Alteration of Signal Intensity on Follow-Up Magnetic Resonance Imaging in Spinal Epidural Inflammatory Myofibroblastic Tumor: A Case Report and Literature Review

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Abstract

Inflammatory myofibroblastic tumor (IMT) in the spine or paraspinal area is extremely rare. Here, we report a rare case of a 56-year-old man who had pathological-proven spinal epidural IMT showing a malignancy-like infiltrative feature on MR imaging with alteration of T2 signal intensity on follow-up from slightly high to low compared with initial MR imaging. To the best of our knowledge, this is the first report to describe paraspinal IMT monitoring with various imaging modalities without treatment. From this case, we were able to gain understanding of the natural course of IMT, and it could be helpful in the differential diagnosis of infiltrative paraspinal masses.

Keywords: Inflammatory Myofibroblastic Tumor, Spine, Computed Tomography, Magnetic Resonance Imaging, Radionuclides Imaging

1. Introduction

Inflammatory myofibroblastic tumor (IMT) is a mesenchymal neoplasm of intermediate biological potential composed of differentiated myofibroblastic spindle cells and an inflammatory cell infiltrate of plasma cells and lymphocytes. IMT commonly involves the lung and the orbit; however, the involvement of the spine or paraspinal area is extremely rare. Most tumors found in the spine are metastases, while primary benign tumors are rare (1).

IMT has a propensity to mimic clinically and radiologically a malignant tumor (2, 3). There are no distinguishable characteristic findings of IMT, and various signal intensity of the mass was reported at one time point (4-14). We report a rare case of pathologic-proven spinal epidural IMT which showed not only changing signal intensity on magnetic resonance imaging (MRI), but also corresponding change on computed tomography (CT) and bone scan, without any treatment over a 2-year follow-up. Understanding the spectrum of imaging findings of IMT is useful in the differential diagnosis of an infiltrative paraspinal mass.

2. Case Presentation

A 55-year-old man was referred to our hospital with a paraspinal mass at T10 level. His initial symptoms were presented 3 weeks before with a sudden-onset of pain and swelling of the left back accompanied by a chilling sensation. His medical history was unremarkable. He had a mild sensory disturbance on the left side of T9 and T10 dermatome during the physical exam. Upon admission, his laboratory tests revealed a high white blood cell (WBC) count of 23700/uL (normal 4,000 - 10,000/uL) with elevated neutrophil count of 19742/uL (83.3%, normal 40% - 74%), a serum C-reactive protein (CRP) concentration of 226.49 mg/L (normal 0 - 5.0 mg/L), and an erythrocyte sedimentation rate (ESR) of 20 mm/hr (normal 0 - 15 mm/hr).

L-spine MRI revealed a left-sided paraspinal soft tissue mass from T9 to L1, showing iso to mild hyperintensity on T2-weighted image relative to the muscle with homogeneous well-enhancement on fat-saturated T1-weighted image after contrast infusion (Figure 1A and B). This mass extended into the left epidural space and neural foramen at T10 - 12 levels, causing mild compression of the dural sac. In addition, the lesion involved the adjacent left side of the vertebral body and posterior column of T11, and 9 - 12th ribs (Figure 1B and 2A). A Chest CT on the following day also demonstrated a 6 cm-sized enhancing infiltrative soft tissue mass on the left lateral aspect of T9 - L1 bodies with associated intracanal extension (not shown) and bone destruction at the left posterior arc of the T11th rib and T11th body (Figure 3). The bone scan performed a week later showed mildly increased bony uptake on the left side of the T11 - T12 vertebral bodies (Figure 4A).

Based on the radiological findings, the initial impression was a nonspecific soft tissue malignancy, such as lymphoma or a metastatic tumor. For the diagnosis, open
A 55-year-old man presented with the sudden-onset of pain and swelling of the left back accompanied by a chilling sensation. A, Initial magnetic resonance (MR) axial scans show iso to slightly hyperintensity left-sided paraspinal soft tissue mass (arrow) at T10 level compared to adjacent muscle with associated intracanal extension with mild dural sac compression on T2-weighted image (TR = 3,300/TE = 80 ms). B, The lesion (arrow) demonstrates relatively homogeneous enhancement on axial fat-saturated post-contrast T1-weighted MR image (TR = 610/TE = 15 ms) in the initial MR axial scan. C, Two-year follow-up MR axial scans show decreased size of the paraspinal soft tissue mass (arrow) at T10 level and signal change to marked hypointensity compared to adjacent muscle on T2-weighted MR image (TR = 3,650/TE = 80 ms). D, The lesion (arrow) demonstrates subtly decreased homogeneous enhancement on axial fat-saturated post-contrast T1-weighted MR image (TR = 610/TE = 15 ms).

excisional biopsy, not resection, was done to confirm the paraspinal mass. The intraoperative finding of the lesion was suggestive of inflammation and granulation tissue in the paraspinal muscle layer. The histopathology disclosed a localized area of inflammatory cell infiltrations intermingled with interlacing bundles of spindle cells in a collagenized background, and confirmed as IMT (Figure 5).

The patient’s symptoms improved after 6 months without any treatment. Follow-up chest CT and L-spine MRI were obtained on the 2-year follow-up visit. Chest CT revealed a decreased size of the mass, but intracanal extension and bony destruction of the left 11th rib and T11 body were still noted. On an MRI on the same day, in addition to the decreased size of the mass with little change of intracanal extension, the mass showed low signal intensity relative to muscle on T2-weighted image (Figure 1C). Furthermore, the extent of bone marrow signal change and enhancement of T11 and 9–12th ribs decreased (Figure 1D and 2B). A bone scan demonstrated decreased bony uptake on the left side of T11 - T12 vertebral body compared to the initial study (Figure 4B). The patient did not have any special treatment after the excisional biopsy. Besides the change of imaging features, the patient complained of no symptom and his laboratory tests showed improvement with a WBC count of 11,000/μL (normal 4,000 - 10,000/μL), neutrophil count of 7760 (70.3%, normal 40% - 74%), and a serum CRP concentration of 23.78 mg/L (normal 0 ~ 15 mm/hr).

3. Discussion

In this case, we observed a spinal epidural IMT initially showing iso to mildly high signal intensity relative to muscle on T2-weighted image changing to low signal intensity and regressing on the 2-year follow-up MRI, CT, and bone scan without receiving treatment. In the literature review, nine cases of spinal epidural IMTs demonstrated characteristic signal intensity at one time point of the disease course, but no case report exist regarding signal changes on follow-up MR images to date (Table 1).
There are conflicting reports concerning signal intensity changes of IMT on T2-weighted image. A few articles have reported that IMT usually has low signal intensity on T2-weighted images with homogeneous or heterogeneous enhancement (5, 7, 8, 10, 13). Han et al. (7) suggested that T2 hypointensity of an IMT might be explained by a relative lack of both free water and mobile protons within a fibrotic lesion; whereas, there are also a few reported cases of IMT demonstrating hyperintensity on T2-weighted images (9, 14). Seol et al. (14) speculated that the signal intensity on T2-weighted images is dependent on the degree of reactive and fibrotic lesions within the IMT. In other words, it is suggested that the area showing abundant inflammatory cells in vascular stroma could be hyperintense on T2-weighted images, and fibrous and collagenous components correlate with hypointensity on T2-weighted images. Briefly, this might be explained that the amount of fibrosis and cellular infiltration diversify the imaging findings of IMT.

Histologically, IMT is characterized by a variably cellular spindle cell proliferation in a myxoid to collagenous stroma with a prominent inflammatory infiltrate composed primarily of plasma cells and lymphocytes, with occasional admixed eosinophils and neutrophils. Coffin et al. (15) described three basic histological patterns of IMT according to the composition of spindled, fibroblastic-

Figure 2. Comparison of coronal fat-saturated post-contrast T1-weighted MR images between initial and two-year follow-up scans. A, Coronal fat-saturated post-contrast T1-weighted MR image (TR = 510/TE = 15 ms) shows enhancement at paraspinal soft tissue mass (arrow) from T9 to L1 with adjacent bone marrow signal change at the T11 body (arrowhead). B, Two-year follow-up coronal fat-saturated post-contrast T1-weighted MR image (TR = 510/TE = 15 ms) shows decreased size of the paraspinal soft tissue mass (arrow) from T9 to L1 and decreased extent of adjacent enhancing bone marrow signal change at the T11 body (arrowhead).

Figure 3. Initial chest computed tomography (CT) scan. Axial chest CT with bone setting shows an infiltrative soft tissue mass (arrow) on the left lateral aspect of T11 body and a bone destructive lesion at the left posterior arc of the 11th rib (arrowheads).

Figure 4. A, Initial Tc-99 m hydroxymethylene diphosphonate (HDP) whole-body bone scan shows mildly increased bony uptake on the left side of T11 - T12 vertebral bodies (arrows) suggesting bony involvement. B, Two-year follow-up bone scan shows no bony uptake on the left side of T11 - T12 vertebral bodies (arrows) compared to initial bone scan.

Figure 5. Microscopic finding of the excision specimen of left-sided paraspinal mass shows a localized area of inflammatory cell infiltration intermingled with interlacing bundles of spindle cells in a collagenized background, confirmed as inflammatory myofibroblastic tumor (Hematoxylin-eosin (HE) × 200).
filtrative feature could be helpful in accurate diagnosis of these imaging findings during the disease course with in¬
position as an evolution of the disease. Recognition of result from the transitional change of histopathologic com-
turbed course of IMT. With this finding, we may assume ical signal change on MRI throughout the natural undis-
consider it as a differential diagnosis of infiltrative paraspinal of this entity with invasive radiologic features, and con-
our case. Although it is rare, radiologists should be aware size of bone marrow enhancement on MRI, and relief of the decreased bony uptake on follow-up bone scan, decreased
infiltrative feature may be required from either a bone destruction or biopsy. However, despite infiltrative fea-
tures such as bone destruction and intracanal extension, decreased bony uptake on follow-up bone scan, decreased size of bone marrow enhancement on MRI, and relief of the symptoms helped to discriminate IMT from malignancy in our case. Although it is rare, radiologists should be aware of this entity with invasive radiologic features, and consider it as a differential diagnosis of infiltrative paraspinal soft tissue mass.

In conclusion, in this case, we observed the chronologi
cal signal change on MRI throughout the natural undis
turbed course of IMT. With this finding, we may assume that the signal change within the IMT lesion might re
sult from the transitional change of histopathologic com
position as an evolution of the disease. Recognition of these imaging findings during the disease course with in
filtrative feature could be helpful in accurate diagnosis of paraspinal IMT.

Footnotes

Authors’ Contributions: Jinyoung Chang was responsible for the study concept and design and drafting of the manuscript. Sung Hye Koh carried out acquisition of data. Analysis and interpretation of data and critical revision of the manuscript for important intellectual content was done by Sun-Yong Park. Administrative, technical, and material support was performed by Sung Hye Koh, and Soo Kee Min. Finally, Kwan Seop Lee supervised the study.

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Table 1. Characteristics of Cases of Spinal Epidural Inflammatory Myofibroblastic Tumor Reported in the Literature

| References         | Year | Age, y/sex | Clinical Symptoms                              | Laboratory Findings | Location | Bone Destruction | Signal Intensity on MR Images Compared to Adjacent Muscle | Treatment                          | Follow-Up, Month |
|--------------------|------|------------|-----------------------------------------------|---------------------|----------|------------------|--------------------------------------------------|------------------------------------|------------------|
| Roberts et al. (4) | 1997 | 58F        | Back pain, gait disturbance, paresthesia       | Normal              | T9-T11   | +                | Iso, Hypo                                       | Resection                         | 6                |
| Gilliard et al. (6) | 2000 | 45M        | Progressive quadriplegia, distal paresthesia   | NA                  | C5-T2    | +                | Iso, NA                                       | Resection + IV steroid             | 2                |
| Roberts et al. (6) | 2000 | 5M         | Back pain, numbness in limbs, gait disturbance| NA                  | T9-T10   | -                | Iso, Hypo                                       | Resection + IV steroid             | 6                |
| Seo et al. (4)     | 2005 | 44M        | Paraplegia, urinary incontinence, progressive back pain | Normal              | T1-T2    | -                | Iso, Hyper                                       | Resection                         | 12               |
| Sailler et al. (13)| 2006 | 78M        | Back pain, weakness in lower limbs             | ESR 35 mm/hr; CRP   | T6-C3    | NA               | NA, Hypo                                       | Resection + IV steroid + CPA       | 7                |
| Sailler et al. (13)| 2006 | 73F        | Back pain, weakness in lower limbs             | ESR 35 mm/hr; CRP   | T5-T7    | NA               | NA, Hypo                                       | Resection + IV steroid + CPA + IVIG| NA               |
| Kato et al. (9)    | 2012 | 63M        | Back pain, numbness of the lower limbs, gait disturbance | Normal              | T5-T6    | -                | Iso, Hypo oligogranular                        | Resection                         | 24               |
| Sato et al. (15)   | 2014 | 42M        | Lower back pain, radiating pain, weakness of right lower limb, mild bladder dysfunction | Normal              | L4-S2    | -                | Iso, Ketonuria                                 | Resection + IV steroid             | NA               |
| Kanagawa et al. (4) | 2015 | 40F        | Numbness of the lower limbs, gait disturbance, sensory change at right T9 dermatome | Normal              | T9-T11   | -                | Hypo                                            | Excision + IV steroid             | 2                |
| Present case       | 2015 | 55M        | Back pain, mild sensory change at left T9-T10 dermatome | ESR 20 mm/hr; CRP   | T9-L1    | +                | Iso, Hyper oligogranular                       | Resection + IV steroid             | 24 (4)           |

Abbreviation: NA, not available; PMN, polymorphonuclear neutrophil; ESR, erythrocyte sedimentation rate; CRP, C-reactive protein; WBC, white blood cell; CPA, cyclophosphamide; IVIG, intravenous immunoglobulin; WBC, white blood cell; CRP, C-reactive protein; ESR, erythrocyte sedimentation rate.

Follow-up imaging with CT, MRI, and bone scan.