

Resolution of “salt and pepper” appearance of the skull with vitamin D therapy

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ABSTRACT

Chronic hypovitaminosis D leads to state of decreased mineralization and generalized osteomalacia. It also results in secondary hyperparathyroidism causing increased bone turn over and decreased bone mass, manifested radiologically as a “salt and pepper” appearance in skull, subperiosteal resorption, bone cysts and lytic lesions. In this case, a young male patient with hypovitaminosis D and secondary hyperparathyroidism, radiological features show resolution of “salt and pepper” appearance of the skull with vitamin D in 11 months and regression of other lytic lesions.

Key words: Osteitis fibrosa cystica, salt and pepper appearance, secondary hyperparathyroidism, vitamin D deficiency

INTRODUCTION

The adequacy of vitamin D is required primarily for calcium homeostasis and bone integrity. Patients with chronic vitamin D deficiency develop secondary hyperparathyroidism. With increasing severity of hypovitaminosis D generalized osteomalacia results and patients progress from states of increased bone turnover and decreased bone mass, to state of near absent mineralization.^[1] We report a case of chronic vitamin D deficiency in a young male patient leading on to secondary hyperparathyroidism, with normocalcemia and radiological features of hyperparathyroidism, with resolution of these features subsequent to vitamin D supplementation, in just 11 months.

CASE REPORT

A 27-year-old male patient, presented with the complaint of pain in legs and difficulty while walking for past 4 months.

He also complained of pain and stiffness in the lower back for the same time. Patient could walk only with support for the past 1 month. There was no history of any trauma to the back. No history suggestive of bladder involvement, no positive or negative sensory symptoms. Patient was on antidepressant medication for the last few years and housebound for the past few months. Patient's examination revealed no features suggestive of spinal cord involvement; however, power in both lower limbs was grade four out of five in the proximal and distal muscles. No features suggestive of sensory involvement were seen. His systemic examination was unremarkable.

Investigations revealed hemoglobin 14.9 g/dl, total leucocyte count 6800/ μ l, platelets 212,000/ μ l, blood urea 20 mg/dl, serum creatinine 0.73 mg/dl, serum sodium 142 mEq/l, serum potassium 3.4 mEq/l, serum chloride 108 mEq/l, serum calcium 9.5 mg/dl, serum phosphorus 2.7 mg/dl, serum alkaline phosphate 2347 U/L, serum albumin 5.1 g/dl. Urine examination was unremarkable. Plasma parathyroid hormone (PTH) was 754.0 pg/ml and serum 25-hydroxyvitamin D₃ (25[OH] D₃) was 4.60 (normal > 20). His X-ray skull showed salt and pepper appearance [Figure 1], X-ray pelvis showed loosers zones with marked subperiosteal resorption in the region of pubic rami [Figure 2], X-ray hand showed resorption of the terminal phalanges with the presence of a cortical cyst in the metacarpal [Figure 3]. X-ray lumbosacral spine showed osteoporotic changes with

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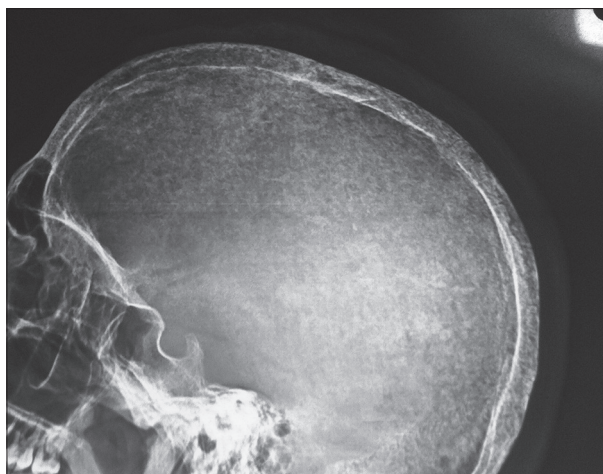


Figure 1: X-ray skull lateral view (July 2012) showing endosteal resorption with reduced mineralization of the outer and inner table of the skull - suggestive of salt and pepper appearance of the skull



Figure 2: X-ray pelvis anteroposterior view (June 2012) showing reduced bone density with areas of endosteal resorption involving the pelvic bones. Areas of reduced density resembling linear cortical defects-involving bilateral superior and inferior rami - suggestive of pseudofractures

central collapse of lumbar vertebrae and biconvex disc spaces [Figure 4]. His computerized tomography (CT) neck with contrast was normal, sestamibi scan showed diffuse uptake in the bones, with no evidence of parathyroid adenoma discernible in the parathyroid scan.

A diagnosis of vitamin D deficiency with a secondary hyperparathyroidism with normocalcemia was made. He was given vitamin D and calcium supplementation and his general condition improved over the next few months. Power in musculature improved with resolution of pain. His serum showed PTH levels, after 9 months, of 59.0 pg/ml and calcium 8.9 mg/dl. X-rays skull, hand and pelvis showed remarkable improvement with resolution of salt and pepper appearance, regression of osteopenic changes and cortical cyst in a span of 11 months [Figures 5-7].

DISCUSSION

Osteomalacia in adults results from inadequate vitamin D levels. Bone pain, tenderness, muscle weakness and difficulty walking are all common clinical manifestations of osteomalacia. Diagnostic work-up involves biochemical assessment of vitamin D status.^[2]

Our patient, a young male had a history of having negligible outdoor activity, for the past many months. He presented with severe osteomalacia with myopathy, with markedly elevated alkaline phosphatase levels, reduced serum vitamin D levels and elevated PTH levels. His serum calcium and phosphorus and creatinine were within normal range. It is rare to see such a biochemical and clinical profile in a young individual, with no significant underlying comorbid condition.

The major source of vitamin D for children and adults is exposure to natural sunlight. Very few foods naturally contain or are fortified with vitamin D. Thus, the major cause of vitamin D deficiency is inadequate exposure to sunlight.^[3] The presence of inadequate exposure to the sun in our patient resulted in vitamin D deficiency.

Deficiency in vitamin D causes secondary hyperparathyroidism, which increases bone resorption through increased osteoclast activity, decreases renal calcium clearance, increases renal phosphorus clearance and increases serum levels of 1,25-dihydroxyvitamin D, stimulating intestinal calcium absorption. This in turn leads to high bone turnover, bone loss, mineralization defects, myopathy and falls.^[4]

Osteitis fibrosa cystica, is a manifestation of secondary hyperparathyroidism. It is characterized by generalized skeletal demineralization, subperiosteal bone resorption leading to cortical thinning, presence of brown tumors (osteoclastomas) that are manifested as lytic lesions and bone cysts that can disrupt the overlying cortex. Skull radiographs reveal a salt and pepper appearance as seen in this patient.^[5]

Vitamin D deficiency is highly prevalent in Indian population. In healthy adolescents in Delhi, 25(OH) D levels were <22.5 nmol/l in 27% and 42% of children of high and low socio-economic status, respectively; boys had higher levels than girls.^[6] In a study of 132 individuals, including people with depigmented skin, physicians, nurses, soldiers and pregnant women and their newborns living in Delhi, serum 25(OH) D levels were suboptimal in all groups except soldiers. Serum 25(OH)



Figure 3: X-ray hand anteroposterior view (July 2012) showing marked subperiosteal resorption with frayed cortical outline involving the metacarpals and the phalanges. Expansile lytic lesion is seen involving 4th metacarpal. No break in cortex seen - suggestive of Brown tumor

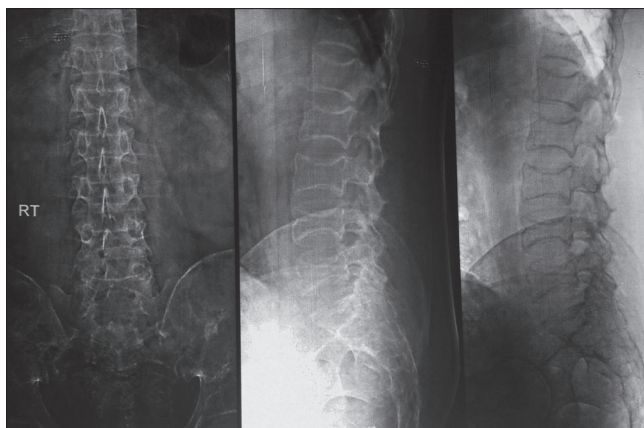


Figure 4: X-ray lumbar spine anteroposterior/lateral view (June 2012) shows central wedging involving L4 and L5 vertebrae with endplate biconcavity. Visualized vertebrae show reduced density with sclerosed end plates giving Rugger Jersey spine appearance



Figure 5: X-ray skull lateral view (May 2013) shows resolution of salt and pepper appearance with reduced areas of resorption and improved bone mineralization



Figure 6: X-ray pelvis anteroposterior view (dated March 2013) shows resolution of lesions with improved bone mineralization



Figure 7: X-ray hand anteroposterior view (May 2013) showing improved bone mineralization with reduced subperiosteal resorption

D levels were related to direct sunlight exposure and skin pigmentation.^[7]

In patients with documented vitamin D deficiency, a cumulative dose of at least 600,000 IU administered over several weeks appears to be necessary to replenish vitamin D stores.^[8]

Our patient was treated with vitamin D supplementation and showed marked improvement in biochemical profile and the resolution of skeletal manifestation in the span of 11 months. The radiographs showed regression in the lesions, with disappearance of the classic “salt and pepper” appearance of the skull. The PTH levels too, were reduced to 59 pg/ml.

A case study using serial CT scan documented resolution of skeletal manifestations of severe secondary

hyperparathyroidism, after oral vitamin D therapy in a patient of chronic kidney disease.^[9] Another showed radiological improvement of the changes of osteitis fibrosa cystica with resolution of brown tumor 38 months after parathyroidectomy in a patient of primary hyperparathyroidism.^[10]

Of the few cited studies, showing the resolution of the manifestations of features of osteitis fibrosa cystica, none have shown the documented resolution of “salt and pepper” appearance, and the regression of other changes of severe osteopenia, in a span of just 11 months.

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