AMERICAN JOURNAL OF DIAGNOSTIC IMAGING, 2018
VOL 3, NO. 1, PAGES 12–14
10.5455/ajdi.20170902061321

eJManager

CASE REPORT

Open Access

Crooked painful sacro-coccygeal region as presenting feature of osteomalacia – A case report

Ganesh Singh Dharmshaktu, Binit Singh, Alamgir Jhan
Department of Orthopaedics, Government Medical College, Uttarakhand, India

ABSTRACT

Osteomalacia is a result of severe and uncompensated vitamin D or phosphate depletion. It has a characteristic clinical, biochemical, and radiological profile. The resultant softening of the bone may lead to varied patterns of deformities affecting axial as well as appendicular skeleton. The unexposed parts of the skeletal system like lower spine may also bear the brunt of deformity due to cyclic physiological loading in the settings of weakened bone. Very few reports are there describing a severe sacral and coccyx deformity along with other radiological features as pointer toward the diagnosis of a generalized metabolic condition like osteomalacia. Radiograph of a young lady with severe coccydynia revealed severe sacro-coccygeal deformity along with other features like tri-radiate pelvis, pseudo-fractures, bilateral secondary protrusion of hip and underlying osteopenia. The deformity might be secondary to bone softening and cyclic loading over the affected area due to long-standing osteopenia.

ARTICLE HISTORY

Received September 02, 2017
Accepted December 13, 2017
Published January 08, 2018

KEYWORDS

Osteomalacia; vitamin D deficiency; coccygodynia; treatment; triradiate pelvis; deformity; coccyx

Introduction

Vitamin D deficiency is a key defect in osteomalacia with less common causes being phosphate or calcium depletion [1]. It has many extrinsic, intrinsic, and acquired causes. In certain parts of the world, gastrointestinal disorders and hypophosphatemia are becoming more common causes. Developing countries witness most number of affected cases despite adequate sun exposure. A careful history and examination coupled with judicious use of investigative battery is a key to early diagnosis and proper treatment.

Case Report

A 19-year-old young girl presented to us with severe coccyx pain since last eight months. She consulted many practitioners of modern as well as indigenous medicine with only transient relief. The pain increased on prolonged sitting and walking and relieved somewhat while recumbent. There was history of vague generalized pain also but not as severe as coccyx pain. There was no radiation, referral, or diurnal variation present. On clinical examination, there was tenderness and pain was localized to coccyx region. There was associated protrusion of coccyx that was abnormal compared to normal people. The further examination could not be done as the patient and attendants refused it. She spent most of her time indoors at ground floor in a crowded locality. There was history of lack of adequate sun exposure due to socio-religious prejudices. She was having difficulty in walking and performing activities of daily living since last month due to severe pain.

The radiograph of the coccyx lateral view showed a deformed and crooked coccyx with increased curvature corresponding to localized prominence clinically. On closer observation, sacral deformity was also noted thus making it deformed sacro-coccygeal deformity (Figs. 1 and 2). The bone quality was poor with signs of osteopenia. Further radiograph of pelvis was evaluated along with relevant biochemical studies. The pelvis was severely osteopenic with narrowed pelvic outlet and severe

Contact Ganesh Singh Dharmshaktu ✉ drganeshortho@gmail.com Department of Orthopaedics, Government Medical College, Uttarakhand, India.

© EJManager. This is an open access article licensed under the terms of the Creative Commons Attribution Non-Commercial License (http://creativecommons.org/licenses/by-nc/3.0/) which permits unrestricted, noncommercial use, distribution and reproduction in any medium, provided the work is properly cited.

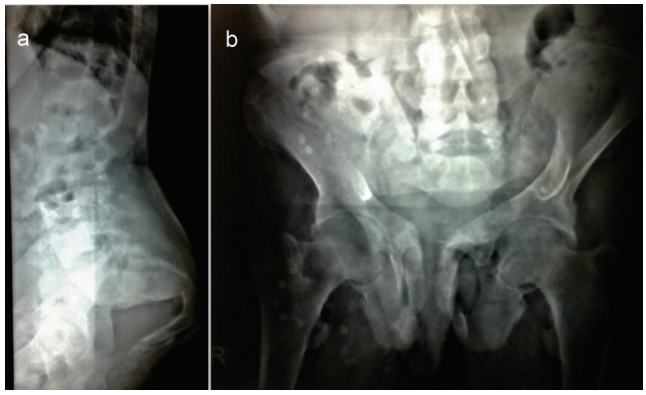


Figure 1. Lateral skin diagram showing severe sacro-coccygeal deformity along with poorly mineralized skeleton (a) and radiograph with characteristic deformities of pelvis with bilateral protrusio actabulii (b).

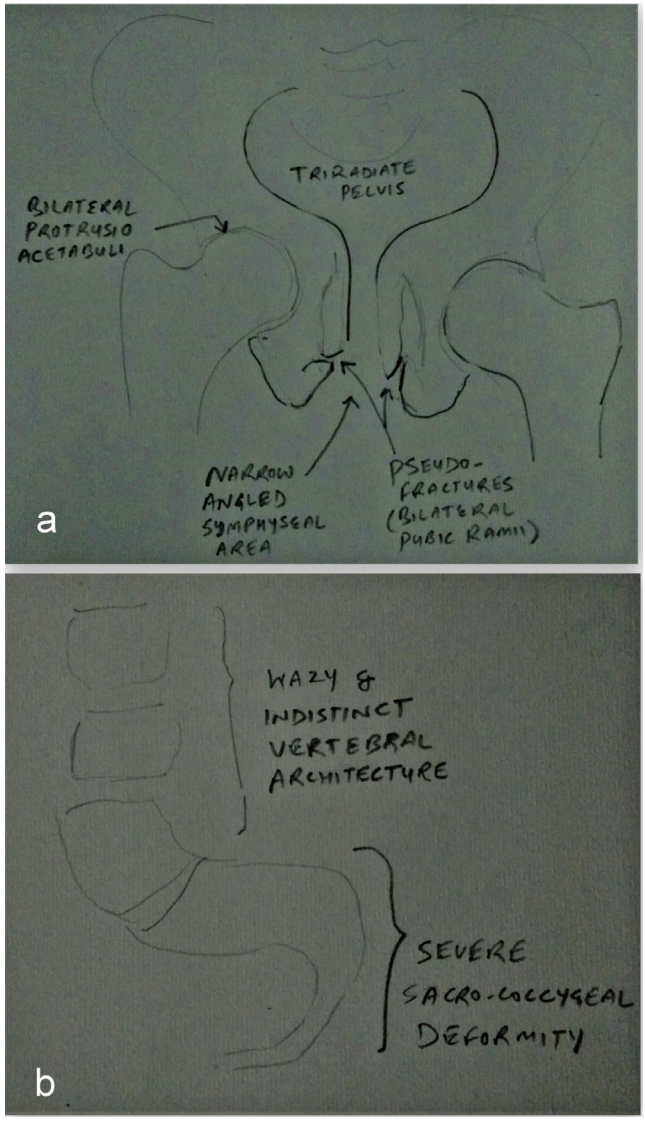


Figure 2. Schematic diagram depicting relevant bony changes due to osteomalacia including tri-radiate pelvis, pseudofractures, and bilateral secondary protrusio in anteroposterior view (a). Lateral view (b) showing severe crooked sacro-coccygeal deformity with osteopenia of vertebral column.



Figure 3. Follow-up radiograph showing improved skeletal density at 2 and 6 months follow-up.

protrusion of bilateral femoral heads into acetabulam along with tri-radiate shape. The picture depicted softened bone and resultant deformities and suggested metabolic disorders affecting the skeleton. The biochemical markers corresponded with hypocalcemia, hypophosphatemia, hypoproteinemia and increased alkaline phosphatase. A provisional diagnosis of osteomalcia was made and appropriate treatment started.

Apart from protein, calorie, and calcium rich diet, early sunbath was advised. The pharmacotherapy was started with 6 lacs IU of vitamin D3 injections (Arachitol) once weekly with daily 1,000 mg of elemental calcium was started. The patient showed a dramatic response in clinical profile with decreased pain over the course of therapy of three months. The therapy was monitored by biochemical markers till within normal limits. The serial radiographs at two and six months showed decreased osteopenia and increased bone density (Fig. 3). The patient was pain free and performing activities of daily living at final follow up at six months. The patient has been instructed for periodic review and adherence to bone healthy habits and advises.

Discussion

Osteomalacia clinically manifests as constellation of symptoms and signs, some of them very characteristic of the condition. In early stages, however, a high index of suspicion necessitates use of appropriate workup to diagnose and treat the affected cases. Most of the clinical features are based on mechanisms yet not fully understood [2]. The clinical complaints may often be very vague and nonspecific. Endemic regions have skeletal deformity cases more commonly than developed ones [3].

Bone pain is often presenting complaint and thought to be caused by hydration of undermineralized matrix stretching the periosteum [4]. Associated muscle weakness adds to the difficulty in using the affected body parts [5]. The common

deformities of skeletal system include tri-radiate pelvis, protrusio acetabuli, coxa vara, kyphosis, and chest wall deformities. The concomitant coccyx deformity has not been attributed in literature. Osteopenia, Looser zones, described deformities with occasional stress or pseudo fractures are widely encountered radiological features. Our case had pelvic deformities with tri-radiate shape and narrow pubic canal, severe osteopenia, and crooked coccyx deformity leading to refractory coccygodynia.

Other defromities that are described are kyphoscoliosis and increased concavity of vertebral endplates in the spine. Pseudofractures or Looser zones are common features and usually involve pubic ramii, scapulae, ribs, long bones, and femoral neck as bands at right angle to long axis of bone. Serum biochemical markers often have hypocalcemia, hypophosphatemia, and increased alkaline phosphatase [1,6]. A low serum 25-hydroxyvitamin D is ancillary and may serve as prognostic marker. Tetracycline-labeled bone biopsy helps in diagnosis in difficult cases [7]. Radionuclide scans and bone mineral density measured by dual energy X-ray absorptiometry (DEXA) scan of relevant sites are used to further evaluate the disorder. DEXA scans are important in classifying the patient as per World Health Organisation definitions but they are also prone to error on estimation between large and small bones and those with cortical and cancellous bones [8]. Quantitative CT is another investigation that does volumetric assessment of trabecular and cortical bones and is helpful in other ways [9]. Micro CT has more resolution thus better visualization of trabecular bones. Magnetic resonance imaging may provide similar finer details but both the techniques have technical challenges and are prone to motion artifacts apart from being expensive and not readily available [10].

The present case highlights the importance of high index of suspicion in cases of prolonged pain with history and clinical profile suggestive of inadequate sun exposure. Apart from it, understanding of uncommon presentation of severe deformity as a result of generalized bone disorder must be a differential diagnosis. Evaluation of other features of metabolic disorders in surrounding or distant sites helps in diagnosis. Comprehensive study of metabolic profile of the patient related to bone health, however, could not be done in our case due to financial limitations. A compliant patient and treatment along with good follow-up positively affects the functional outcome.

References

[1] Parfitt AM. Osteomalacia and related disorders. In: Avioli LV, Krane SM (eds) Metabolic bone disease and clinically related disorders, Saunders, Philadelphia, pp 329–96, 1990.

[2] Bhan A, Rao AD, Rao DS. Osteomalacia as a result of Vitamin D Deficiency. Endocrinol Metab Clin North Am 2010; 39:321–31.

[3] Mathew JT, Seshadri MS, Thomas K, Krishnaswami H, Cherian AM. Osteomalacia: fifty five patients seen in a teaching institution over a 4 year period. J Assoc Physicians India 1994; 42:692–4.

[4] Holick MF. Vitamin D deficiency. N Engl J Med 2007; 357:266–81.

[5] Russell JA. Osteomalacic myopathy. Muscle Nerve 1994; 17:578–80.

[6] Basha B, Rao DS, Han ZH, Parfitt AM. Osteomalacia due to vitamin D depletion in the US. Am J Med 2000; 108:296–300.

[7] Ramser JR, Frost HM, Frame B, Arnstein AR, Smith R. Tetracycline-based studies of bone dynamics in rib of 6 cases of osteomalacia. Clin Orthop Relat Res 1966; 46:219–36.

[8] Guglielmi G, Muscarella S, Bazzocchi A. Integrated imaging approach to osteoporosis: state-of-the-art review and update. Radiographics 2011; 31(5):1343–64.

[9] Adams JE. Advances in bone imaging for osteoporosis. Nat Rev Endocrinol 2013; 9(1):28–42.

[10] Burghardt AJ, Link TM, Majumdar S. High-resolution computed tomography for clinical imaging of bone microarchitecture. Clin Orthop Relat Res 2011; 469(8):2179–93.