Evaluation of Different Clinico-Radiological Changes In Endemic Fluorosis and Correlation with Duration of Exposure at Nasipur Village of Birbhum District, West Bengal

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Abstract

Background: *Chronic fluoride toxicity mainly causes dental and skeletal fluorosis.*
Aims and Objectives: *The study describes the spectrum of clinical and radiological features in 204 patients suffering from endemic fluorosis, residing inNasipur village of Birbhum district of West Bengal.*
Results and Analysis: *The patients mainly belonged to the middle age group (41-50 years, 38.24%), and were mostly males (male : female – 2.4:1) and labourers by occupation (51.9%). Low back pain was the most common (97.05%) and crippling deformity of spine was the least common (14.7%) complaints. Diminished chest expansion was the most common (39.21%) observation on general survey. The most common abnormality on regional examination was rigidity and stiffness of spine (79.41%). Radiologically, majority of the patients had osteosclerosis (94.12%) while only 2.94% had osteopenic changes. Most patients had moderate skeletal fluorosis (49.48%), who mostly belonged to the middle age-group (41-60 years). The severity of the symptoms were seen to be directly proportional to the duration of exposure to the source.*
Conclusion: *It is necessary to diagnose fluorosis early and differentiate it from mimicking conditions.*
Keyword:*Fluorosis, low back pain, stiffness of spine, osteosclerosis.*

I.   Introduction

Chronic exposure to high level of fluoride for prolonged duration results in chronic toxicity to fluoride or fluorosis. The estimated safe and adequate intake of fluoride in adults is 1.5 - 4 mg/day, and it is less in children and those with renal disease [1,2,3]. But in endemic regions, the daily intake has been found to vary from 10 to 35 mg% and can be even higher in summer months [4].Fluorosis can affect almost all the systems of the body, with profound effects on the bones and teeth. High level of fluoride for prolonged duration leads to degradation of collagen and ground substance in bones and teeth, and thus leads to symptoms of fluorosis. In early stages, the disease may be confused with rheumatoid arthritis, ankylosingspondylosis. Later, generalised sclerosis of the bone has to be differentiated from osteopetrosis, secondaries from prostatic carcinoma, Paget's disease, rickets, osteodystrophies, etc. Thus, it is important to know the spectrum of clinical and radiographic features of fluorosis, for the diagnosis and prognosis of fluorosis and also differentiate it from other mimicking conditions. Various countries have been identified for the problem of fluorosis [5] and the problem has reached an alarming proportion in India, affecting 19 states [6,7,8] which includes West Bengal. Birbhum, Bankura, Burdwan and Purulia are the four affected districts in Bengal [9,10]. The people of our study village, Nasipur, in the Birbhum district, were found to be affected with dental and skeletal fluorosis since 1978, after four deep tube wells were established. However, after these tube wells came to be known as the source of fluorinated water, the people were stopped from drinking the water of these tube wells since last 17 years. This study assesses the magnitude of the problem of fluorosis in the Nasipur village of Birbhum district, delineates the spectrum of clinical and radiological features of fluorosis in the affected patients, and correlates disease symptoms with the duration of exposure to the fluorinated water.

II.   Material And Methods

This is a cross-sectional observational study that was conducted in the village of Birbhum district of West Bengal by the Department of Orthopaedics, Medical College, Kolkata, during the period of January 2014 to July 2014. The study includes a sample of 204 patients who were residents of Nasipur village and were suffering from endemic fluorosis. The patients in the study were evaluated by a detailed history, clinical examination and radiological examination. Detailed history included patient's identification, occupational history, personal history including water consumption and food intake, chief complaints and their duration, and past history of any major illness including hospitalisation. A through clinical examination included general survey, examination of teeth and simple diagnostic tests to note for pain and stiffness in the neck, backbone, shoulder joints and hip. Chest expansion was noted. A detailed physical examination of the back and spine, and of each and every joint of upper limb and lower limb was done. A detailed radiological examination of cervical spine, dorso-lumbar spine and lumbo-sacral spine in antero-posterior and lateral views; X-ray chest in postero-anterior view, and radiological examination of pelvis, knee, both bone leg, shoulder, elbow, both bone forearm and wrist in antero-posterior view was done. Radiographs provided the spectrum of radiological features in fluorosis and also enabled grading of skeletal fluorosis into mild, moderate and severe.[11]

III.   Results And Analysis

This study, which was done for a period of one year, on a sample of 204 patients, revealed various interesting findings. The age wise distribution of the study population showed that a maximum number of patients belonged to the 41-50 years age group, accounting for 38.24% and the average patient age was found to be 52.02 years. The youngest patient was a male 20 years and oldest patients were two females of 70 years. The sex wise distribution showed that the males outnumbered females (males - 70.58%, females - 29.42%), with a male : female ratio of 2.4 : 1(table 1, table 2).

The patients presented with a variety of clinical features including back pain, knee joint pain, leg pain, difficulty to squat, deformity of lower limbs, painful and restricted forearm movement, neurological symptoms, crippling deformity of spine, breathing difficulty, and dental fluorosis. The analysis showed that low back pain was the most common complaint (97.05%) and crippling deformity of spine was the least common complaint (14.7%) (table 4).Various abnormalities were found on general survey, of which diminished chest expansion (<2.5cm) is present in most of the patients (39.21%), followed by anaemia (<26.47%)

A spectrum of abnormalities were noted upon performing detailed regional examination of the shoulder, elbow, wrist, hand, hip and knee. Presence or absence of generalised muscle wasting was noted grossly. It was observed that maximum number of patients (79.41%) suffered from rigidity and stiffness of spine, followed by restriction of hip movement (70.78%). (table 7)
The spectrum of radiological features that were noted included osteosclerosis, osteopenia, thoracic kyphosis, irregular cortical thickening, obliteration of marrow cavity, diaphysealwidening, interosseous membrane and ligament calcification and osteophytic changes. Majority of the patients had osteopenic changes.

Skeletal fluorosis was graded radiologically into mild, moderate and severe using radiograph. The distribution of the various grades of fluorosis into different age groups was noted. It was seen that the maximum number of patients (49.48%) had moderate skeletal fluorosis while only 21.87% patients had severe skeletal fluorosis. Majority of the moderate skeletal fluorosis patients belonged to the age group of 41-60 years while most of the severe skeletal fluorosis patients belonged to the older age group of 61-70 years. (table-9)

Analysis was done, correlating the duration of exposure to fluoridated water and development of dental and skeletal fluorosis. It was seen that none of the patients in our study population were below the age of 20 years, who had an exposure period of $\leq$ 4 years to fluoridated water. Some people in the age group of 21-30 years (exposure period of fluoridated water 5-14 year) had developed dental fluorosis but not skeletal fluorosis. All patients affected by skeletal fluorosis belong to the age group of 31-70 years (exposure to fluoridated water $\geq$ 15 years).

IV.   Discussion

Fluorosis affects many systems of the body, but its main effects are on the bone and teeth. Excess fluorine interferes with collagen formation in bone and collagen, characterised by mottling of teeth (dental fluorosis). Hence, the focus of our study is on dental and skeletal fluorosis.
The people of our study village, Nasipur, in the Birbhum district have been known to suffer from endemic fluorosis since 1978, because in that year, the culprit "deep tube well" were established.

Before 1978, the villagers used to drink surface water and nobody had suffered from this type of disease. However, in 1997 these tube wells were identified as source of fluorinated water and since then the people were stopped from drinking water of these wells. Nevertheless, the problem of fluorosis is still existing in the village even after elimination of sources, thereby establishing the fact that fluorosis is an incurable disease, once exposure to the source for sufficient duration has occurred.

Our study illustrates the spectrum of clinical and radiological features in the study population and provides useful information that aids in the early diagnosis of fluorosis, and enables its differentiation from common mimicking conditions like spondylo-arthropathy. An analysis of the epidemiological characteristics of the present study show that majority of the affected patients belong to the age group of 41-50 years (38.24%), male sex (M : F – 2.4 : 1) and labourer class (51.9%). Male patients outnumbered the females in all age groups except 61-70 years of age. The possible explanation for males outnumbering the females in the below 60 years age group may be due to the fact that males in this age group mostly used to go outside home for working purpose and were more frequently exposed to the flurorinated deep tube well water as compared to females in the similar age groups. But the above 60 years age group, the number of males were lower than the number of females, due to the death of the male patients affected by fluorosis, and greater survival of the female patients as they were less affected by fluorosis. However, due to the small sample size, these results regarding the distribution according to sex may be confounding, and a larger representative population is necessary to substantiate this observation.The labourers were the main occupational group affected by fluorosis because of their greater exposure to the fluorinated deep tube well water than the other occupational groups.A similar male and labourer class predominance was noted by Jolly SS et al, Assefa G. [12, 13]

The patients in the study, presented with a variety of clinical features, of which low back pain (97.05%) was the commonest complaint followed by dental mottling (94.11%). Knee pain, leg pain, difficulty to squat, painful and restricted forearm movement, genu valgum, genu varum, neurological symptoms, breathing difficulty were the common complaints. Crippling deformity of spine was least common complaint (14.7%). Our observation regarding the mode of presentation is more or less similar to the findings made by Jolly SS, Yanzhao Wang et al, Shivashankara AR et al and Assefa G et al. [5,12,13,14]

On general survey, diminished chest expansion (< 2.5 cm) was the commonest observation (39.21%) and this finding correlates with the previous workers like Jolly SS. [12]Anaemia was the second most common observation. These anaemic patients had history of recurrent chest infection, diminished chest expansion clinically and white chest cage and spine radiologically. Considering these findings, it may be inferred that anaemia developed in skeletal fluorosis probably due to the absence of red bone marrow of ribs and spine and thus impaired erythropoiesis. [15]

Oedema, pitting in type was another important observation which was also observed in the study by Xiang QY et al. [16]Few patients also presented with jaundice. The occurrence of jaundice and pitting oedema in fluorosis could not be explained otherwise.

Spine was found to be the most commonly affected site on regional clinical examination. Restriction of spinal movement was the most common abnormality and 9 patients had no movement al all, which could be attributed to the ossification of ligamentumflavum in these 9 patients. These findings are corroborative to a previous study done by Wang W. [17]

Restriction of abduction and external rotation of shoulder joint was seen, and gross restriction was present in 5 patients, which was due to ligamental calcification, as evident by x-ray.

Varying degrees of restriction of supination-pronation movement of forearm was seen which correlates with varying degrees of calcification of inter-osseous membrane of forearm. Surprisingly, these patients had almost full range of flexion-extension without any deformity or localised tenderness. These findings are corroborative to studies done by Nayak B, Ray MM et al. [9]

Restricted wrist and hand movements, wasting of muscles of hand and difficulty to make a fist were noted in 29.41%, 23.52% and 23.52% cases respectively. Restricted movements occurred due to soft tissue and ligamental calcification around the wrist. Similar findings were reported by investigator Pandit CG et al. [18]

Varying degrees of restriction of hip (70.58%) and knee (32.35%) movements were present in our study. Some patients also presented with fixed flexion deformity of hip (67.64%), fixed adduction deformity of hip (5.88%), genu valgum and genu varum. Similar observation wasreported by investigators Teotia M et al and Shivashankara et al. [14,19]

Generalised muscle wasting, mainly involving quadriceps femoris and calf muscle were present in 14.7% patients, correlating with the observation made by investigator Witford GM. [20]

A spectrum of radiographic findings were reported in the study, of which osteosclerosis (94.12%) followed by irregular cortical thickening (58.82%) were common features and osteopenia (2.94%) was the least common feature. Our study results are corroborative to that of previous investigators like Jolly SS, Teotia M et al, Xing QY et al. [12,16,19]

Radiographic grading of skeletal fluorosis showed that majority of the patients (49.48%) had moderate skeletal fluorosis who mostly belonged to the middle age group (41-60 years) while few patients had severe skeletal fluorosis (21.87%) who mostly belonged to older age group (61-70 years). This corroborate the fact that severity of skeletal fluorosis increases with advancing age, and correlates with the observations made by Teotia M et al, Teotia SPS et al and Xiang QY et al. [11,16,19,]

Our study also correlates symptoms of fluorosis to the duration of exposure to fluorinated water. It was seen that none of the people below 20 years in Nasipur village had symptoms of fluorosis. This was because these people had been exposed to fluorinated water for ≤ 4 years, which is not sufficient for the development of fluorosis. Patients in the age group 21-30 years, had features of dental fluorosis only, as they had been exposed to fluorinated water for 5-14 years, which is the exposure period required for development for dental fluorosis but not skeletal fluorosis, once again corroborating the fact that fluorosis affects the dentine tissue earlier than the skeleton. [18,21,22,23] Patients in the age group of 31-70 years had developed features of skeletal fluorosis as they had been exposed to fluorinated water for ≥ 15 years, which is sufficient for the development of skeletal fluorosis.

Again severity of skeletal fluorosis increased with increasing age, as the duration of exposure to fluorinated water was also more for the advancing age groups. These observation imply thatfluorosis is a cumulative exposure related disease, with the severity of symptoms progressing with increased duration of exposure to the source. Thus, elimination of the source, may not cure the disease, but will at least prevent the further deterioration of symptoms in affected patients and most importantly, will prevent the occurrence of disease in yet unaffected residents.

V.   Summary
Our study, a cross-sectional observational one, gives a vivid delineation of the clinical and radiographic features that may be seen in fluorosis. This will enable early diagnosis of this not so common condition, and help to differentiate it from other diseases which often mimic fluorosis in its early stages.

This study also emphasises that fluorosis is a cumulative exposure related disease, with symptoms deteriorating with increasing years of exposure, and that it cannot be cured, once symptoms have developed, even if the source is eliminated. However, early diagnosis of the disease in any region, may enable undertaking of measures for identification and elimination of sources, which can in turn prevent the further progression of symptoms in affected patients, and also save the unaffected people from developing the disease.

Our study group represents only a small proportion of fluorosis victims of Nasipur village. So it is recommended that a thorough epidemiological study with long term follow-up, be undertaken in Nasipur and adjacent villages to find out the extent of the disease and the exact magnitude of the problem.

Finally, from the devastating effects of fluorosis observed in Nasipur village, it may be suggested that any water source should not be open for drinking purpose until proper testing for fluoride as well as arsenic and other impurities have been done by some authentic representative.

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Clinical and radiological pictures

Figure 1: Dental Fluorosis

Figure 2: Dental Fluorosis

Figure 3: Skeletal fluorosis : Genu valgum(Left)

Figure 4 :Skeletal Fluorosis: Kypho-Scoliosis

Figure 5:Skeletal Fluorosis: Crippling patient

Figure 6:Skeletal Fluorosis: Osteosclerosis, calcification of sacro-tuberous and sacro-spinous ligament and osteophytes.
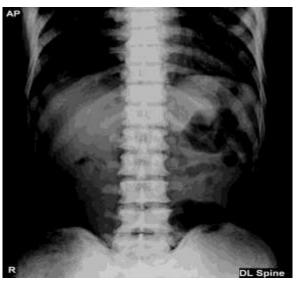
Figure.7: Skeletal Fluorosis: Osteosclerosis of vertebras and ribs

Figure 8 :Osteosclerosis, obliteration of marrow cavity, ossification of interosseous membrane of leg and osteophytes
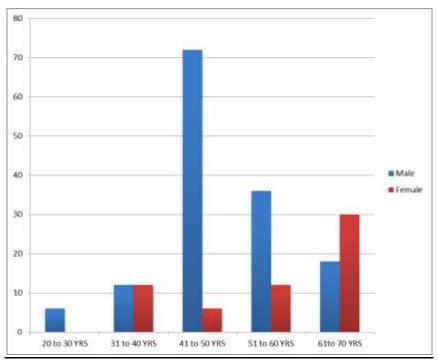
Figure 9: Skeletal fluorosis : Involvement of pelvic bones and femur: osteoscelorosis of epiphysis, metaphysis and diaphysis.. obliteration of medulla

Figure 10: Skeletal Fluorosis : Thickening of cortex, obliteration of medullary cavity of Radius and ulna; ossification of inter-osseous membrane of forearm

Table 1 : Proportion of patients belonging to different age groups in both sex

Age in Years	Sex		Total	Percentage
	Male	Female		
20 - 30	6	x	6	2.94
31 - 40	12	12	24	11.76
41 - 50	72	6	78	38.24
51 - 60	36	12	48	23.53
61 - 70	18	30	48	23.53

Proportion of Patients belonging to Different Age Groups in Both Sexes

Table 2: Distribution of patients according to their occupation

Occupation	No of Cases	Percentage
Student	6	3.7
Labourer	84	51.9
Cultivator	24	14.8
Grocer	18	11.1
Housewife	30	18.5

Table 3: Types of Clinical presentation according to different sex

SL. NO.	Complaints	No. of Cases		Total	Percentage
		Male	Female		
1	Back pain	138	60	198	97.05
2	Knee joint pain & leg pain	120	60	180	88.23
3	Difficulty to squat	96	54	150	73.52
4	Deformity of lower limbs	72	36	108	52.94
5	Painful & restricted forearm movement	90	54	144	69.23
6	Neurological symptoms	48	30	78	38.23
7	Crippling deformity of spine	12	18	30	14.70
8	Breathing difficulty	48	30	78	38.23
9	Dental fluorosis	138	54	192	94.11

Table 4: Abnormalities on regional clinical examination

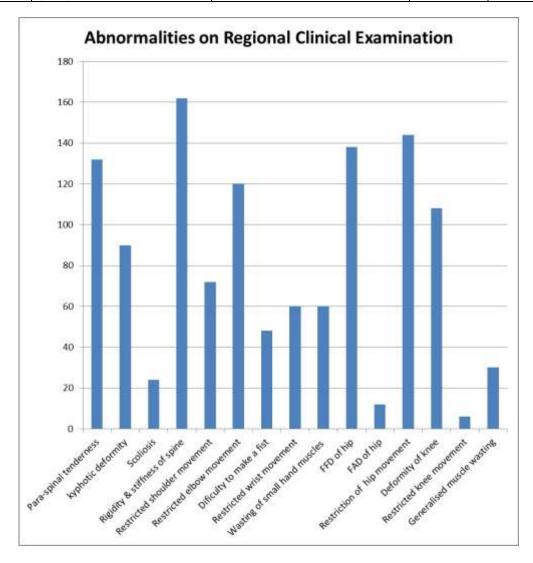| Sl. No	Region	Abnormality	No. of patient	Percentage
1	Spine	Para spinal tenderness	132	64.70
		Kyphosis	90	44.11
		Scoliosis	24	11.76
		Rigidity and stiffness of spine	162	79.41
2	Shoulder	Restriction of movement	72	35.29
3	Elbow	Restriction of movement (Pronation-Supination)	120	58.82
4	Wrist and hand	Difficulty to make a fist	48	23.52
		Restriction of movement	60	29.41
		Wasting of small muscles	48	23.52
5	Hip	Fixed flexion deformity(FFD) (10°-45°)	138	67.64
		Fixed adduction Deformity(FAD)	12	5.88
		Restriction of movement	144	70.58
6	Knee	Deformity(valgus-varus deformity)	108	52.94
		Restriction of movement	66	32.35
7	Generalised muscle wasting (Gross)		30	14.70

Table 5: Distribution of patients according to radiological findings

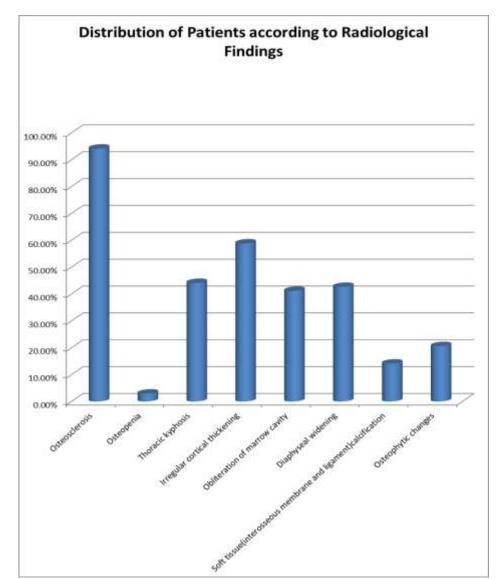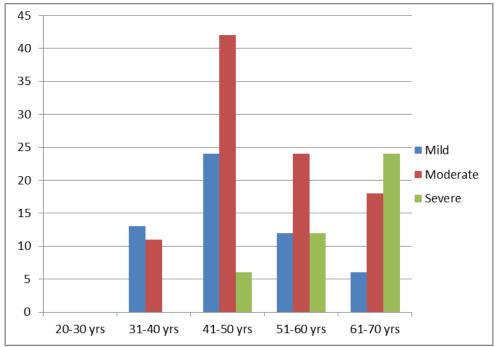| Sl. No.	Radiological findings	No of patients	Percentage
1	Osteosclerosis	192	94.12
2	Osteopenia	6	2.94
3	Thoracic kyphosis	90	44.12
4	Irregular cortical thickening	120	58.82
5	Obliteration of marrow cavity	84	41.18
6	Diaphyseal widening	87	42.65
7	Soft tissue (Interosseous membrane and ligament) calcification	29	14.22
8	Osteophytic changes	42	20.59

Table 6: Distribution of patients according to different radiological grading in different age groups

Age group (in years)	No of patients according to radiological grading		
	Mild	Moderate	Severe
20-30	0	0	0
31-40	13	11	0
41-50	24	42	6
51-60	12	24	12
61-70	6	18	24
Total number	55	95	42
Percentage	28.65%	49.48%	21.87%

Distribution of Patients according to Different Age Groups and Radiological Grading