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Case Report

MRI findings of serous atrophy of bone marrow with postirradiation changes: A case report☆

*Wai Kit Kung, MBBS, FRCR**, *Wing Yan Chin, MB ChB, FRCR, FHKCR*

Department of Radiology and Imaging, Queen Elizabeth Hospital, Rm 9, G/F, Block K, 30 Gascoigne Rd, Kowloon, Hong Kong

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ABSTRACT

This case report presents serous atrophy of bone marrow (SABM) in a cachexic patient with metastatic squamous cell carcinoma who had undergone radiotherapy. The unique magnetic resonance imaging (MRI) findings of SABM, known as the "flip-flop" phenomenon, were observed in both the irradiated and nonirradiated areas, a finding previously unreported in the literature. The report highlighted the characteristic features of SABM in various MRI sequences, which can be easily misinterpreted as technical errors, leading to unnecessary repetition of MRIs.

Introduction

Serous atrophy of bone marrow (SABM) is a rare disorder of the bone marrow that is associated with many underlying conditions, most commonly anorexia nervosa and cachexia. Magnetic resonance imaging (MRI) findings of SABM, known as the "flip-flop" phenomenon, can be misleading and frequently misinterpreted as technical errors, resulting in unnecessary repeated imaging [1].

This article reports a case of SABM in a cachexic patient with metastatic squamous cell carcinoma who had undergone radiotherapy, with post-irradiation changes in the thoracic spine and sacrum. Notably, these irradiated areas also exhibited the "flip-flop" phenomenon, a finding which has not been previously reported in the literature to the best of our knowledge.

Case presentation

The patient was a 68-year-old Chinese man with a history of squamous cell carcinoma of the skin in left buttock treated with radical excision done 21 years ago. Six months after the surgery, he was found to have left inguinal nodal metastasis with radical groin dissection and right rectus myocutaneous flap performed. Adjuvant radiotherapy (60 Grays in total) was also given to the left groin region for better local control.

☆ Competing Interests: The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

* Corresponding author.
E-mail address: allenkwk@gmail.com (W.K. Kung).

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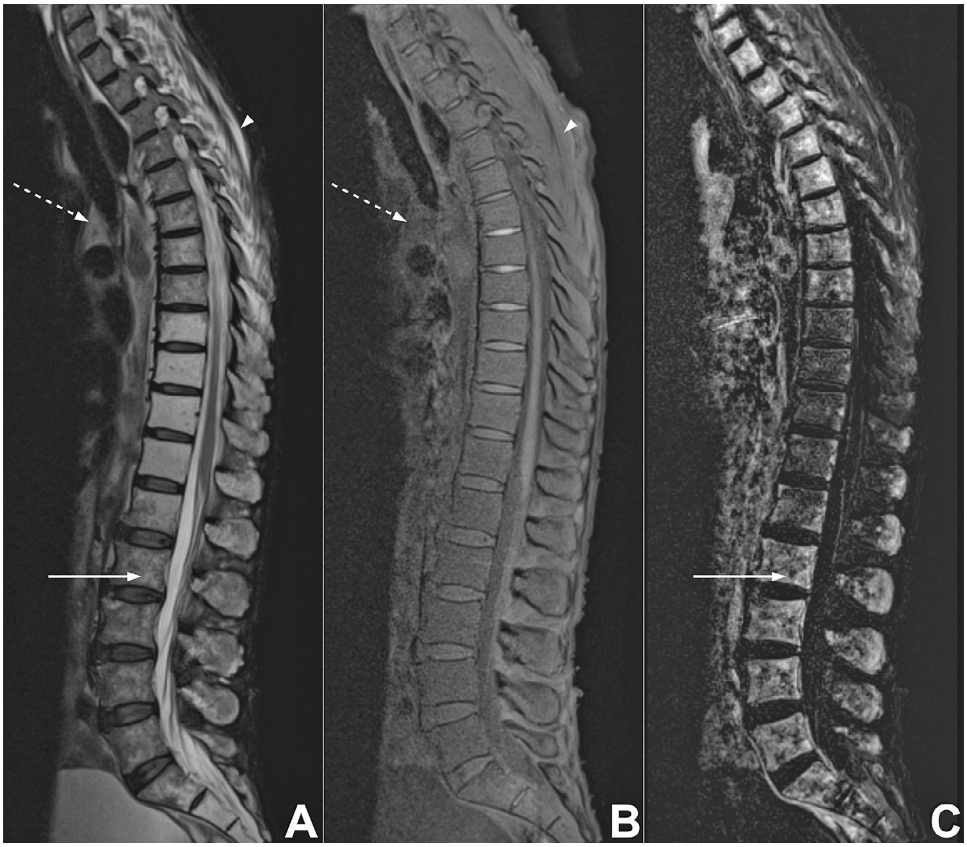

Fig. 1 – Sagittal T2W TSE STIR (A), sagittal T1W TSE (B) and contrast-enhanced T1W TSE with subtraction (C) images of MRI of the thoracolumbar spine show patchy T2W hyperintense, nonenhancing areas in the bone marrow (arrows), compatible with heterogeneous involvement of SABM, which is a common pattern in the axial skeleton. Relative homogeneous T2W hyperintense signals are seen in the postirradiated areas in mid thoracic spine and sacrum, in keeping with more diffuse involvement by SABM. The expected areas of subcutaneous fat in the upper back (arrowheads) and visceral fat (dotted arrows) are abnormally T1W mildly hypointense and T2W hyperintense.

Two years ago, an incidental finding of an enlarged gastrohepatic lymph node up to 2.8 cm was detected in a contrast-enhanced computed tomography of the abdomen and pelvis. It was subsequently confirmed to be nodal metastasis of squamous cell carcinoma by endoscopic ultrasonography-guided fine needle aspiration. No new primary is detected in the positron emission tomography-computed tomography (PET-CT). The gastrohepatic nodal metastasis was deemed unresectable and treated with intensity-modulated radiation therapy (50.4 Grays in total).

A follow-up PET-CT, 4 months postradiotherapy, revealed mild shrinkage of the gastrohepatic nodal metastasis but several new nodal metastases were detected in the abdominal para-aortic, right retrocrural and left supraclavicular regions. Palliative chemotherapy was then given. The patient soon complained of progressive dysphagia and weight loss. The next PET-CT showed a marked enlargement of the gastrohepatic lymph nodes, which had invaded the gastroesophageal junction. He eventually became dependent on nasogastric tube feeding. He was cachectic with a body mass index of 14.6 kg/m^2.

In the latest episode of care, the patient complained of progressive bilateral lower limb weakness. There was no urinary or fecal incontinence. Physical examination revealed bilateral lower limb power of grade 3/5 with slightly lax anal sphincter tone, while the lower limb and perineal sensation remained intact. MRI of the thoracolumbar spine with gadolinium contrast administration was carried out to investigate the underlying cause (Figs. 1–3).

The bone marrow of the included spine was diffusely T1-weighted (T1W) mildly hypointense relative to muscles. In T9 to L1 vertebra and the left-sided sacrum, which were the known irradiated areas, showed homogeneous hyperintense signals with sharp borders in T2-weighted (T2W) and short-tau inversion recovery (STIR) sequences. In contrast, heterogeneous T2W hyperintense signals were noted in the rest of the bone marrow. These T2W hyperintense signals are nonenhancing in the contrast-enhanced T1W sequence with subtraction. The bone marrow showed no definite signal drop out in the opposite-phase images as compared to the in-phase images in T2W Dixon sequences. The expected areas of subcutaneous and visceral fat showed abnormal T1W hypointense and T2W hyperintense signals, signifying severe fat depletion. The imaging features were consistent with SABM. There was no evidence of bony metastasis or cord compression.

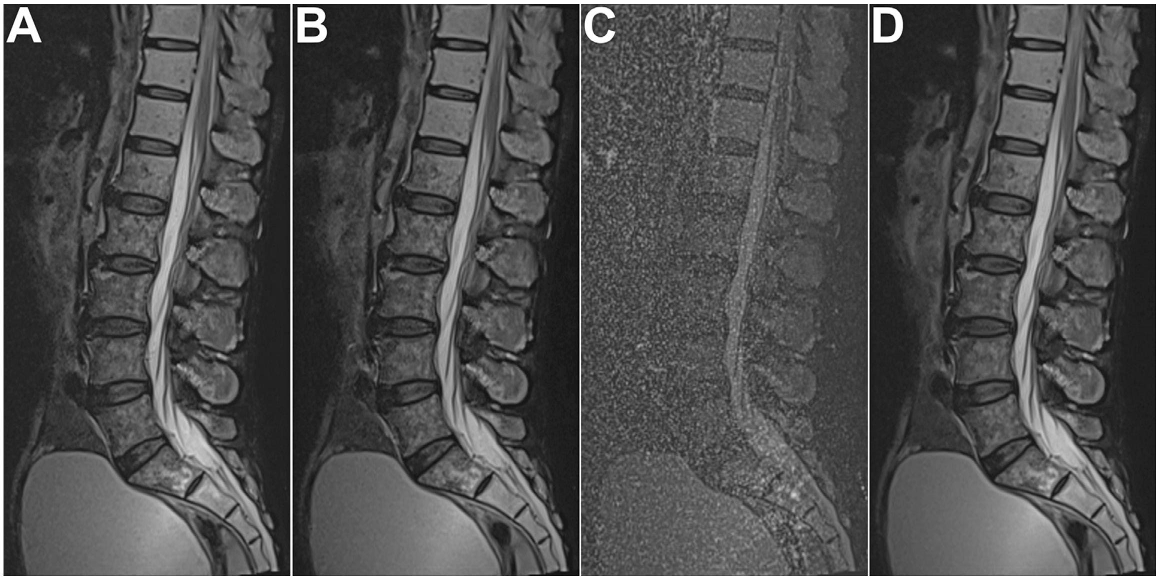

Fig. 2 – T2W TSE Dixon sequence with opposed-phase (A), in-phase (B), fat-only (C) and water-only (D) images of MRI of the thoracolumbar spine. There is no visible difference between the opposed-phase and in-phase images due to the lack of fat in the patient's body, resulting in minimal chemical shift artefacts. The bone marrow signals are expected to be hypointense in water-only images as normal adult bone marrow is mainly composed of fat.

Fig. 3 – T1W TSE (A), contrast-enhanced T1W TSE (B), and contrast-enhanced fat-saturated T1W TSE (C) images of MRI of the thoracolumbar spine. There is no visible difference in the contrast-enhanced images before and after the application of fat-saturation, indicating that the patient's body lacks normal fat.

Discussion

SABM, a rare condition also known as gelatinous bone marrow transformation or starvation marrow, is characterized by atrophy of fat cells, reduction of hematopoietic cells, and accumulation of extracellular gelatinous substance rich in hyaluronic acid-based mucopolysaccharides [2].

This disorder primarily affects adults, with a higher frequency in males. SABM often manifests as a morphological sign of a generalized severe illness and is commonly associated with significant weight loss and cachexia. The condition has a variety of underlying causes including malnutrition, malignancy, malabsorption, alcoholism, chronic heart failure, chronic renal failure, and chronic infections like acquired immunodeficiency syndrome and tuberculosis [2,3].

During periods of negative energy balance, the body mobilizes subcutaneous and visceral fat with a paradoxical increase in bone marrow fat. This might lead to a preferential differentiation of mesenchymal stem cells towards adipocytes rather than osteoblast lineage. The bone marrow adipose tissue appears to be resistant to lipolysis until other fat reservoirs have been depleted, which typically occurs in the late stages of starvation. SABM develops when bone marrow fat stores are mobilized under these extreme conditions, leading to the extracellular space in the cancellous bone becoming filled with hyaluronic acid-rich mucopolysaccharides. A histopathological diagnosis of SABM can be established by identifying the presence of extracellular gelatinous substances with a significant decrease in the number and size of hematopoietic and fat cells [1].

In MRI, the affected bone marrow is mildly hypointense on T1W images and hyperintense on fat-suppressed fluid-sensitive images relative to muscles [1]. The expected areas of subcutaneous and visceral fat also display abnormal T1W hypointense signals and hyperintense signals in fat-suppressed fluid-sensitive sequences.

SABM typically begins in the appendicular skeleton before spreading to the axial skeleton. As such, homogeneous involvement in the peripheral skeleton and heterogeneous involvement in the axial skeleton are common [3], as observed in our case. This pattern differs from diffuse bone marrow infiltration conditions or reconversion marrow disorders which primarily affect the axial skeleton and progressively extend to the appendicular skeleton. In our case, the heterogeneous hyperintense signals in the affected bone marrow in fluid-sensitive sequences are nonenhancing in the contrast-enhanced T1W sequence with subtraction. This contrasts with other bone marrow infiltrative conditions like malignancies and infections, which generally exhibit contrast enhancement in hyperintense areas on fluid-sensitive sequences. This distinction serves as another useful tool in differentiating between SABM and other infiltrative diseases.

The postradiotherapy signal changes in bone marrow depend on the duration and dosage of the treatment. In the acute phase, the bone marrow develops hyperintense signals in fluid-sensitive sequences attributable to edema, vascular congestion, and capillary injury to the fine vasculature. Contrast enhancement is also noted and possibly related to dilatation of sinusoids. In the chronic phase, depletion and replacement of the hematopoietic cells and blood vessels by the yellow fat cells occur. The fatty marrow replacement is seen as early as 2 weeks after the initiation of the radiation therapy [4,5]. In our case, there were 2 distinct irradiation fields - one targeting the left groin region, delivered over 2 decades ago, and the other focusing on the gastrohepatic nodal metastasis, conducted approximately 2 years prior to the MRI study. Under normal circumstances, one might anticipate fatty replacement within the bone marrow of these irradiated areas, characterized by hyperintense signals on T1W sequences and complete signal suppression on fat-suppressed sequences. However, contrary to these expectations, the bone marrow within the irradiation region displayed homogeneous hyperintense signals in fluid-sensitive sequences and hypointense signals in T1W sequences. In the context of serous atrophy of bone marrow, we postulate that the fatty marrow within the irradiation field underwent a transformation akin to that observed in the non-irradiated bone marrow, that is, lipolysis accompanied by the deposition of gelatinous materials.

It is not uncommon to attribute the abnormally hyperintense signals of the bone marrow, subcutaneous fat and visceral fat in fat-suppressed sequences to technical errors such as failed fat suppression, leading to repeated imaging [1]. In our center, T2W turbo spin-echo (TSE) Dixon sequence is routinely performed in the spinal MRI protocol. In our case, the in-phase and opposed-phase sequences have nearly identical images with chemical shift artefacts hardly observed. It can be due to the fact that the bone marrow and soft tissues of the patient lack normal fat, resulting in similar signals when the protons in water and fat molecules are in-phase and out-of-phase. Given the confusing marrow signal characteristics, repeated imaging with additional STIR and contrast-enhanced T1W sequences (with and without fat suppression) were performed. Subtraction technique was also adopted to better assess the marrow enhancement.

The distinctiveness of our case lies in the depiction of SABM within the postirradiation area, a phenomenon not previously documented in existing literature to the best of our understanding. A variety of sequences were employed to demonstrate the pathology, including Dixon method, STIR and contrast-enhanced fat-saturated T1W sequence. The baffling picture in Dixon method in SABM is also rarely reported and discussed in the literature.

Limitations of this case report include the lack of histological confirmation. Nevertheless, it would be unethical to obtain unnecessary biopsy in this patient with end-stage cancer under palliative care, particularly when the MRI imaging features are indicative of SABM. Another limitation is the absence of prior MRI studies to illustrate the gradual alteration in bone marrow signals. Ideally, an MRI covering the appendicular skeleton would have provided a more holistic view of the distribution of SABM across the entire body.

Conclusion

Recognizing the characteristic MRI features of SABM is crucial as misinterpretation often results in erroneous attribution of the findings to technical errors, leading to unnecessary repetition of MRIs. In addition, radiologists should be aware of the potential involvement of SABM in the post-irradiated areas to facilitate more accurate imaging interpretation.

Patient consent

I confirmed that the written informed consent for publication of this case report was obtained from the patient.

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