors suggested that the MR fluid sign is a more sensitive and powerful diagnostic finding with regard to vertebral osteonecrosis [6].

Although the IVC sign is currently not an extremely rare radiologic finding, only a few cases have been reported regarding bone scan findings. Bhalla and Reinus [3] evaluated 11 patients with the IVC sign, 3 of whom underwent a bone scan. The bone scans of all three patients showed “increased uptake” in the collapsed vertebrae with IVC, although the exact bone scan timing after trauma was not described. Van Eenenaam and el-Khoury [10] reported a patient with delayed vertebral collapse with the vacuum phenomenon. The patient had a normal plain radiograph 3 weeks after the initial back injury, while a bone scan showed “increased uptake” at the affected vertebra. The vertebra was collapsed with the vacuum phenomenon 9 weeks after the initial back injury. Another case report described a patient with delayed vertebral collapse who may have had Kummel’s disease [11]. In this case, the bone scan showed “iso-uptake” of the affected vertebra; however, there was no IVC sign in the collapsed vertebra. A similar case was reported by Javier et al. [9].

In the present case, the collapsed vertebra with an IVC sign showed a prominent cold defect on the bone scan, in contrast with previous studies. One possible explanation is that the cold defect is caused by the IVC itself because it may be difficult to deliver the radiotracer to the IVC area considering the air density cavity on spinal CT, suggesting the absence of significant bone tissue and a related blood supply. However, in this case, the whole vertebra was seen as a cold defect, not the part of the affected vertebra. The extent of the cold defect seen on the bone scan was larger than the IVC on the CT image. Therefore, we assumed that the extent of the cold defect could not be fully explained by the IVC itself. We believe that this large defect may have been due to the early timing of performing the bone scan. In the present case, an acute compression fracture was found on plain radiography on the same day as the patient’s accident, and the spine CT showed a compression fracture with an IVC sign on the next day. Two days after the minor trauma, a bone scan was performed to evaluate

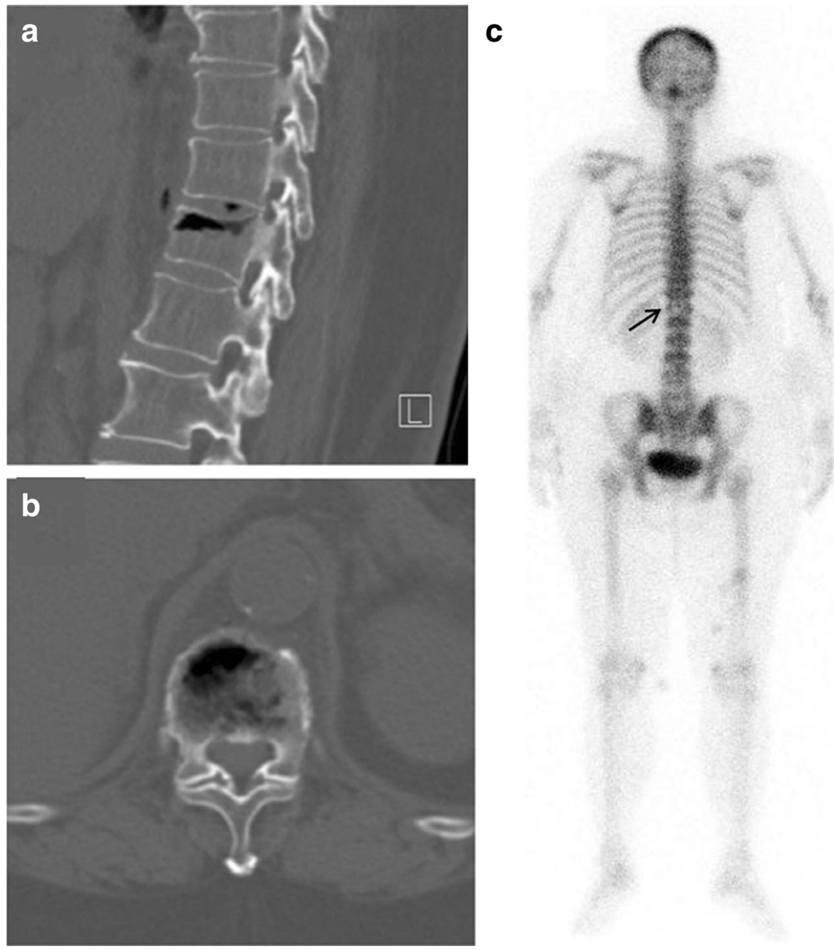

Fig. 3 Spine CT and bone scan after the minor trauma. The intravertebral vacuum cleft sign of the T12 vertebra (**a** and **b**) is shown, which was not visible on the initial spine radiograph. A bone scan using Tc-99m hydroxydiphosphonate (HDP) acquired the following day showed a prominent cold defect of the T12 vertebra with no other significant abnormalities (**c**)

unrecognized fractures. Conversely, the timing of the bone scan in a previous report was 3 weeks after the trauma [10]; however, the accurate timing of bone scans was not described in several previous articles [3, 9, 11]. Additional bone SPECT or SPECT/CT images might be helpful to correlate the cold defect to the IVC in our patient; unfortunately, it was not available in the present case.

Acute vertebral fracture with IVC leads to vascular disruption of the injured vertebral body, especially in the ventral zone, namely the anterior portion of the vertebral body [12]. Vertebral osteonecrosis begins with vascular disruption along with bone regeneration [6]. Bone regeneration includes mesenchymal cell proliferation, neovascularization, dead bone resorption by osteoclasts, and osteoblastic reaction. The amount of blood supply and the osteoblastic reaction are related to the intensity of uptake on the bone scan [19]. In an acute vertebral compression fracture with IVC, the blood supply is interrupted, and the fractured vertebra may not be revascularized. In addition, the effect of the osteoblastic reaction may be minimal in earlier phases. In the context of our patient, the lack of blood supply and osteoblastic reaction of the earlier period after the trauma may have led to the cold defect on the bone scan. The bone scan in our study was performed much earlier than that of Van Eenenaam et al. [10] . If the bone scan was performed at several weeks after the compression fracture with IVC, the fractured vertebra would show increased uptake by the revascularization and osteoblastic reaction. Thus, we postulated that the lack of reported cases of scintigraphically cold lesions is primarily due to both the scarcity of compression fractures with IVC signs and the need for an early bone scan.

Most compression fractures with the IVC sign are benign in nature [2]. However, vertebral collapse with the IVC sign has been reported in some multiple myeloma patients [2, 20]. Multiple myeloma is a well-known malignant disease that can produce a cold defect on bone scans [21]. We performed a bone biopsy of the pedicle to exclude the possibility of multiple myeloma, which was negative on the bone biopsy specimen. The histopathologic analysis also did not show evidence of osteonecrosis in our case. We hypothesized that the discordant result between the imaging (MRI and CT) and histopathologic findings may have been due to the bone biopsy site. Specifically, the bone biopsy site of other previous studies was the affected vertebral body [4, 6], while the pedicle was used as the biopsy site in our case.

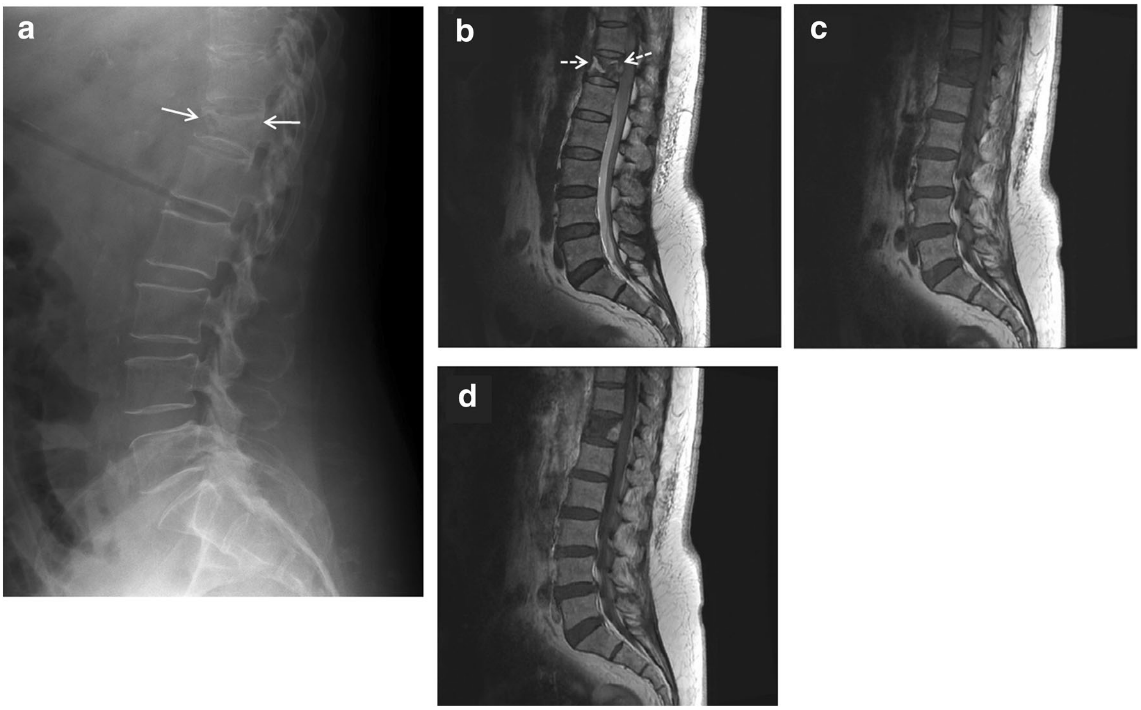

Fig. 4 Follow-up plain radiograph and MRI of the L-spine. About 3 weeks (day 24) after the minor trauma, an intravertebral vacuum cleft (IVC) was suspected on the follow-up radiograph, located near the upper end plate of the T12 vertebral body along with a wedge-shaped compression fracture (**a**, arrows). A spine MRI was performed on the same day (**b**, **c**, **d**). Just below the upper end plate of the T12 vertebra, the T2-weighted image (**b**) showed a sharply demarcated linear high-signal-intensity lesion (dotted arrows), while the T1-weighted image (**c**) showed low signal intensity at the IVC. These findings suggested that the IVC had been replaced with fluid. Mild gadolinium enhancement of the posterior body was noted, possibly indicating non-specific hyperemia (**d**)

In conclusion, a vertebral compression fracture with an IVC sign can produce a cold defect on the bone scan if it is performed during the early phase of repair, possibly within several days of the trauma. In the early phase, the blood supply of the vertebral body may be reduced and an osteoblastic reaction may not yet have been initiated. Ultimately, this diminished vascularity and lack of osteoblastic reaction may result in a cold defect in the compression fracture with IVC, which can also be an indicator of vertebral osteonecrosis.

Compliance with ethical standards None

Conflict of interest The authors declare that they have no conflict of interest.

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