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Case Report

Melorheostosis of a rib

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ABSTRACT

Melorheostosis is a rare sclerosing bone dysplasia, characterized by cortical and medullary hyperostosis with typical “dripping candle wax” appearance, usually involving the long bones. Here, we present a case of melorheostosis of a rib, incidentally discovered in an asymptomatic adult patient.

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Case presentation

A 68-year-old previously healthy male was found to have an exophytic sclerotic costal lesion on chest radiograph performed for the assessment of acute minor thoracic trauma (Fig. 1). We could neither obtain any information as to the mechanism and precise location of the thoracic injury, nor to the nature of the patient’s presenting symptoms. Neither was there any history of remote thoracic trauma, which could have produced callous formation around rib fractures or have resulted in myositis ossificans. The lesion remained stable on a follow-up chest radiograph obtained 4.5 months later. By that time, the patient had no symptoms related to the rib lesion.

Subsequent computed tomography (CT) chest performed 5 months after the original chest radiograph, confirmed the presence of a sclerotic lesion involving the lateral aspect of the right seventh rib (Fig. 2). The lesion had a lobulated, ex-

ophytic, extraosseous component extending along the external cortex of the rib with a “dripping candle wax” appearance, characteristic of melorheostosis.

As the possibility of an aggressive lesion, such as an osteosarcoma, was entertained at the time of the initial CT interpretation, the patient had undergone additional investigation with magnetic resonance imaging (MRI) of the chest performed 2 weeks following the CT. The MRI showed the lesion to have a uniformly hypointense T1W and T2W signal, and no enhancement following intravenous gadolinium injection (Fig. 3).

The 18-F sodium fluoride whole body positron emission tomography/CT bone scan performed 3 weeks after the MRI, demonstrated that the lesion featured moderately intense uptake of tracer (Fig. 4). No other active bone lesion was identified on the positron emission tomography bone scan.

Assessment of the patient by a multidisciplinary team reached the conclusion that the rib lesion was benign.

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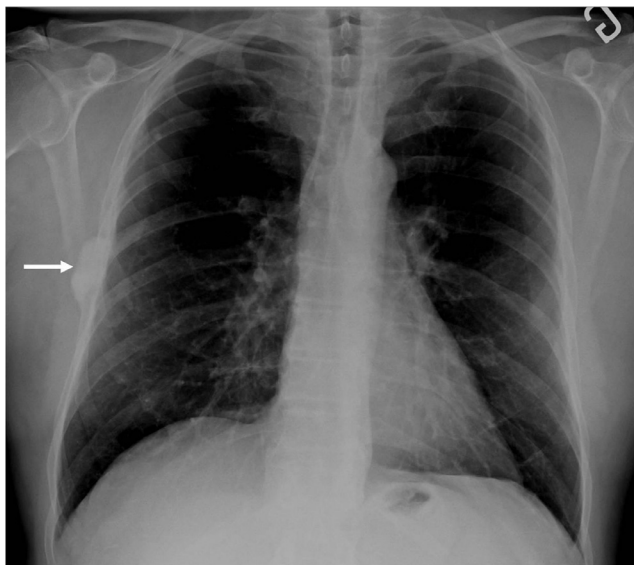


Fig. 1 – Postero-anterior chest radiograph of a 68-year-old previously healthy male performed for the assessment of a minor chest trauma demonstrates a dense lesion arising from the right lateral ribs (arrow).

The follow-up chest CTs performed up to 2 years following initial presentation, demonstrated stability of the rib lesion. The patient remained asymptomatic.

Hence the combination of typical radiological appearance, absence of symptoms and long-term stability of the lesion yielded the diagnosis of melorheostosis. Other bone lesions which could have an extrasosseous calcified component were excluded based on imaging and clinical findings. The extrasosseous component of the lesion in this case demonstrated diffuse, homogenous dense calcification without ossification or medullary continuity with the underlying rib, excluding the possibility of mature heterotopic ossification and osteochondroma. The location, presence of sclerosis of the parent bone, dense homogeneous calcification, and homogeneously hypointense T1 and T2 signal with lack of enhancement made bizarre parosteal osteochondromatous proliferation unlikely, as bizarre parosteal osteochondromatous proliferation the lat-

ter most commonly occurs in the hands and feet, contains varying amounts of calcifications, and demonstrates intermediate to high signal on T2-weighted sequences and enhances postgadolinium [1]. Imaging features were not consistent with tumoral calcinosis neither, which on radiographs, typically appears as periarticular, amorphous, cystic and lobulated calcifications, and demonstrates T2 hyperintensity on MRI [2].

Discussion

Melorheostosis is a rare sporadic sclerosing bone dysplasia. It was first described by Leri and Joanny in 1922 as *hyperostose en coulée*—flowing hyperostosis [3]. Henceforth, it is also referred to as "Leri disease." The name "melorheostosis" is derived from the Greek words "melos"—meaning "limb" and "rhein" which means "flow," in reference to the characteristic appearance of flowing hyperostosis. The etiology of melorheostosis remains unknown.

Melorheostosis predominantly affects the long bones of the upper and lower extremities. Vast majority of patients are asymptomatic and are usually diagnosed incidentally during imaging for unrelated reasons. In symptomatic cases, most common clinical presentation of melorheostosis is limb stiffness or pain [4]. Surgical excision of the lesions can be performed for symptomatic cases.

The "dripping candle wax" appearance on conventional radiography and CT is usually pathognomonic of melorheostosis [3–5]. MRI of melorheostosis typically shows the lesion to be of low signal intensity on all pulse sequences [6]. On radionuclide bone scanning, melorheostosis commonly shows moderate increased uptake [7]. A biopsy is unnecessary because the diagnosis can be made from the radiologic appearance alone [6].

Melorheostosis is uncommon in the axial skeleton and is very rare in the ribs [5,8]. When occurring in atypical anatomical location, melorheostosis can be misinterpreted as an aggressive lesion, as was the case of the initial CT of our patient. However, typical "dripping candle wax" sign and absence of aggressive features, such as cortical disruption and soft tissue component, should make imaging-based confident diagnosis of melorheostosis possible even outside the appendicular skeleton.

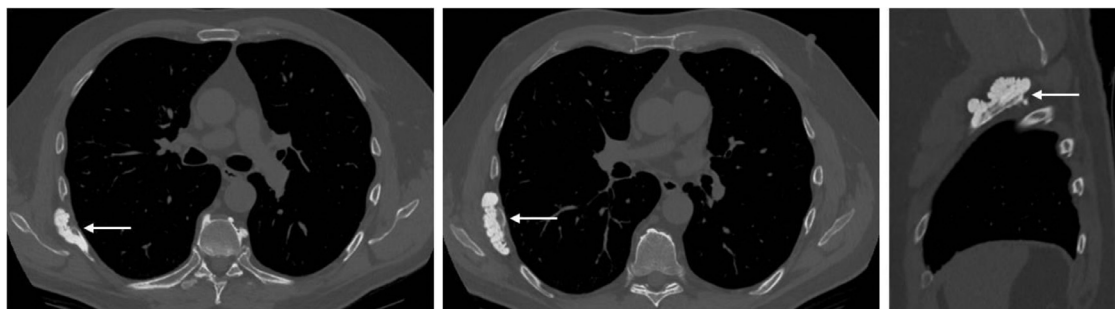


Fig. 2 – Axial and sagittal images of a CT chest show a sclerotic lesion arising from the lateral aspect of a right rib, extending along its external cortex, with a "dripping candle wax" appearance (arrows).

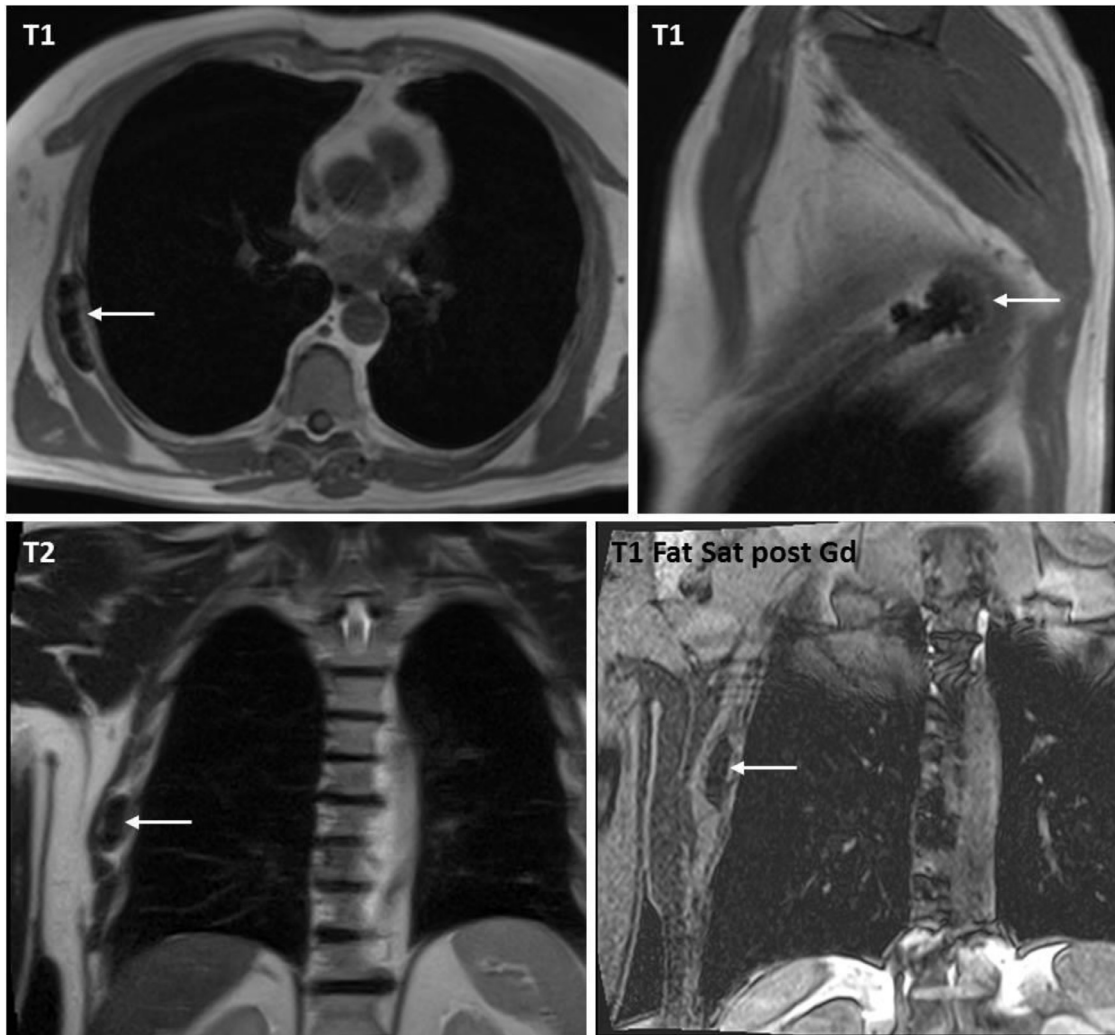


Fig. 3 – MRI of the chest. Axial and sagittal T1WI, coronal T2WI, and coronal T1 fat suppressed postintravenous gadolinium injection images demonstrate uniformly hypointense signal of the right rib lesion (arrows).

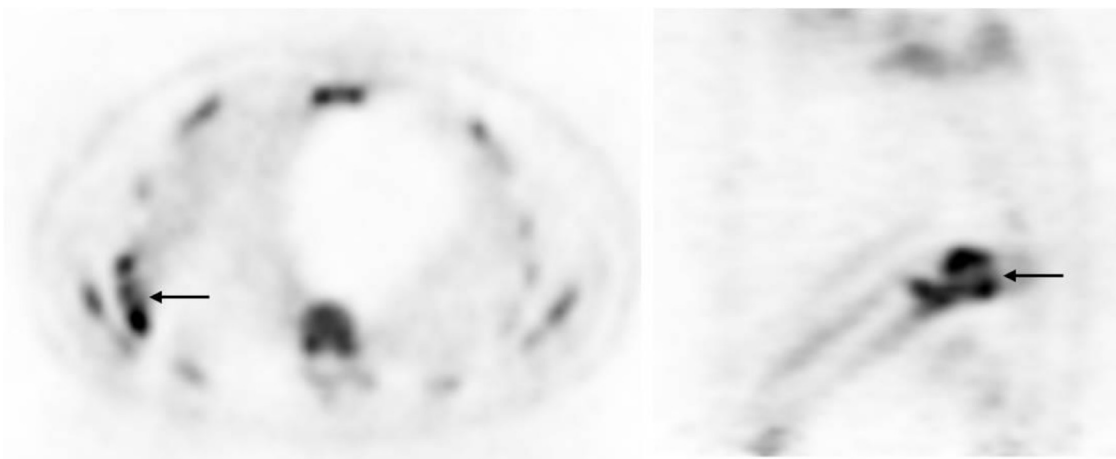


Fig. 4 – Axial and sagittal images of ^{18}F -sodium fluoride total body bone positron emission tomography/computed tomography (PET/CT) show moderately intense uptake of tracer by the right rib lesion (arrows).

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