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ENDEMIC GENU VALGUM AND OTHER BONE DEFORMITIES IN TWO VILLAGES OF MANDLA DISTRICT IN CENTRAL INDIA

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SUMMARY: An epidemiological investigation was undertaken in the villages of Tilaipani and Hirapur located in Mandla District of Central India to determine the cause and extent of a peculiar skeletal deformity characterised by knock knee (genu valgum) occurring mainly among children. In Tilaipani, 74.4% of children and adolescents below age 20 had dental mottling, and 51.2% were afflicted with genu valgum. In Hirapur the figures were 56.9% and 6.25%, respectively. Radiological examination revealed extensive osteosclerosis with coarse trabecular pattern. In a few cases juxta-articular bone resorption, periosteal reaction, and bony exostoses were also present. Biochemical parameters were generally within normal limits, but dietary surveys indicated that calcium and vitamin C intakes were significantly below recommended levels. In Tilaipani the fluoride content of the drinking water ranged from 9.22 to 10.83 ppm and in Hirapur from below 1 ppm to 13.5 ppm. The mean urinary fluoride concentration in the cases of skeletal deformities was 4.42 ppm.

The typical fluorotic dental mottling, the radiological findings, and the high fluoride content in the urine and drinking water all point to high fluoride, probably enhanced by deficient nutrition, as the cause of the genu valgum and other bone deformities.

Keywords: Bone deformities, Calcium deficiency, Dental fluorosis, Genu valgum, Hirapur, Malnutrition, Mandla District, Skeletal fluorosis, Tilaipani, Vitamin C deficiency, Water fluoride.

INTRODUCTION

In April 1995, we received a request from the Collector and the Chief Medical Officer of Mandla District for us to investigate the cause of a mysterious disease characterised by pain and deformity (genu valgum) in the lower limbs, mostly among children and adolescents below age 20 in two villages of the district. Thus this epidemiological investigation was undertaken with the objective of finding the cause of the knock-knee deformity.

MATERIALS AND METHODS

The investigation was carried out in two affected villages of Mandla District (Figure 1). The first village is Tilaipani, located about 12 km east of the district headquarter Mandla; the second village is Hirapur, which is about 50 km southwest of Mandla City. Tilaipani has a population of 542 in various age groups, and Hirapur has about 620. Evaluation of data comprised obtaining and recording medical histories, detailed clinical and radiological examinations, biochemical parameters of blood, dietary surveys, and fluoride levels in urine and in all drinking water sources.

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Biochemical investigations included serum alkaline phosphatase,[1] inorganic phosphorus,[2] and serum calcium.[2] Dietary surveys were conducted using the 24-hr recall method.[3] Fluoride levels were estimated using a fluoride ion selective electrode (Orion Research Inc.).

Figure 1. Map of District Mandla (not to scale)

S.No.	Index	S.No	Index	S.No	Index
1	Pacca road	4	District H.Q.	7	P.H.C.
2	District boundary	5	Tahsil H.Q.	8	Hospital
3	Study area	6	River	9	Railway line

OBSERVATIONS

Clinical: The percentage distributions of knock knee (genu valgum) and dental mottling in Tilaipani and in Hirapur are given in Tables 1 and 2, respectively. In Tilaipani the overall prevalence of genu valgum (Figure 2) below 20 years of age was 51.2% with more among males than females (2:1). The prevalence is significantly higher ($p<0.05$) in this age group than above age 20 ($Z = 4.99$). Dental mottling (Figure 3) was seen in 74.4% of children below age 20 and was significantly higher ($p<0.05$) among persons above age 20 ($Z = 5.81$).

In Hirapur, the incidence of dental mottling in individuals below age 20 was 56.9%, and no cases of dental mottling were seen above that age. The prevalence of genu valgum was only 6.25% among children below age 20. In Hirapur, however, this deformity was much more severe than in Tilaipani. Apart from genu valgum, severe anterior bowing and flattening of tibia and fibula were also evident (Figure 4).

Table 1. Age and sex distribution of knock knee (genu valgum) and dental mottling in the village of Tilaipani, Mandla District

Age	Sex	No. examined	No. with G.V.[a]	Prevalence %	Distribution of G.V.[a] %	No. with D.M.[b]	Prevalence %	Distribution of D.M.[b] %
0-5	M	7	0	0	0	0	0	0
	F	7	2	28.6	10.5	3	42.9	9.1
	T	14	2	14.3	3.8	3	21.4	3.9
6-10	M	20	16	80.0	48.5	19	95.0	43.2
	F	13	7	53.8	36.8	11	84.6	33.3
	T	33	23	69.7	44.2	30	90.9	39.0
11-20	M	25	13	52.0	39.4	21	84.0	47.7
	F	14	6	42.8	31.6	10	71.4	30.3
	T	39	19	48.7	36.5	31	79.5	40.3
21+	M	32	4	12.5	12.1	4	12.5	9.1
	F	34	4	11.7	21.1	9	26.5	27.3
	T	36	8	22.2	15.4	13	36.1	16.9
Total	M	84	33	39.3	100.0	44	52.4	100.0
	F	68	19	27.9	100.0	33	48.5	100.0
	T	152	52	34.2	99.9	77	50.7	100.1

[a]G.V. is genu valgum; [b]D.M. is dental mottling.

Table 2. Age and sex distribution of knock knee (genu valgum) and dental mottling in the village of Hirapur, Mandla District

Age	Sex	No. examined	No. with G.V.[a]	Prevalence %	% distribution of G.V.[a]	No.with D.M.[b]	Prevalence %	% distribution of D.M.[b]
0-5	M	15	0	0	0	2	13.3	3.4
	F	7	0	0	0	2	28.6	6.1
	T	22	0	0	0	4	18.2	4.4
6-10	M	60	6	10.0	85.7	49	81.7	84.5
	F	49	2	4.1	28.6	25	51.0	75.8
	T	109	8	7.3	57.1	74	67.9	81.3
11-20	M	13	0	0	0	7	53.8	12.1
	F	16	2	12.5	28.6	6	37.5	18.2
	T	29	2	6.9	14.3	13	44.8	14.3
21+	M	17	1	5.9	14.3	0	0	0
	F	21	3	14.3	42.9	0	0	0
	T	38	4	10.5	28.6	0	0	0
Total	M	105	7	6.7	100.0	58	52.2	100.0
	F	93	7	7.5	100.1	33	35.5	100.1
	T	198	14	7.1	100.0	91	46.0	100.0

[a]G.V. is genu valgum; [b]D.M. is dental mottling.

Figure 2. A group of children from Tilaipani with genu valgum

Figure 3. A 10-year-old child with severe dental mottling

Figure 4. An 8-year-old girl with anterior bowing and flattening of the tibia and fibula

Radiological: Radiographs of upper extremities, lower extremities, spine and hip bones were taken of a few affected children. Osteosclerosis with a coarse trabecular pattern was seen in most of the cases. A few X-rays showed juxta articular bone resorption of metacarpals and phalanges. Periosteal reaction with thickening of cortex was seen in most of the lower limb X-rays. In a few cases severe bowing of tibia and fibula (Figure 5) as well as radius and ulna (Figure 6) were seen. Multiple horizontal lines of trabeculae, which are known as growth-arrest lines suggesting repeated interruption of osteoblastic activities, were visible in long bones. Bony exostoses, which are considered to be a severe form of skeletal fluorosis,[4] were seen in two cases. No spinal or forearm calcification was found.

Figure 5. X-ray showing bowing of tibia and fibula with thickening of cortex and multiple growth arrest lines

Biochemical: A total of 86 blood samples from Tilaipani and 84 from Hirapur were collected and subjected to biochemical analysis. The mean value for serum alkaline phosphatase was 16.6 ± 6.4 KA Units, 3.4 ± 0.9 mg% for serum inorganic phosphorus, and 9.5 ± 0.5 mg/100 ml for serum calcium.

Dietary surveys: Dietary surveys were conducted in 22 households in Tilaipani and in 26 in Hirapur. The average intake of calories, calcium and iron in both villages was significantly ($p<0.05$) less than the recommended dietary allowances (RDAs).[5] Protein consumption was higher than the RDA in Hirapur but significantly lower in Tilaipani (Table 3). Vitamin C intake was also significantly lower in Tilaipani ($p<0.05$).

The average consumption of cereals and pulses was higher than the RDAs in both villages, whereas that of green leafy vegetables, sugar and jaggery, milk and milk products, and oil and fats was significantly ($p<0.05$) below the RDAs in Tilaipani. In Hirapur the intake of other vegetables, sugar and jaggery, milk and milk products and oil and fats was also below ($p<0.05$) the RDAs (Table 4).

Table 3. Average nutrient intake in gram/consumption unit/day in Tilaipani and Hirapur

Nutrients	RDA	Tilaipani intake (n = 22)	t-value d.f. = 21	Hirapur intake (n = 26)	t-value d.f. = 25
Energy	2425 Kcal	1889 ± 559.5	4.49*	2124 ± 528.8	2.89*
Protein	60 gm	48 ± 16.2	3.18*	60.47 ± 17.9	0.13
Fats	20 gm	20 ± 11.2	0.39	15.44 ± 6.4	3.66*
Calcium	400 mg	195 ± 65.7	14.59*	266.8 ± 147.9	4.59*
Iron	28 mg	9.9 ± 4.7	18.06*	14.05 ± 5.2	13.63*
Vitamin C	40 mg	27.1 ± 27.2	5.79*	34.06 ± 15.3	1.98

*Statistically significant, p<0.05.

Table 4. Average consumption in g/cu/day of foodstufs in Tilaipani and Hirapur

Food stuffs	RDA	Tilaipani intake in g (n = 22)	t-value d.f. = 21	Hirapur intake in g (n = 26)	t-value d.f. = 25
Cereals	460	506 ± 194	1.11	600 ± 177	4.03*
Pulses	40	71.2 ± 48.4	3.03*	62.0 ± 65.0	1.24
Green leafy vegetables	40	6.0 ± 16.4	9.72*	19.0 ± 52.8	2.03
Other vegetables	60	74.5 ± 90.5	0.75	114.3 ± 92.8	2.67*
Roots and tubers	50	35.7 ± 37.3	1.80	89.8 ± 61.0	3.33*
Sugar and jaggery	30	13.0 ± 9.7	6.95*	10.9 ± 10.8	19.20*
Milk and milk products	150	52.0 ± 66.1	8.22*	20.7 ± 34.3	9.01*
Oil and fats	40	15.8 ± 9.8	11.60*	11.0 ± 7.3	20.28*

*Statistically significant, p<0.05.

Fluoride content of water: Water samples from five different hand pumps in Tilaipani contained fluoride levels ranging from 9.22 ppm to 10.83 ppm. In Hirapur four hand pumps and three well water samples were analysed. The fluoride levels were below 1 ppm in all the sources except one hand pump where the fluoride content was 13.5 ppm.

Fluoride content of urine: To determine fluoride content, a total of 34 spot urine samples from Tilaipani and 10 from Hirapur were collected from individuals with skeletal deformity. The mean fluoride level was 4.42 ppm ± 2.87.

DISCUSSION

Endemic genu valgum associated with fluorosis was reported from the State of Andhra Pradesh in 1973.[6] Madhya Pradesh, on the other hand, is considered to be a low endemic area for fluorosis,[7] and Mandla District has not been previously reported as an endemic area for fluorosis. Recently, two children in

Nairobi were reported with genu valgum or knock knee due to exposure to high fluoride (10 ppm).[8] Sporadic cases of genu valgum associated with excess fluoride have been reported from a few more states of India,[4] though now there are 15 states declared affected because of fluorosis.[6] In Tilaipani, the significantly higher trend of the prevalence of genu valgum ($p<0.05$) among children below 20 years of age compared to individuals above age 20 could be because all five drinking water sources (hand pumps) in Tilaipani contain high fluoride (9.22 to 10.83 ppm). In Hirapur, though the prevalence is not so high as in Tilaipani, the trend is more towards children below 20 years. This could be because there is only one hand pump containing high fluoride (13.5 ppm). Since skeletal deformity is directly proportional to the level of fluoride intake,[9,10] probably for this reason the deformity in Hirapur children was much more severe than in the children of Tilaipani.

Figure 6. X-ray showing bowing of radius and ulna

The etiological factor in the 1973 report[6] was fluoride contamination of surface water by underground water caused by a change in soil strata, where top soil became underground soil and vice versa due to construction of a dam in a nearby area.[11] However, in the present study, fluorosis occurred due to consumption of water from deep bore wells, even though such wells were proposed as a method to protect individuals from endemic fluorosis.[12]

Poor nutrition also plays an important role in aggravating endemic fluorosis.[13] Though serum calcium values were normal in the children in our study, dietary calcium intake was much lower than the RDA ($p <0.05$) in both villages. Normal serum calcium values in skeletal fluorosis and genu valgum have also been reported previously.[14] Thus, low-calcium intake could also be another factor responsible for aggravating the problem of genu valgum.[15] Apart from calcium, dietary intakes of vitamin C, iron, green leafy vegetables, milk

and milk products, sugar and jaggery, and oil and fats in Tilaipani were much lower than the RDAs ($p < 0.05$). Likewise in Hirapur, the consumption of iron, milk and milk products, sugar and jaggery, and oil and fats was also much less than the RDAs ($p < 0.05$). Obviously, the possible role of these factors needs to be studied in greater depth.

Except for one child in Tilaipani, there were no obvious clinical signs of rickets. The high fluoride levels in the water and urine, together with the typical clinical and radiological findings, thus favour fluoride toxicity as the cause of the deformities. Earlier reports[11] have shown that fluorosis *per se* can cause genu valgum and rickets-like radiological features. An extensive survey is now indicated in this area to map out the fluorosis-affected locations in order to provide the people with safe drinking water.

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