A CASE OF ENDEMIC FLUOROSIS OF BONE IN THE CAPE PROVINCE

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Fluorosis of bone is recognized as occurring in South Africa from the consumption of drinking water containing a high level of sodium or calcium fluoride. In other countries such as the United States of America and India it has also been recorded in areas where the drinking water was similarly affected. Industrial poisoning due to the inhalation of dust containing fluoride has not been recorded in South Africa, but in other countries where cryolite, a fluoride of sodium and aluminium, is used in the production of aluminium it is well known. Similarly, workers with rock phosphate are apt to suffer from fluoride poisoning.

Ockerse,[1] in a dental survey among the population of the

Cape Province, noted changes in the teeth of school children, typical of fluorosis. In another investigation,[2] in the Pretoria district, advanced changes due to fluorosis were found in the skeletons of 6 Natives whose teeth were free of fluorosis. For 19 years these Natives had drawn their drinking water from a bore-hole containing a high fluorine content. Fichardt *et al.*[3] reported another such case from the Pretoria district, where the water contained 11·78 parts per million of fluorine, but in this teeth were affected.

Because of the widespread incidence of dental fluorosis in the children of the North-West Cape and because of the high fluorine content of the water, Ockerse[1] predicted that ultimately cases of severe bone changes similar to those recorded in India by Shortt *et al.*[4] and in the US would be discovered. The case we describe belongs in this category.

L.F., a non-European female aged 30 years, was admitted to hospital on 15 August 1957 complaining of a cramplike pain in the right iliac fossa, present for about 10 days and becoming progressively worse. Vomiting commenced 3 days after the onset and on admission the vomitus was frankly faeculent. About 12 years ago bilateral ureteric transplants had been performed elsewhere, after attempted repair of a severe vesico-vaginal fistula had failed. Until the onset of the present illness she had considered herself reasonably fit and well, and was employed as a domestic servant.

On examination the patient was found to be severely dehydrated, drowsy, and in a state of collapse and quite unable to give a rational account of herself. The abdomen was generally distended and rigid, the site of maximal tenderness appearing to be located in the left flank. Bowel sounds were absent. B.P. 100/60 mm. Hg. Pulse rate 140 per minute. W.B.C. 38,000 per c. mm. Hb. 10·5 g. %. Straight X-ray revealed multiple fluid levels in the small and large bowel. The appropriate fluids were replaced and the patient made reasonably fit for operation.

Operation. The abdomen was entered through a right paramedian incision and immediately a great deal of foul-smelling pus was encountered. Dense adhesions were found in the region of the pelvic colon and a perforation was located in the sigmoid. The site of the previous ureteric transplant was not identified as the patient's condition did not warrant a prolonged search being made. The perforation in the colon appeared to be quite unrelated to the previous operation and, in fact, a satisfactory cause was not found. There was no evidence of diverticulitis. The perforation was closed and a transverse colostomy performed. The wound was drained and the abdomen closed in layers. The patient made an uneventful recovery, and the colostomy was closed intraperitoneally on 22 October.

When the patient became ambulant, it was observed that her gait was peculiar, and examination revealed an almost complete loss of spinal flexion (Fig. 1), with limitation in varying degree of all spinal movements.

Fig. 1. Showing maximum degree of flexion of spine.

An attempt was made to link this clinical finding with the radiological appearance of the bones. The patient's teeth examined by Mr. W. Rosenblatt were reported as being free of the chalky patches and pitting usually associated with fluorosis.

X-ray Changes

1. *Skull:* No lesions.
2. *Cervical Vertebrae:* Moderate increase in density with trabeculation largely obscured by new bone deposited between trabeculae. Slight calcification of anterior longitudinal ligament.
3. *Thoracic Vertebrae:* Moderate increase in density with a pronounced degree of calcification of muscular and ligamentous attachments to the transverse processes and other prominences (Fig. 3). Some kyphosis of the thoracic region. The disc spaces are a little ballooned so that some mild osteoporosis of the bodies is likely.
4. *Ribs:* Upper ribs very little involved but the lower ribs show extensive calcification of ligamentous and muscular attachments without any substantial increase in rib density (Fig. 4).
5. *Lumbar Vertebrae:* Very marked increase in density with almost complete obliteration of bone structure of the bodies in the A.P. view. There is some calcification of ligamentous attachments
6. *Pelvis:* Very marked increase in bone density but some trabecular structure persists. Ligamentous attachments calcified,

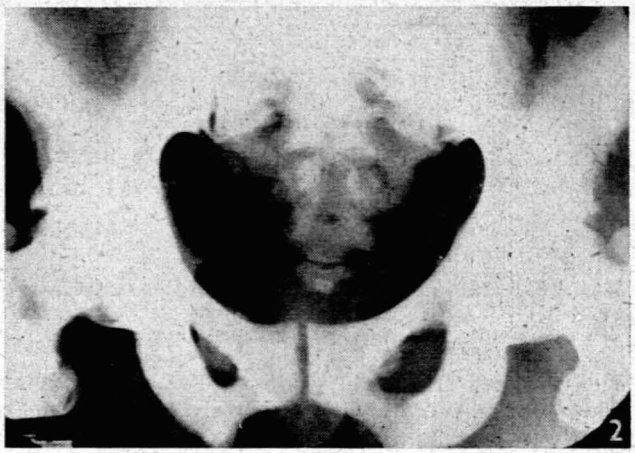

Fig. 2.

but particularly the sacro-tuberous and sacrospinous ligaments at their outer ends (Fig. 2), as described by Stevenson.[9]

7. *Femora:* Slight increase in density of femoral heads, especially the right, and prominence of the linea aspera; otherwise normal.
8. *Tibia and Fibula:* Some increase in density of upper ends of tibiae and fibulae on both sides, with substantial calcification of muscular and ligamentous attachments of upper halves of both bones. Symmetrical changes.
9. *Feet:* Normal.
10. *Humeri:* Normal scapulae. Ligamentous and muscular calcification on axillary borders.
11. *Radius and Ulna:* Marked calcification of attachments of inter-osseous membrane in upper halves (symmetrical). (Fig. 5).
12. *Hands:* Normal.
13. *Teeth:* Upper and lower incisors show normal peri-apical bone.

DIAGNOSIS

Fluorosis of the skeleton can be recognized by a progressive rigidity of the trunk in patients who have been exposed to high fluoride drinking-water for a period of approximately 20 years. In patients who have been exposed to dust containing fluoride in industrial processes, the period is reduced to about $2\frac{1}{2}$-3 years.[5] Where children have been continuously drinking water with a high fluorine content, dental changes will occur if the absorption of fluoride occurred before calcification of the permanent teeth, i.e., before 8 years of age.

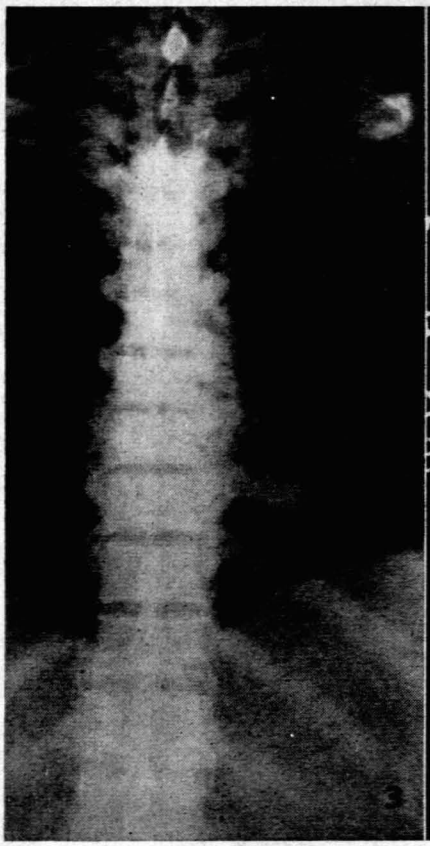

Fig. 3.

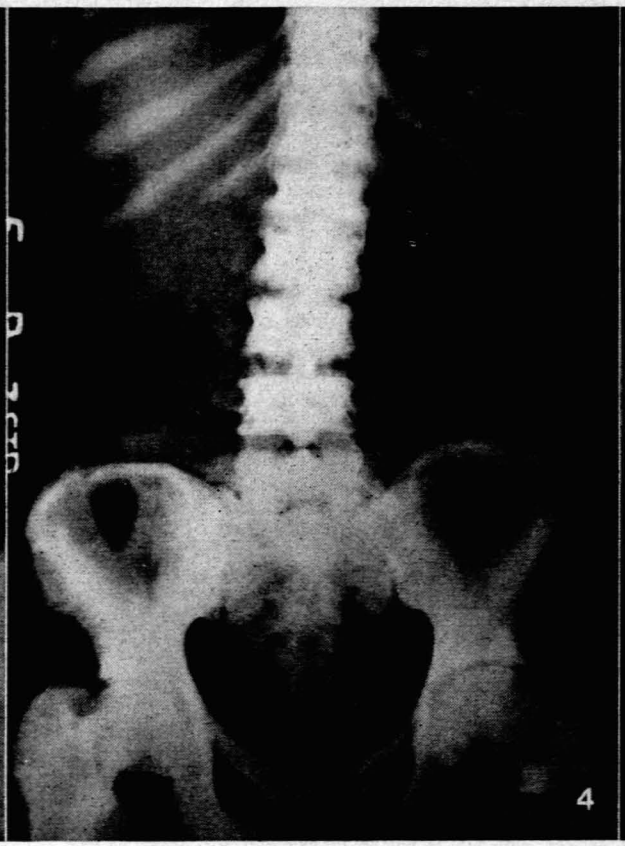

Fig. 4.

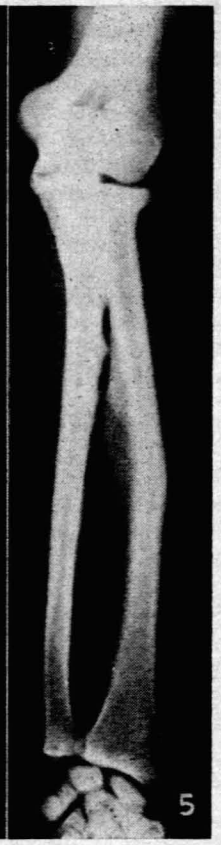

Fig. 5.

With continued drinking of such water the skeletal changes follow 20 years later, so that it is at the age of 30 years that the patient presents the clinical changes.[4] The absence of dental changes accords with this patient's story that she came to the farm as an '*opgeskote kind*' and that her drinking water was drawn from the same spring for 20 years subsequently until a new bore-hole was sunk about a year ago.

Ideally, to prove fluorosis it is necessary to show that the patient's water supply contained a high level of fluorine, that the bone contained excess fluoride, and that the urinary excretion of fluoride was raised. In our case no data are available on any of these points, and they will be dealt with in turn.

(*a*) *Water Supply.* She states that the spring was filled in a year or more ago, so that a sample cannot be obtained. The location of the farm is in the South-West Karoo on the boundary of the Ceres and Sutherland districts, where fluoride is reported by Ockerse[1] (quoting Wasserstein) as occurring in all horizons of the Karoo sediments. However, in his dental survey he did not investigate this particular area as thoroughly as he did the Kenhardt and Gordonia areas, so that accurate estimations of the fluorine in the water are not available. Allowance must also be made for seasonal variations in the fluorine content of the water derived from bore-holes (Ockerse[1]). This patient is not aware of stiffness as an endemic complaint in her area, nor has she heard mentioned the occurrence of dental changes in farm animals.

(*b*) *The Fluorine Content of Bone.* It did not seem justifiable to recommend a bone biopsy to determine the fluorine in the ashed bone, for this procedure would not have assisted her treatment. Leone *et al.*[6] examined the bones of a woman of 52 years who throughout her lifetime had been exposed to a high level of fluorine in her drinking-water and who died of a cerebral haemorrhage. She was one of a sample of members of the population of a Texas town whose domestic water supply contained 8 parts per million of fluorine. This investigation formed part of a 10-year comparative study by the US Public Health Service of two populations, one with a high fluorine content in the water supply and one with a normal fluorine content, with a view to assessing the development of the bone changes of fluorosis. In this case, where the bone fluorine content was 0·6 mg. % (6 times the normal) no histological change was reported, but it is not stated specifically that the Haversian systems were not filled with calcium-fluoride crystals as found in animal feeding experiments by Moller and Gudjonnson[7].

(*c*) *Urinary Examination.* In this case, because of the bilateral transplantation of the ureters into the colon, it was not possible to obtain a sample of urine for assay of the fluorine content. The method used by Shortt *et al.* is a laborious one—the zirconium nitrate, sodium alizarin sulphonate indicator method—and they mention that they present their results with some reserve because of doubt of the adequacy of the technique and the method. Krebs[8] assessed the fluorine content of urine in Wilkie's two cases in Sheffield, but the method he used is not stated.

Green[9] mentions the use of a urinary fluorine assay (to detect fluorine poisoning near a factory concerned with the calcining of ironstone in Bedfordshire) in cases where possible X-ray changes are not yet present. Her method also is not disclosed.

DISCUSSION

Diagnosis in this case must, therefore, depend on the clinical features and the X-ray appearances. Essential radiological features are osteosclerosis and calcification of the ligaments and muscular attachments. In particular, the sacro-tuberous and sacro-sciatic ligaments are affected.

Stevenson and Watson[10] maintained that they had not seen this in any other condition. There are many diseases in which osteosclerosis is a feature, such as marble bones, osteoblastic neoplastic deposits etc. and there are many different causes of ligamentous and muscular calcification, but a disease state where both osteosclerosis and ligamentous calcification are prominent features is recognised only in fluoride intoxication, or, as Flemming Moller and Gudjonsson[7] pointed out, 'the changes are of a kind to which no analogy can be found in any former roentgenological experience'.

It is known that in rare cases generalized osteosclerosis may occur in renal failure, but without calcification of ligamentous or muscular attachments. This patient presented with transplantation of the ureters into the colon, and chloride retention with hyperchloraemic acidosis is known to occur in these circumstances. However, normal sodium (142, 143, 139 mEq.), potassium (2·9, 3·5,— mEq.) and chlorine (115, 102, 106 mEq.) serum estimations were recorded on 3 occasions and a raised blood urea on only one occasion, viz on admission, while dehydrated.

Shortt *et al.*[4] point out that there are 3 degrees of involvement. In the first stage of increased bone density and exaggerated trabeculation with medullary narrowing, the other causes of osteosclerosis will have to be differentiated. However, once the ligamentous and muscular attachments calcify, the disease enters the second stage, in which our case falls. The condition is progressive, so that in the third stage the trabecular structure is completely hidden by the chalk-like appearance which may extend to the skull and the metacarpals and phalanges, which are usually the last to be affected.

Because of the possibility that the patient's drinking-water may now contain a more normal fluoride content, it remains to be seen whether this patient will end up in the next 10 years in the final stages of fluoride intoxication with complete immobilization of the thorax. Breathing is then entirely abdominal and the chest becomes barrel-shaped and flattened anteriorly. Spinal cord pressure may also result from bony encroachment, with loss of sphincter control. The patient becomes bedridden but mental powers are preserved throughout.

SUMMARY

A case of endemic fluorosis of bone, but without dental lesions, occurring in a young coloured woman resident in the Ceres-Sutherland area of the Cape Province is presented.

Thanks are due to the Medical Superintendent and the Head of the Division of Surgery (Prof. J. H. Louw) for permission to record this case.

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