CASE REPORT

Osteopoikilosis mimicking osteoblastic bone metastasis: a case report

Mariha Aslam[1*], Owais Bin Qadeer Gill[2], Shahar Bano[1], Muhammad Babar Imran[2], Shahzad Afzal[2]

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ABSTRACT

Background: Osteopoikilosis (OP) is a benign, uncommon disorder that is inherited by the autosomal dominant gene. Multiple benign enostoses are present in this specific kind of sclerosing skeletal dysplasia, which can be mistaken for osteoblastic skeletal metastases.

Case Presentation: We present here a case of a 50-year-old male patient with pain in the lower back for 3 months. His pelvic X-ray showed a few sclerotic rounded lesions in the ilium, acetabulum, and proximal femur. His magnetic resonance imaging (MRI) pelvis showed T1 and T2 weighted image hypo intense focal lesions in the ilium and acetabulum. His ^{99m} Technetium methylene diphosphonate bone scan was done showing no focal abnormal uptake in the lesion reported on pelvic X-ray and MRI. These features are suggestive of OP, ruling out osteoblastic skeletal metastasis.

Conclusion: OP is usually an incidental finding on radiograph. Both X-ray and MRI findings can mimic skeletal metastasis, but a normal bone scan can rule out metastasis and is useful if metastatic disease is thought to be a possibility.

Keywords: Osteopoikilosis, ^{99m}Tc-MDP bone scan, enostoses, skeletal scintigraphy, case report.

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Correspondence to: Mariha Aslam
Punjab Institute of Nuclear Medicine (PINUM) Cancer Hospital, Faisalabad, Pakistan.
Email: mariha.aslam@gmail.com.
Full list of author information is available at the end of the article.

Introduction

Osteopoikilosis (OP) is a rare, congenital, autosomal dominant, benign type of sclerosing bone dysplasia. It is usually an incidental finding in asymptomatic patients, but rarely some patients can present with pain at the location site. The coexistence of OP with pain in the pelvic region is rarely reported along with findings on magnetic resonance imaging (MRI) and ^{99m}Technetium methylene diphosphonate (Tc-MDP) bone scan. These imaging modalities play a critical role in distinguishing it from osteoblastic bone metastasis.

Case Report

A 50-year-old male patient complained of lower back pain that had been bothering him for 3 months. The pain did not radiate to the legs, was dull in nature, and came on gradually. He had a history of attending the gym for exercise for a few months. There is no history of trauma, tuberculosis, or any other co-morbid condition. His pelvic X-ray showed few, well-defined, round, tiny radio-dense foci in ilium, acetabulum, and proximal femur (Figure 1). His pelvis showed T1 weighted image (T1WI) and T2 weighted image (T2WI) hypo intense focal lesions in ilium and acetabulum, and proximal femur (Figures 2 and 3). After 3 days, his ^{99m}Tc MDP bone scan was done for further evaluation which showed non homogenous radiotracer uptake in the spine more marked in the L5 vertebra; however, no focal abnormal uptake was noted in lesions reported on pelvic X-ray and MRI (Figure 4). Based on all these findings diagnosis of OP was made.

Discussion

OP, also known as osteopathia condenses, asymptomatic bone dysplasia, and spotted bone disease, is a rare, congenital, benign type of sclerosing bone dysplasias [1].

This rare disease was first described by Albers-Schonberg in 1915 [2] with an incidence of 1 in every 50,000 subjects [3]. OP is inherited as an autosomal dominant disorder [4] and is seen in all age groups [5]. It occurs in both genders but some suggest that there is slight male predilection. This disease is featured by multiple, discrete round sclerotic lesions usually involving cancellous bone that especially involves epiphysis and metaphysis of long tubular bones, carpals, tarsal, and pelvis. The axial skeleton is largely spared and skull involvement is rare.

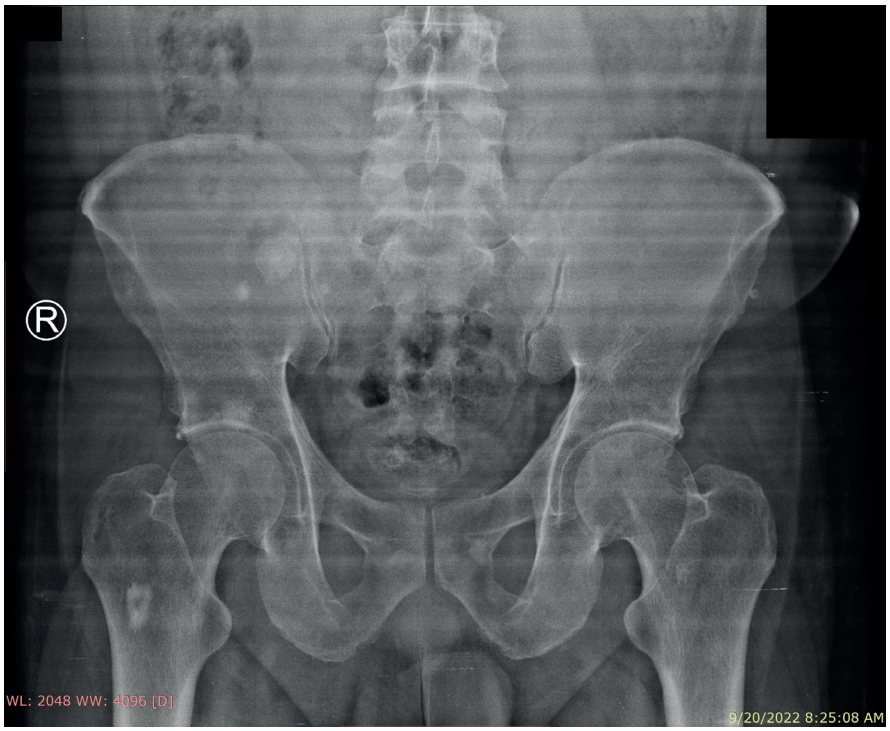

Figure 1. *Tiny radio-densities in ilium, acetabulum, and proximal right femur.*

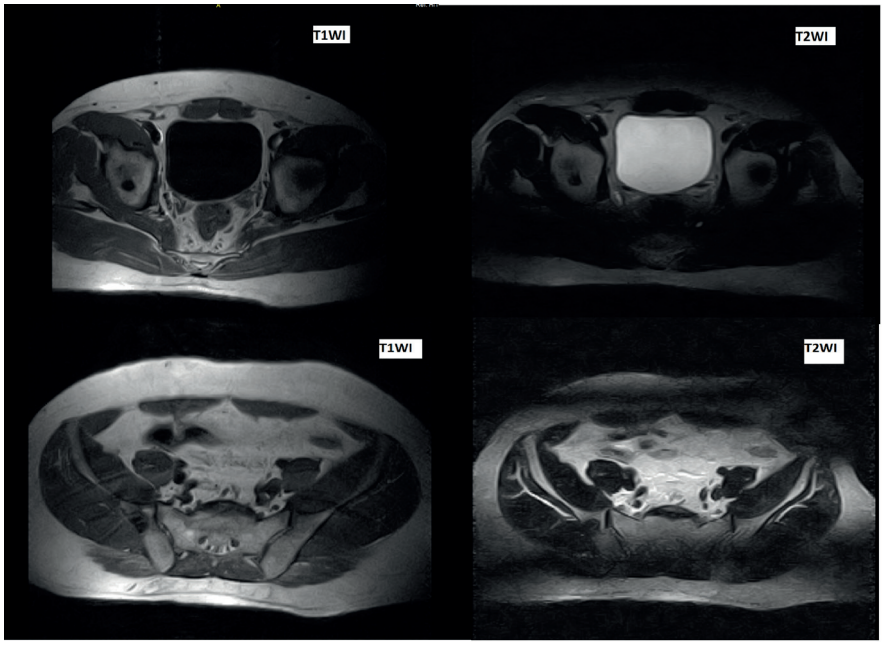

Figure 2. *Pelvic MRI (Transaxial slice) showing T1WI and T2WI hypo intense focal lesions in right ilium and acetabulum.*

OP is found along side with melorheostosis, osteopathia striata and is associated with Gunal-Seber-Basaran syndrome in which there is OP with dacrocystitis and Buschke-Ollendorff syndrome, also known as disseminated dermatofibrosis lenticularis, comprising OP associated with disseminated connective tissue and cutanoeous yellowish nevi, predominantly on the extremities and trunk [6].

Plain X-ray usually shows a homogeneously dense, sclerotic focus in the cancellous bone with distinctive radiating bony streaks ("thorny radiation") that blend with the trabeculae of the host bone, creating a feathered or brush-like border. They are presented on computed tomography as high attenuating areas with a mean attenuation threshold of >885 Hounsfield unit (HU) and maximum attenuation of >1,060 HU [7]. On MRI, they appear hypo intense on both T1WI and T2WI sequence. ^{99m}Tc MDP bone scan shows no uptake in these lesions and it helps in differentiating it from the more aggressive

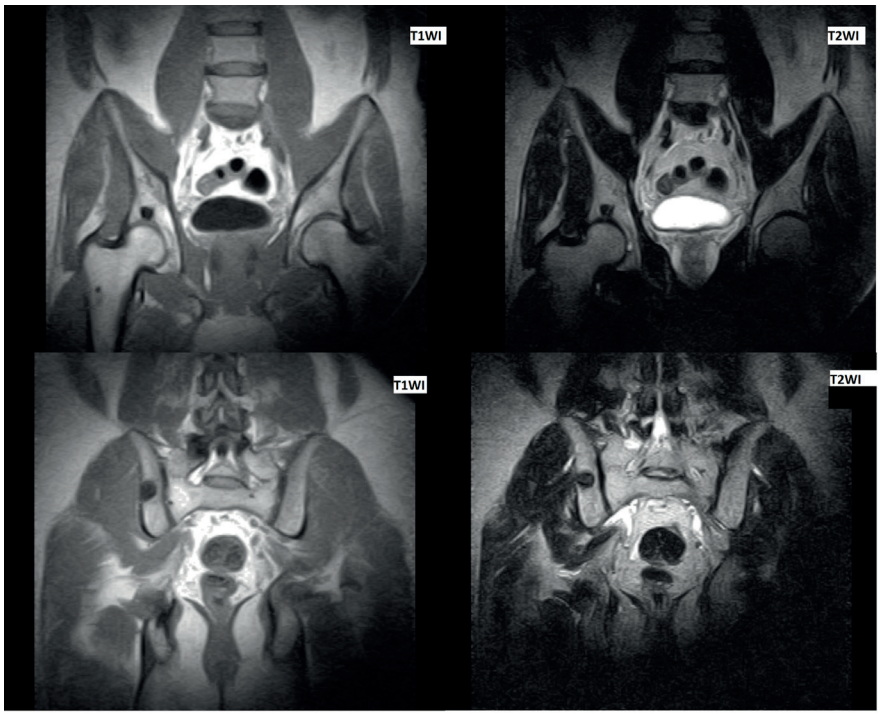

Figure 3. *Pelvic MRI (Coronal slice) showing T1WI and T2WI hypo intense focal lesions in right ilium and acetabulum.*

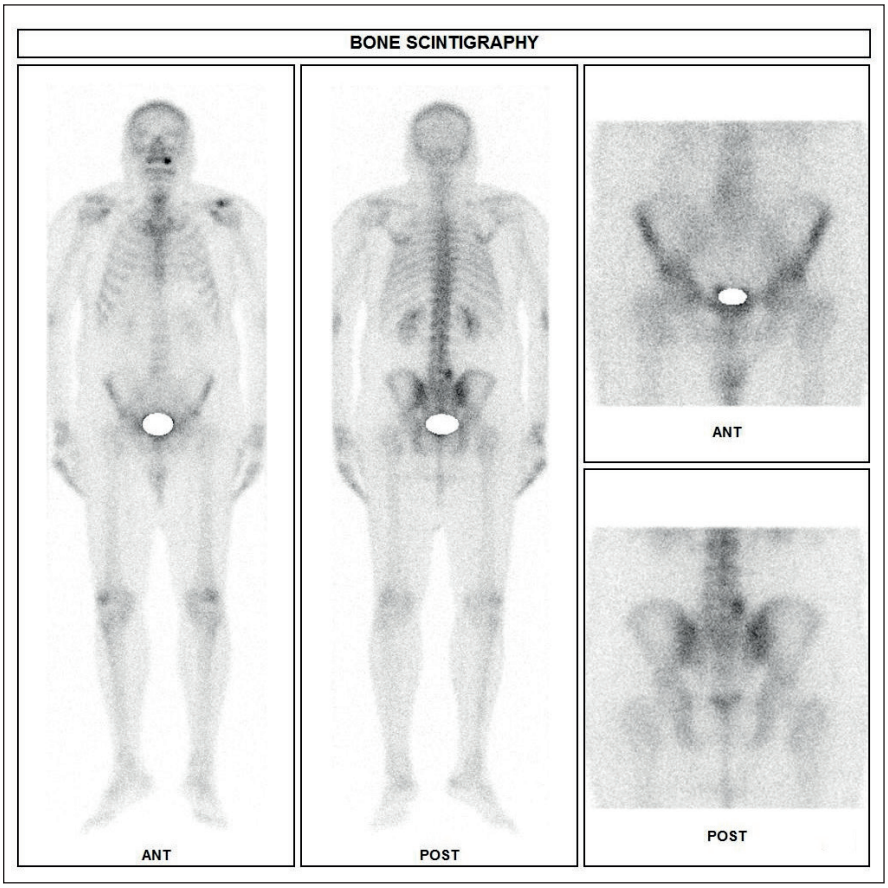

Figure 4. *^{99m}Tc MDP bone scan showing non homogenous radiotracer uptake in spine more marked in L5 vertebra. Lesions reported on X-ray and MRI are silent.*

and grave etiologies such as osteoblastic metastasis which really change the course of patient management.

It should be differentiated from other differential diagnosis on the basis of radiological and bone scan findings. D/D includes [5]:

Sclerotic bone mets: (They will show intense uptake on bone scan)

Osteopathia striata: (Osteopathia striata is typically bilateral, although occasionally it can be unilateral, typically in tubular bones. Radiographically prominent vertical striations predominate in the metaphysis and epiphysis of the long bones (celery stalk metaphysis).

Melorheostosis (periosteal cortical thickening is characteristic, but endosteal thickening is also seen in many cases. There are thick undulating ridges of bone, reminiscent of molten wax or flowing candle wax appearance. Bone scan will show increase uptake).

Osteoid osteoma (rarely multiple, X-ray show nidus with intense uptake on bone scan)

Erdheim Chester disease (bilateral, symmetric metaphyseal and diaphyseal sclerosis with cortical thickening, increased uptake on Tc-MDP bone scan).

Paget disease (Pelvic Paget's: cortical thickening and sclerosis of the iliopectineal and ischiopubic lines results in the pelvic brim sign and leads to obliteration of Köhler's teardrop. A bone scan will show intense uptake).

Other differentials include incidental bone islands, sclerotic bone dysplasias, osteosarcoma, lymphoma, and chronic multifocal sclerosing osteomyelitis.

OP is termed as 'DON'T TOUCH LESION'. Non-steroidal anti-inflammatory drugs, analgesics such as acetaminophen and opioids can also be used. Rare active lesions can be treated with bisphosphonates therapy.

Conclusion

In OP X-ray and MRI findings can mimic skeletal metastasis but a normal bone scan can rule out mets and is helpful if metastatic disease is considered in the differential.

List of Abbreviations

HU	Hounsfield unit
MRI	Magnetic resonance imaging
OP	Osteopoikilosis
T1WI	T1 weighted image
T2WI	T2 weighted image
Tc-MDP	Technetium methylene diphosphonate

Conflict of interests

The authors declare no conflict of interest regarding the publication of this article.

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Consent for publication

Informed consent was obtained from the patient for the publication of this case.

Ethical approval

Ethical approval is not required at our institution for publishing a case report in a medical journal.

Author details

Mariha Aslam[1], Owais Bin Qadeer Gill[2], Shahar Bano[1], Muhammad Babar Imran[2], Shahzad Afzal[2]

1. Post Graduate Resident (Nuclear Medicine), Punjab Institute of Nuclear Medicine (PINUM) Cancer Hospital, Faisalabad, Pakistan
2. Punjab Institute of Nuclear Medicine (PINUM) Cancer Hospital, Faisalabad, Pakistan

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