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Case Report

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Diagnosis and management of skeletal fluorosis in current scenario: a case report

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ABSTRACT

Skeletal fluorosis is a rare form of toxic metabolic disease of bone characterised by increased incorporation of fluoride in bone. Skeletal fluorosis generally occurs in endemic regions where there is increased ingestion of fluorine in water and or food over a long period of time. Fluorosis is a spectrum of disease ranging from dental, non-skeletal fluorosis to skeletal fluorosis. The disease is characterised by typical clinical and radiological features that raise the suspicion towards diagnosis of the disease. Radiographic features are characterized by generalised osteosclerosis and later ossification of ligaments, tendons and interosseous membranes. Skeletal fluorosis can be easily confused with other rheumatologic disorders. People exposed to large amounts of fluoride show dental changes much earlier than the skeletal effects. Management of fluorosis generally focuses on symptomatic treatment.

Keywords: Osteosclerosis, Ossification, Skeletal fluorosis

INTRODUCTION

Skeletal fluorosis is a type of metabolic bone disease occurring due to chronic ingestion or rarely inhalation of fluoride ions in an endemic region.[1] The most common mode of ingestion is by drinking water that has high concentration of fluoride dissolved in it or if the drinking water is stored in earthen pots (world health organisation limit of 1.5 mg/l groundwater rich in fluoride).[2] Fluorosis has a wide range of presentation and can easily be mistaken for other rheumatological bone disease. Fluorosis can be dental, non-skeletal and skeletal fluorosis. Majority of the patients remain asymptomatic and it is in the late course of disease that symptoms begin to appear in the form of skeletal pain, decreased mobility, chronic low back pain and dental changes.[2]

Skeletal fluorosis is an endemic disease in some parts of the world, especially India, where it was first noted in 1930s.[4] Since then skeletal fluorosis has been reported from all parts of the world. Skeletal fluorosis is common in regions where the ground is rich in phosphaturic acid or due to industrial exposure of toxic waste rich in fluoride.[4] Nearly 6 million people in India are disabled because of fluorosis.[5]

Skeletal fluorosis is a chronic disease that develops from prolonged exposure of fluoride over decades. At low

doses, fluoride has a therapeutic action and was previously prescribed as an osteoporosis medication.[5] At higher doses, it has toxic effects, causing dental and skeletal fluorosis.

Diagnosis of fluorosis in radiological, clinical and biochemical. Skeletal fluorosis has characteristic radiological findings that include generalised osteosclerosis, ossification of ligaments and tendons.[6] There is increased osteophytosis. Ossification of interosseous membrane of forearms is a characteristic sign.[7] Neurological complications are common in advanced skeletal fluorosis with the incidence being around 10%.[8] These complications occur due to spinal cord compression from osteophytes, calcification of posterior longitudinal ligament and ligamentum flavum. These complications are progressive and lead to muscle atrophy, radiculopathies and in severe cases can cause paraparesis and paraplegia.[8]

Management of skeletal fluorosis in mainly based on symptomatic management consisting mainly of analgesic and physiotherapy. Patients with advanced arthritis of hip and knee may require arthroplasties. Myelopathy and radiculopathies can be managed with decompressive laminectomies. Some studies report reversal of radiological changes years after stopping the exposure of fluoride.[9]

CASE REPORT

We report a case of skeletal fluorosis in a 47-year-old male belonging to a village in Haryana, India who presented in the outpatient department. The chief complaint of the patient was chronic low back with radiation to both the lower limbs associated with decreased mobility for 5 years.

The patient was a daily wage labourer in occupation who started to develop pain in his lower back which was insidious in onset and progressive in nature. Initially the pain was relieved on medication for which the patient took treatment in the form of analgesics. The pain then started radiating to both of his lower limbs since about 4 years ago for which he took medical attention and was again prescribed analgesics and supportive treatment. The patient continued his daily activities but over the course of one and a half year the pain increased progressively and the patient was not able to attend to his duties and was hence forced to be home bound. The patient kept on taking analgesics during this period on and off. The patient then went to see a general practitioner and was referred to tertiary care center for further management. The patient on presentation was ambulatory with the help of walker. The patient could only walk a distance of 5 to 6 meters before taking rest.

On examination, there was deep tenderness in lower back at around the region of lumbar third and fourth vertebrae along with joint line tenderness in bilateral knees. Straight leg raise test was positive bilaterally. There was global decrease in range of motion at bilateral hips, knees and ankle. Recorded range of motion are presented (Table 1).

Table 1: Range of motion at hip, knee and ankle.

Variables	Left	Right
Hip		
Flexion	30	35
Extension	5	5
Abduction	15	15
Adduction	10	10
Internal rotation	5	5
External rotation	10	15
Knee		
Flexion	50	45
Extension	0	0
Ankle		
Dorsiflexion	15	15
Plantarflexion	10	10

On detailed history the patient elicits that the distance that he could walk had progressively decreased over the past six months and he was now able to walk 5 to 6 meters that to with the help of a walker. The pain is associated with paraesthesia and tingling sensations in both the lower limbs. Neurological examination was done and higher mental functions were normal. Power in both the lower limb is presented (Table 2).

Table 2: Medical research council grading of power in various group of muscles.

Muscle group	Left	Right
Hip flexors	3/5	3/5
Hip abductors	5/5	5/5
Hip adductors	5/5	5/5
Quadriceps	3/5	2/5
Knee flexors	5/5	5/5
Ankle dorsiflexors	4/4	4/4
Extensor hallucis longus	5/5	5/5
Ankle plantar flexion	5/5	5/5

Dermatomal examination revealed decreased sensation of touch on the medial side of leg bilaterally. Pain, temperature, joint position sense and sense of vibration were normal bilaterally. There was absent patellar reflex bilaterally and rest of the reflexes were normal. Plantar reflex was normal bilaterally and there was no bladder bowel involvement. Varus of about 5 degrees was present in both the knees with no associated instability or laxity with decrease in range of motion as described in (Table 1).

The patient was advised radiological investigation-radiographs of pelvis with bilateral hips (Figure 1), standing radiographs of bilateral knees in both anteroposterior and lateral position (Figure 2). Radiographs of lumbar spine (Figure 3) and bilateral ankles (Figure 4).

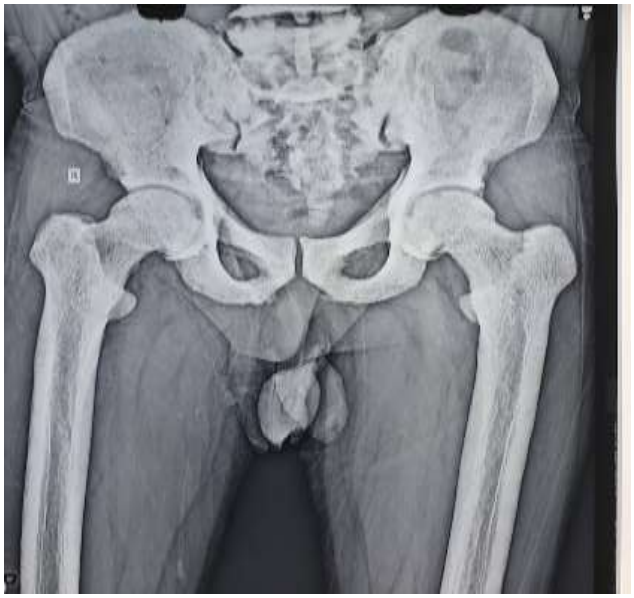

Figure 1: Radiograph pelvis with bilateral hips AP view of generalised increase in bone density, diffuse calcification of multiple ligaments including Sacro tuberous and sacrospinous ligament with global over coverage of bilateral femoral heads along with possible joint space narrowing more on left side.

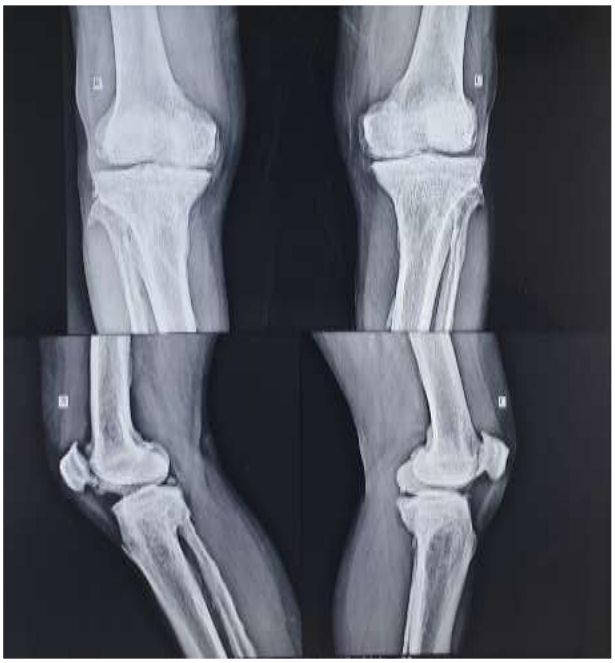

Figure 2: Radiograph bilateral knee AP and lateral view of diffuse increase in bone density, ossification of lateral meniscus in bilateral knees, markedly reduced joint space bilaterally, tibial, femoral and patellar osteophytes present, calcification of interosseous membrane right more than left. Ossification of bilateral patellar tendon, loose bodies in the knee joints bilaterally and varus malalignment in both the knees.

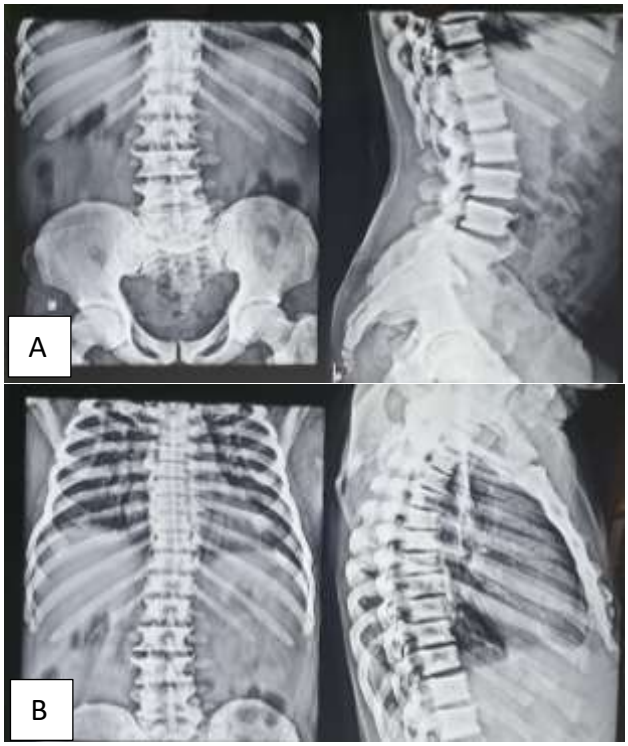

Figure 3: (A) Radiograph dorsolumbar spine AP and lateral view of diffuse osteosclerosis predominantly involving the upper and lower end plates of the vertebrae. Multiple osteophytes present. Osteophytosis and osteosclerosis of the visualised vertebral bodies along with calcification of posterior longitudinal ligament, (B) additional syndesmophytes resembling flowing wax appearance involving dorsal 5, 6, 7, 8 and 9 vertebrae.

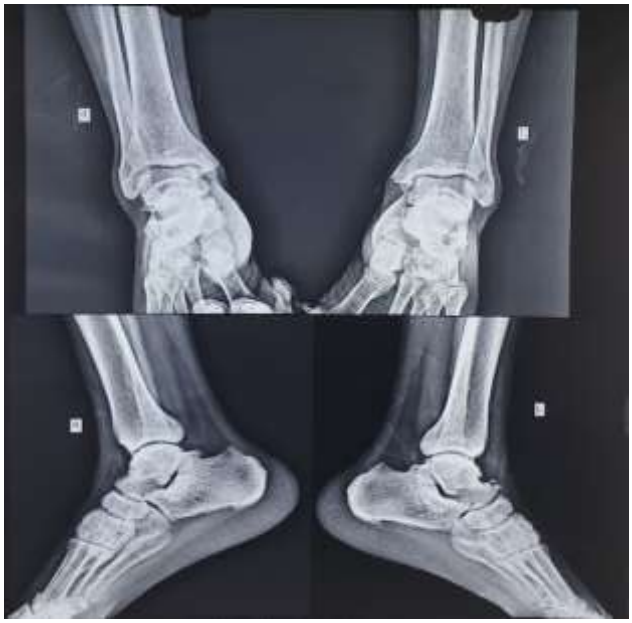

Figure 4: Radiograph bilateral ankle AP and lateral view of diffuse increase in bone density with bilateral calcaneal spurs. Multiple osteophytes present.

The above radiographs raised a high suspicion of metabolic/rheumatological disorder for which additional investigations were done. RA factor, HLA B27, Serum alkaline phosphatase and vitamin D3 levels were all normal. A radiograph of bilateral forearm (Figure 4) was also done which showed calcification in interosseous membrane that confirmed the diagnosis of skeletal fluorosis. An MRI of lumbar spine (Figure 5) was also performed because of motor weakness and radicular symptoms for which the patient presented and were the chief complaints.

Diagnosis of skeletal fluorosis with compressive myelopathy due to calcification of ligamentum flavum and diffuse disc bulge affecting was hence made involving mainly fourth lumbar nerve root and patients was planned for lumbar canal decompression surgery. Lumbar decompression was done by removing calcified ligamentum flavum and herniated disc material using laminectomies at lumbar third and fourth vertebrae along with posterior spinal interbody fusion. Analgesics, neuropathic drugs and vitamin C were started and the patient was counselled about the disease, its cause, and the further natural course of the disease and was advised to stop drinking water stored in an earthen pot and to use a water filtration device. Following decompression, the patient reported symptomatic relief in lower back pain and improved motor control of bilateral lower limb. He was put on physiotherapy and discharged on twelfth post-operative day. On further follow up patient reported complete relief in his lower back pain with slight improvement in range of motion at his hip, knee and ankle. He could now ambulate with the help of walker for longer distances.

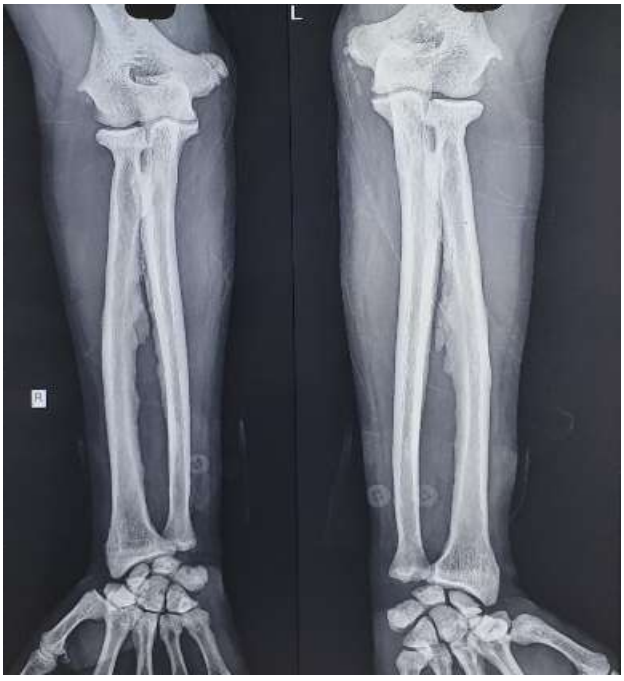

Figure 5: Radiograph of calcification of interosseous membrane in both forearms with increase in bone density.

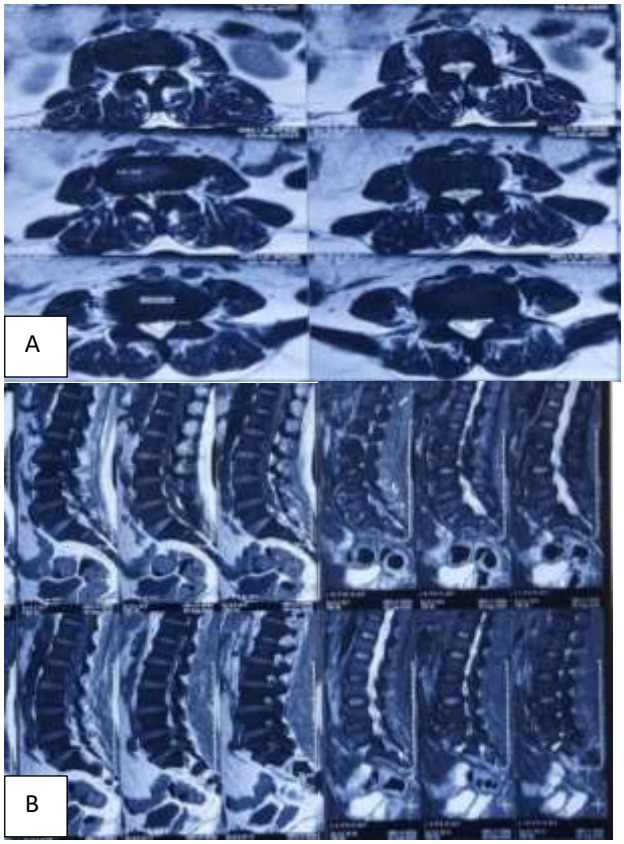

Figure 6: MRI of lumbar spine (A) axial sections through lumbar spine: diffuse disc bulge at L2-L3 and L3-L4 level causing compression of thecal sac and exiting nerve roots more on the right side, (B) T1W and T2W images: Diffuse sclerosis with loss of marrow fat in dorsolumbar spine, oOssification of posterior longitudinal ligament and ligamentum flavum and features suggestive of fluorosis.

DISCUSSION

Skeletal fluorosis is a type of chronic metabolic disease, which can lead to serious complications if not timely diagnosed and managed. Patients with skeletal fluorosis have generalised osteosclerosis with ossification of ligaments and tendons.[6] Though the diagnosis can be made clinically and radiologically, the presence of ossification of interosseous membrane of forearm is a characteristic sign pointing towards diagnosis.[7] Such radiological sign along with characteristic history of endemic origin or a history pertaining to exposure of high concentrations of fluoride can help make diagnosis. Patients generally have prolonged exposure over the years and decades and present late. Skeletal effects of fluoride are due to the incorporation of fluoride into hydroxyapatite crystal by replacing the hydroxide ion which in turn changes the biomechanical properties of the structure. Fluoride once incorporated in the crystal has a prolonged half-life of about 7 years. Moreover, fluoride alters the structure and strength of bone by altering the interaction of surface molecules i.e. RANKL and transcriptions factors like Runx2.[10]

There is ossification of posterior longitudinal ligament that leads to radicular pain and low backache.[6] These changes in bones lead to progressive decrease in range of motion that finally lead to crippling of the patient. Skeletal fluorosis in more advanced stages can lead to spinal deformities including kyphosis and canal stenosis. It also predisposes to osteoarthritis of multiple joints most commonly involving hips. There is calcification of interosseous membranes. Ossification of interosseous membrane of forearm is a diagnostic feature of the disease.[7,8] Biochemical investigations include increase in level of blood and urine fluoride levels. 24 hours urine fluoride level are specific for increased fluoride intake and help in making diagnosis of fluorosis. However, gold standard in making a diagnosis remains quantitative bone ash fluoride analysis done on bone biopsy.[4] Clinical characteristic feature is mottling of teeth due to incorporation of fluoride in dental enamel, this being the earliest clinical signs of fluorosis.[11]

Complications are many and include arthritis of hip, knee and ankle; most these patients develop radicular symptoms and myelopathy. Treatment involves cessation of exposure to fluoride along with analgesics and vitamin C.[9] In late stages there may be a need for operative intervention including joint replacements and decompressive laminectomies.

CONCLUSION

Report a case of skeletal fluorosis with late complications. Patient on presentation had diffuse osteosclerosis with ossification of ligaments and interosseous ligaments, arthritis of bilateral hip knee and ankle along with compressive myelopathy leading to progressive paraparesis. Patient was treated with decompressive laminectomy at two levels and symptomatic management including analgesics and physiotherapy. Hence, it is necessary to timely diagnose and manage fluorosis to prevent complications and improve quality of life of those affected. Screening is hence necessary in endemic areas and necessary measures accordingly.

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