CASE REPORT

Pyogenic vertebral osteomyelitis in a breast cancer patient: report of a case

Hisamitsu Zaha · Mai Onomura · Yukiko Nishikuramori

Received: 21 April 2011/Accepted: 15 August 2011/Published online: 1 March 2012 © Springer 2012

Abstract We herein report a rare case of pyogenic vertebral osteomyelitis (PVO) coexisting with breast carcinoma. A 71-year-old female presented with neck pain without fever. Magnetic resonance imaging (MRI) showed suspected metastatic lesions in her neck (C7 and Th1). Fluorine-18 fluorodeoxyglucose positron emission tomography (FDG-PET) showed increased FDG uptake in the neck spines and in the left breast. A core needle biopsy of the left breast revealed the presence of invasive ductal carcinoma. Our first tentative diagnosis of the patient was left breast carcinoma with bone metastases, and first-line endocrine therapy was started. However, surgical intervention for the spines had to be considered, because her neurological symptoms progressed. A repeated MRI scan showed a narrowing of the disc space and fluid accumulation around the vertebrae. This suggested the presence of PVO rather than metastases. Surgery confirmed the presence of PVO in C7 and Th1, and a culture of the abscess yielded Escherichia coli. The patient's neurological symptoms dramatically improved after surgery. Breast conserving surgery was performed 3 months after the surgery for PVO. The patient is well and has no clinical evidence of disease 18 months after the breast conserving surgery. PVO is rare, but should be included in the differential diagnosis in patients presenting with early breast carcinoma.

H. Zaha (⊠) · M. Onomura Department of Breast Surgery, Nakagami Hospital, 6-25-5 Chibana, Okinawa, Okinawa 94-2195, Japan e-mail: hisamitu@nakagami.or.jp

Y. Nishikuramori Department of Radiology, Nakagami Hospital, 6-25-5 Chibana, Okinawa, Okinawa 94-2195, Japan

 $\underline{\underline{\mathscr{D}}}$ Springer

Keywords Pyogenic osteomyelitis · Bone metastasis · Breast cancer

Introduction

Pyogenic vertebral osteomyelitis (PVO) is relatively rare and often subtle in its clinical presentation [1, 2]. It is difficult to make a correct diagnosis when it coexists in breast carcinoma patients, because it can mimic vertebral bone metastasis [3]. A misdiagnosis or delayed diagnosis can cause serious clinical problems [1–5]. We herein report a case of PVO that was difficult to differentiate from vertebral bone metastases from breast carcinoma.

Case report

A 71-year-old Japanese female with diabetes mellitus and a past history of pyelonephritis presented at another hospital with neck pain lasting for 3 months. Magnetic resonance imaging (MRI) suggested the presence of metastatic lesions in her neck (C7 and Th1) (Fig. 1), and the patient was referred to our hospital in November 2008. The patient was afebrile and complained of rapidly worsening sensory disturbance of both hands. A complete blood count showed a hemoglobin value of 12.1 g/dl, hematocrit at 36.6%, and a white blood cell count of 4570/mm³ with 31.1% lymphocytes. The patient's fasting serum blood glucose level was 267 mg/dl and her hemoglobin A1c was 7.6%. Her C reactive protein (CRP) level was slightly elevated up to 2.6 mg/dl. Fluorine-18 fluorodeoxyglucose positron emission tomography (FDG-PET) showed increased FDG uptake in the neck spines (C7 and Th1) and the left breast (Fig. 2). The patient was referred to our Breast Surgical Department.

Fig. 1 Sagital magnetic resonance imagings demonstrated abnormal signals in the C7 and Th1 vertebrae. a T1, b short T1 inversion recovery sequence (STIR-MRI)

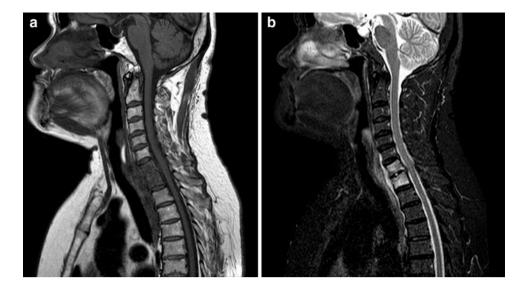
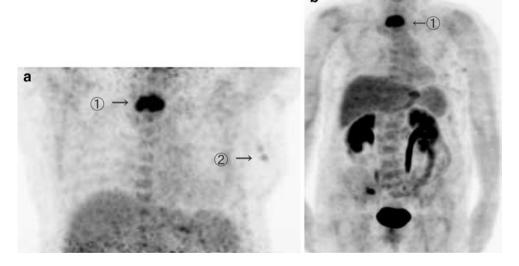


Fig. 2 a FDG-PET images showed markedly increased uptake of 18 FDG in the neck (C7, Th1), and small uptake in the left breast. b No other abnormal uptake in other bones except for C7 and Th1



Mammography, breast ultrasonography and breast MRI found an irregular-shaped tumor of about 2×1 cm in the left lower-outer quadrant of the left breast (Fig. 3), and a core needle biopsy of the tumor revealed an invasive ductal carcinoma. Immunohistochemical studies revealed positive staining for the estrogen receptor (ER) and progesterone receptor (PgR), and was negative for HER2/neu overexpression (HER2). The patient's serum levels of tumor markers such as CEA, CA15-3 and NCC-ST439 were all within the normal ranges. Although favorable tumor characteristics did not suggest the existence of metastatic lesions, bone metastases could not be excluded according to the MRI and FDG-PET findings. First-line endocrine therapy combined with bisphosphonate therapy was planned, along with a careful observation of the patient's neurological symptoms in December 2008. However, urgent surgical intervention for the spines had to be considered because her neck pain and sensory disturbance of both hands rapidly progressed 3 weeks after the initiation of endocrine therapy.

A repeated MRI scan was performed, and demonstrated rapidly destructive vertebrae with compression of the spinal cord and narrowing of the disc space throughout the C7 and the Th1 vertebrae (Fig. 4). Because a narrowing of the disc space and fluid accumulation around the vertebrae became obvious, pyogenic osteomyelitis was suspected, rather than metastases. On the day of admission, administration of antibiotics was started, and surgery on the spines was performed via an anterior approach on the 4th hospital day in January 2009. The abscess was mainly located in the disc space between C7 and Th1, and debridement of the disc and vertebrae of C7 and Th1 was performed. After debridement, a fragment of the iliac bone was harvested and transplanted between the C7 and the Th1 for fixation. A culture of the abscess yielded Escherichia coli. The patient's neurological symptoms dramatically improved



after the surgery. The patient continued receiving endocrine therapy for 3 months, and the tumor in her left breast decreased in size. Breast conserving surgery was performed in April 2009. Histopathologically, the tumor was an invasive ductal carcinoma 1.5 cm in diameter, and was negative for lymph node metastasis. The immunohistochemical analysis was positive for ER and PgR, and negative for HER2. The patient is now doing well and has no clinical evidence of disease at 24 months after undergoing breast conserving surgery.

Discussion

It is generally difficult to differentiate inflammatory changes from vertebral metastases in breast carcinoma

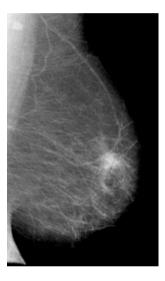
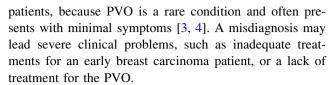


Fig. 3 Mamamography showed irregular shaped tumor with 2×1 cm in diameter, which was suspected malignancy

Fig. 4 Repeated MRI demonstrated sever destruction of the disk between the C7 and Th1, and fluid accumulation around the vertebrae, which suggested pyogenic osteomyelitis rather than metastatic lesions. a T1, b short T1 inversion recovery sequence (STIR-MRI)



The conditions most commonly underlying PVO are diabetes mellitus, ethanol abuse, renal and hepatic failure, malignancy, immunosuppression and intravenous drug abuse [1, 2]. Most cases of PVO result from hematogenous dissemination from a remote site of infection, most commonly the urinary tract [1]. Our patient was at relatively high risk of PVO, because of the presence of diabetes mellitus and a history of pyelonephritis, in addition to her malignancy of the breast.

The most common symptom of PVO is back pain, followed by fever. However, a fever is often absent at the presentation, thus distracting the clinician from the possibility of infection and delaying the diagnosis [1, 2]. Mylona et al. [1] examined a systematic review of PVO, and reported that 40% of PVO patients did not have a fever at presentation. In our case, the absence of fever and leukocytosis were taken as evidence against the presence of a suppurative process, and a misdiagnosis of the condition as bone metastases was made. The leukocyte count is the least useful among the inflammatory markers for PVO; it is high in only one-third to one-half of affected patients [2]. Unlike the leukocyte count, the erythrocyte sedimentation rate (ESR) and CRP level are sensitive markers, which are elevated in the majority of cases with PVO [2].

Since the symptoms and clinical findings of PVO are often nonspecific and minimal, imaging is an essential part of the diagnostic workup. Plain radiographic changes often take weeks to months to appear. The earliest radiographic sign may be an ill-defined vertebral endplate. Enlargement of the perivertebral soft tissues may also be detected in







association with bony impairment [6]. PVO, however, causes a narrowing of the disc space, in contrast to metastatic bone disease, in which the disc space is preserved [7]. Therefore, a lack of disc involvement is a reliable sign of vertebral metastasis in the overwhelming majority of cases [8].

Computed tomography (CT) and MRI have improved the sensitivity over the simple radiography [1–3], however, their findings are nonspecific, and in some instances, may be indistinguishable from those of tumor [3, 9]. Gadolinium enhancement of discs, vertebrae and surrounding soft tissues improves the accuracy of MRI, particularly in early infections when other changes may be subtle, and also helps to differentiate infectious lesions from degenerative changes or neoplasms [10]. FDG-PET has shown promise as a very sensitive modality [11]. It can effectively distinguish infection from degenerative changes even when MRI is inconclusive [12]; however, it shows a low specificity for neoplasms [11].

In our case, careless reading of the MRI led to a suspicion of metastatic vertebral lesions at first, because the disc space was relatively preserved. When, FDG-PET was performed to find the primary tumor for the bone metastases, a co-existing early breast carcinoma was detected. Finally, the vertebral lesions were correctly diagnosed as PVO. The treatment strategy for the breast carcinoma had to be changed because of the rapid worsening of PVO, which consequently resulted in successful breast conserving surgery after preoperative endocrine therapy. In such a complicated case, preoperative endocrine therapy might be a good option to give a priority to the treatment for severe PVO, if the breast carcinoma has endocrine responsiveness. Unlike chemotherapy and surgery, endocrine therapy does not influence the complex infectious disease or the host immune system, and recent clinical trials suggest that preoperative endocrine therapy might be a good treatment option for breast carcinomas. However, when PVO coexists with an endocrine non-responsive breast carcinoma, a treatment delay for the breast carcinoma may be necessary.

Careful reading of the radiographic results with regard to the vertebral discs and surrounding soft tissues should be done when metastatic vertebral lesions are suspected, especially when continuous vertebrae are involved. Fineneedle aspiration biopsy and culture of suspicious lesions should be performed to make a rapid and definite diagnosis when necessary [3].

In conclusion, we herein reported a case of PVO, which was initially misdiagnosed as bone metastases from breast carcinoma. PVO is rare, but should be included in the differential diagnosis in patients with early breast carcinoma.

Conflict of interest Hisamitsu Zaha and co-authors have no conflict of interest.

References

- Mylona E, Samarkos M, Kakalou E, Fanourgiakis P, Skoutelis A. Pyogenic vertebral osteomyelitis: a systematic review of clinical characteristics. Semin Arthritis Rheum. 2009;39(1):10–7.
- Gouliouris T, Aliyu SH, Brown NM. Spondylodiscitis: update on diagnosis and management. J Antimicrob Chemother. 2010;65 Suppl 3:iii11–24.
- Voravud N, Theriault R, Hortobagyi G. Vertebral osteomyelitis mimicking bone metastasis in breast cancer patients. Am J Clin Oncol. 1992;15(5):428–32.
- Khan IA, Vaccari AR, Zlotolw DA. Management of vertebral diskitis and osteomyelitis. Orthopedics. 1999;22(8):758–65.
- Goldrick A, Harnett PR. Vertebral osteomyelitis in advanced breast cancer. Aust N Z J Med. 1993;23(4):406.
- Balériaux DL, Neugroschl C. Spinal and spinal cord infection. Eur Radiol. 2004;14 Suppl 3: E72–83 (review).
- Adams JE, Isherwood I. Conventional and new techniques in radiological diagnosis. In: Stoll BA, Parbhoo S, editors. Bone metastasis: monitoring and treatment. New York: Raven Press, Ltd.; 1983. p. 107–48.
- 8. Norman A, Kambolis CP. Tumors of the spine and their relationship to the intervertebral disc. AJR. 1964;92:1270-4.
- Easton EJ, Powers JA. The spine. In: Thorofare NJ, editor. Musculoskeletal magnetic resonance imaging. New Jersey: Slack Inc.; 1986. p. 93–121.
- Dagimanjian A, Schils J, McHenry MC, Modic MT. MR imaging of vertebral osteomyelitis revisited. AJR Am J Roentgenol. 1996;167:1539–43.
- Schmitz A, Risse JH, Grünwald F, Gassel F, Biersack HJ, Schmitt O. Fluorine-18 fluorodeoxyglucose positron emission tomography findings in spondylitis: preliminary results. Eur Spine J. 2001;10(6):534–9.
- Stumpe KD, Zanetti M, Weishaupt D, Hodler J, Boos N, Von Schulthess GK. FDG positron emission tomography for differentiation of degenerative and infectious endplate abnormalities in the lumbar spine detected on MR imaging. AJR Am J Roentgenol. 2002;179(5):1151–7.

