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Painful Osteopoikilosis: A Rare Case Presentation

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Abstract- Osteopoikilosis (OPK) is a painless rare hereditary disorder of autosomal dominance, characterized by radiological detection of multiple round or oval small osteosclerotic patches, clustered around the periarticular region in appendicial skeleton. OPK is an asymptomatic condition with a rare incidence of 1:50,000 and association of pain as a presenting symptom is still extremely rare. We present a case of this rare entity with review of literature.

Key words: Osteopoikilosis, Asymptomatic, Pain.

Case Presentation: A 22 year young healthy male, presented to the Out-Patient, Department of Orthopaedic, Government Medical College Jammu (J&K) with the prime complaint of pain both hands off and on after exertion of mild to moderate intensity, usually lasting for few hours to few days for a period of one year. There was no history of Trauma, other joint pains, morning stiffness, fever or weight loss. He was non smoker, non alcoholic, non diabetic, vegetarian and not a known case of any Systemic disease .His father had similar lesions in hands and feet but no pain and his mother and sister had no such lesions. General physical and systemic examination was within normal limits his pain score on visual analog was 5. His hand functions were normal, no local signs of inflammation and distal neurovascular status was normal. X-RAYS of both hands revealed both radii, carpals, bases of metacarpals and periarticular areas of MCP joints studded with multiple round and oval radiodense dots 3 to 6 mm in size. The architect of the bones was well preserved without any osteolytic erosions (Fig.1).

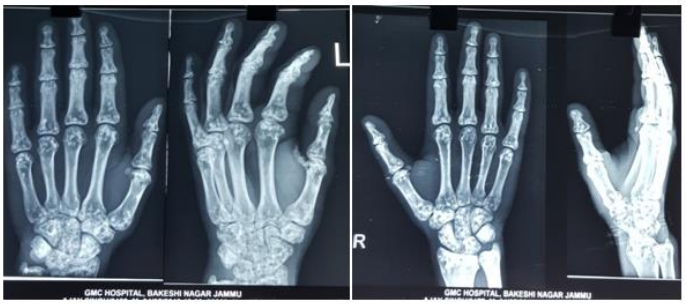

Fig1: Multiple sclerotic foci in both anterposterior and lateral views of variable size typical of Osteopoikilosis in both hands

Typical alkaline phosphatase, radiological findings in otherwise healthy individual leads us to diagnose it as a case of OPK. However, in view of rare presentation of pain, the patient was thoroughly investigated and a review of literature was done.

Investigations: The laboratory tests like complete blood count, sedimentation rate, CRP, Rhaeumatoid Factor, calcium, phosphate levels, serum uric acid, blood sugar, PSA and Thyroid function tests were done and all were within normal limits. X-Ray of pelvis, spine, skull shoulders and ankles were normal because of being benign asymptomatic entity. The patient was explained about its harmless nature and put on analgesics (Nsaids, Etoricoxi B 60mg) for a week to which he responded well. He was followed up three months for one year and was advised to take any analgesics SOS .He was satisfied and occasionally required painkillers. His father was asymptomatic and hence given no treatment.

Discussion

Osteopoikilosis is a benign, autosomal dominant sclerosing dysplasia of bone characterized by the presence of numerous bone islands in the skeleton[1]. OPK is also known as Osteopathia condensans disseminata, assymptomatic bone dysplasia, spotted bone disease [2]. It is a rare disease with occurance of 1:50,000 .The sex distribution as reported in the literature is 1:1, however male dominance by some authors was reported due to males reporting more often than females.The condition was first time diagnosed by ÄLBERS SCHONBERG" in 1915 [3]. The typical lesions are radiodense small ,round or oval in shape and occur in epiphysiometaphysial regions of appendicial skeleton. Rarely it may involve axial skeleton (dorsolumbar spine) as reported by Weisz [4]. Lesions are not known in skull. The importance of knowledge about Osteopoikilosis lies in being able to distinguish it from osteoblastic, Metastatic malignant lessions , so as not to raise an alarm and hence save patient from unnecessary agony. Usually, the condition is asymptomatic, but our patint presented with pain that lead us to extensive differential diagnosis, investigations and review of literature. The hypothesis by different authors to explain the cause of pain as reported is (a) increased localized metabolism,(b) irritation of joint capsule, attachment at sclerotic areas and (c) increased intraosseous pressure due tovenous stasis [5,6]. Weisz has reported an incidence of pain and joint effusin as 20%. MRI does not contribute to diagnosis but can help to rule out malignancy [7]. Each lesion of OPK on MRI is a small dark spot on both t1 and t2 weighted images as it is composed of mature dense bone. Bone scan findings are usualy normal.

Conclusion

The discovery of bone spots on X Rays is always disturbing and more commonly benign lesions must be in mind before jumping on to diagnose serious malignant conditions, notably osteoblastic metastasis on the basis of lack of internal architecture, irregular margins,the periarticular distribution and relative sparing of skull. Therefore it is important that an accurate diagnosis is made. Despite the fact that osteopoikilosis is a very rare condition that most physcians are not familiar with, It is valuable to take it into consideration, particularly when diagnostic issues on bone radiography occur and severe pain at the adjacent joints co exists.

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Conflict of Interest

No conflict of interest.

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