

# Diffuse Skeletal Fluorosis Revealed By Tetraplegia: A Case Report

M Gaye, J Jabang, I Fondo, Y Sakho

## Citation

M Gaye, J Jabang, I Fondo, Y Sakho. *Diffuse Skeletal Fluorosis Revealed By Tetraplegia: A Case Report*. The Internet Journal of Neurosurgery. 2017 Volume 13 Number 1.

DOI: [10.5580/IJNS.52421](https://doi.org/10.5580/IJNS.52421)

## Abstract

**Objectives:** To report the first case of diffuse bone fluorosis revealed by tetraplegia in Senegal

**Setting:** Hospital General Grand Yoff Dakar (HOGGY) BP 3270 Dakar/ Sénégal

**Methods:** A 38 years old peasant living in a rural area has been hospitalized for a progressive tetraplegia in a context of brownish tooth coloration. Skeleton plain X rays demonstrated a diffuse densification of the entire bone. Neuro imaging studies with CT scan and MRI of the cervical spine showed spinal cord compression with bony spurs and calcification and thickening of the posterior longitudinal ligament. Biochemistry studies demonstrated a very high level of fluoride in the blood and urine.

**Results:** We performed a cervical laminectomy, but unfortunately the patient later from a bronchopneumonia caused by aspiration.

**Conclusion:** This report gives us the opportunity to make a literature review and to discuss the pathophysiology of the spinal cord compression. The stiffness of the spine increases the surgical risk.

## INTRODUCTION

Unfortunately, high levels of fluoride exist in the Western area of Senegal leading to the contamination of underground water and to continuous ingestion of such water. As a result, human fluorosis is endemic in this region. The most common occurring sign is brown discoloration of the teeth of the people living in areas with contaminated fields and water. Vertebral involvement of fluorosis is rare. Few articles have been published on the topic of spinal fluorosis. This is a first published sub-Saharan case of cervical myelopathy revealing diffuse skeletal fluorosis.

## CASE REPORT

A 38-year-old man, living in a rural area 100 km from the capital Dakar, has been complaining from neck pain for 2 years and recent left upper limb neuralgia and progressive weakness of the four limbs with neurogenic claudication. For the past 3 months he was unable to walk. All his close family members have brownish dirty discoloration of the teeth.

The patient was alert and well oriented. His dentition was

poor with a brownish discoloration. He suffered from incomplete spastic tetraplegia, sphincter dysfunction with constipation, and bladder retention.

Plain cervico-thoracic spine X rays and CT scan showed (fig 1, 2) an important bone densification and hypertrophy of this segment we call “vertèbre d’ivoire”.

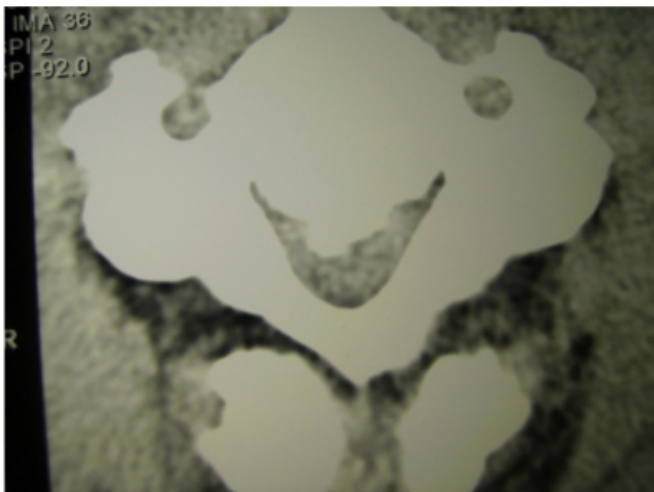
**Figure 1**

X rays of the thoracic spine showing bone densification



**Figure 2**

Axial CT scan: bone thickening which narrows dramatically the spinal canal.



We noticed calcification and ossification of the posterior longitudinal ligament narrowing the vertebral canal (fig 3). Whole skeleton X rays showed a diffuse bone densification affecting all the spine, pelvic and femoral bones. Radiography of the skull showed a thickening of the skull.

**Figure 3**

Sagittal reconstruction CT scan: bone and posterior longitudinal thickening which narrows dramatically the spinal canal.



Cervical spine MRI (fig 4) demonstrated a narrowing of the cervical canal and T2 intra medullary hyper signal.

**Figure 4**

T2 Sagittal MRI of the cervical spine: narrowing of the canal and presence of an intramedullary hypersignal.



Blood and urine fluoride levels were very high: blood fluoride level was 293,8  $\mu\text{g/l}$  (usually below 119  $\mu\text{g/l}$ ), and urine fluoride level was 19194,8  $\mu\text{g/l}$  (normal rate below 500  $\mu\text{g/l}$ ) Blood calcium was 88.54 mg/l, phosphorus was 24mg/l, protein electrophoresis was normal, protein level

was 81.70g/l and creatine was 8.89mg/l.

A medullogram didn't show any cellular proliferation or infiltration. Prostatic serum antigen was 0.610ng/l. No anaemia or leucocytosis was found.

The patient underwent laminectomy of C3 to C5 and during surgery we noticed that the posterior arch of the spine had hypertrophy and was very stiff.

Unfortunately, 24 hours post-surgery he suffered from a bronchopneumonia after aspiration leading to a respiratory distress and death.

### **DISCUSSION**

Skeletal fluorosis is a serious health problem in Senegal. It is caused by a prolonged intake of excessive amount of fluoride through water or food or both. Sometimes fluoride intoxication may be revealed by neurological manifestations. Vertebral bone, ligament flavum, and posterior longitudinal ossification as well as hypertrophy can lead to spinal cord or root compression in the stenotic canal [2]. Fluoride stimulates the osteogenic process. Fluoride intoxication includes three features which occur isolated or combined: dental fluorosis, diffuse calcification of bone, and calcification of bony insertion of many ligaments like intervertebral discs or inter osseous membrane [5]

Indian authors [1, 2, 3] published several articles on this issue. Some cases reports came from North Africa and others countries [4, 5]. Joly [1] from Punjab in 1968 described 62 cases of fluorotic myelopathy. In 2008, Younes and all [5] in Tunisia published a cervical myelopathy revealed by bone fluorosis.

To our knowledge our case is the first case of myelopathy due to skeletal fluorosis reported in the sub-Saharan countries. One can only speculate on the frequency of this pathology which may be under estimated. It means that we need a national survey in the affected regions to get a better picture of this problem.

A high index of suspicion of skeletal fluorosis exists when plain bone X rays demonstrate bone thickening and hyperdensity. Dental pigmentation is typical. One should rule out some others conditions like hyperparathyroidism, Paget disease or prostatic vertebral metastatic cancer.

Spine CT scan and MRI is helpful to make critical to

manage neurological complications. They show calcifications and hypertrophy of the posterior longitudinal ligament and or ligamentum flavum which potentially compromise the spinal canal.

Cervical myelopathy is a frequent neurological manifestation of skeletal fluorosis. Jolly and all. demonstrated that fluorotic myelopathy was mainly located in the cervical spine (60%) followed by thoracic spine (19%). In our case, a diffuse skeletal fluorosis lead to tetraplegia.

Our patient showed very high blood and urinary fluoride levels. Reddy said that urinary fluoride levels are the best indicators of fluoride intake. 24 hours sampling of urine are a reliable way to measure such levels.

Spinal surgery is difficult to perform in cases of fluorotic myelopathy as stated by Reddy. In fact, the rigid cervical spine does not allow for easy intubation and a good positioning; bone resection is hard and a high risk of post-operative complications exists because of restrictive chest movement. Our patient died 24 hours after surgery from a bronchopneumonia due to aspiration. Reddy noted that surgery is of little help but Subba [3] made a report of 6 patients who underwent surgery. In his series, 3 patients improved their spasticity and for 3 it worsened.

### **CONCLUSION**

Skeletal fluorosis can involve the spinal column and give rise to a severe myelopathy. The management of this neurological complication is still challenging. In fact, in an endemic area with high fluoride containing food and water, decision makers should implement a program to build defluoruration plants to limit the population's exposure.

### **References**

1. Joly SS, Singh BM, Math MC (1968): Epidemiological, Clinical, Biochemical study of skeletal fluorosis in Penjab. British Medical Journal, Oct: 425-428
2. Reddy R D (2009): Neurology of endemic skeletal fluorosis. Neurology in India, 57(1)
3. Subba Rao B, Jaraknath (1992): Ossification of the posterior ligament and fluorosis. Journal of Bone Joint Surgery, 74: 469-470
4. Wenbao Wang, Linghua Kong, Heyuan Zhao, Ronghua Dong, Jianjiang Li, Zhanhua Jia, Ning Ji, Shucui Deng, Zhiming Sun, Jing Zhou (2007): Thoracic ossification of ligamentum flavum caused by skeletal fluorosis. Eur Spine J 16:1119-1128
5. Younes M, Mohamed I. (2008): Cervical myelopathy revealed bone fluorosis. Revue neurologie, 164: 185-188

**Author Information**

**Magatte Gaye, MD; Assistant Professor and Consultant in Neurosurgery**

Université Cheikh Anta Diop (UCAD) and Hospital General Grand Yoff

Dakar, Senegal

**John Jabang, MD; Chief Resident in Neurosurgery**

UCAD

Dakar, Senegal

**Ibrahima Aladji Oumar Fondo, MD; Consultant in Neurosurgery**

Hospital General Grand Yoff

Dakar, Senegal

**Youssoufa Sakho, MD; Professor in Neurosurgery**

UCAD; Head of the Department of Neurosurgery Hospital General Grand Yoff

Dakar, Senegal