

GENU-VALGUM (KNOCK-KNEE) SYNDROME IN FLUOROSIS-ENDEMIC RAJASTHAN AND ITS CURRENT STATUS IN INDIAShanti Lal Choubisa,^{a,*} Darshana Choubisa^b

Jaipur and Udaipur, India

ABSTRACT: During a recent observational survey of chronic fluoride (F) intoxication in school children in the F endemic villages of the Dungarpur district of Rajasthan, India, a case of genu-valgum (knock-knee) was detected in Katisor village. The drinking waters of this village have F in the range of 1.75 to 3.75 ppm. A male tribal subject, aged 54 years, was found to be afflicted with a genu-valgum (knock-knee) skeletal deformity, characterised, when standing, by a medial deviation of the thighs (upper legs) so that the knees touched and more widely separated ankles. He had moderate dental fluorosis and the health complaints of pain in the neck, back, and joints, abdominal pain, constipation, bloating, frequency of urination, excessive thirst, and extreme muscle weakness. The F content in the urine and blood samples was not evaluated. To the best of our knowledge, after examining the literature, this case is the first case of genu-valgum syndrome to be reported from Rajasthan (India) where hydrofluorosis is hyperendemic. In the present communication, the current status of genu-valgum in F or fluorosis endemic areas in India is also critically reviewed.

Keywords: Fluoride; Genu-valgum (knock-knee); Hydrofluorosis; Katisor; Rajasthan; India.

INTRODUCTION

It is well established that prolonged fluoride ion (F) exposure, either through drinking water with high F levels or industrial F pollution, induces a range of adverse or undesirable health effects in both man¹⁻³ and domestic animals.⁴⁻⁷ Dental mottling (dental fluorosis) and osteal changes (skeletal fluorosis) are the main visible pathognomic signs of chronic F intoxication. In subjects afflicted with advanced skeletal fluorosis, the most common bony deformities include crippling, kyphosis, scoliosis, flexion deformity of the knee joints, paraplegia, quadriplegia, and genu-varum (bow legs).⁸ However, in India, in certain fluorosis endemic states where F in drinking waters is relatively high (>3.0 ppm), another form of skeletal deformity, genu-valgum (knock-knee), has also been detected and reported.⁹⁻²²

In India, Rajasthan is one of the states where the drinking groundwater in the rural areas of 33 districts is highly contaminated with F.²³ In one of these districts, Dungarpur district, almost all the drinking water sources (hand-pumps and wells) are highly contaminated with F (up to 10.0 ppm).^{24,25} Many epidemiological studies on chronic F intoxication (hydrofluorosis) have been conducted on the tribal individuals²⁶⁻²⁹ and the domestic animals³⁰⁻³⁴ in several of the F endemic villages of this tribal district and diverse dental and skeletal deformities have been reported. However, not a single case of genu-valgum (knock-knee) skeletal deformity was identified in any

^aDr Shanti Lal Choubisa, Emeritus Professor (Biotechnology), Department of Advanced Science and Technology, National Institute of Medical Science and Research, NIMS University Rajasthan, Jaipur-303121, India; Former Associate Professor and Head (Zoology), Govt MG College, Udaipur-313001, Rajasthan, India, and Regional Editor of *Fluoride* for India, Bangladesh, Bhutan, Maldives, Myanmar, Nepal, and Sri Lanka; E-mail: choubisasl@yahoo.com; ^bDepartment of Prosthodontics, Pacific Dental College and Research Centre, Bhilo Ka Bedla, Pratap Pura, Udaipur-313001, Rajasthan, India; E-mail: dr.darshanachoubisa@gmail.com.

*For correspondence: Dr Shanti Lal Choubisa, Emeritus Professor (Biotechnology), Department of Advanced Science and Technology, National Institute of Medical Science and Research, NIMS University Rajasthan, Jaipur-30312, India; Telephone: +91 9460028724. E-mail: choubisasl@yahoo.com

of these studies or in the studies conducted in other districts of Rajasthan.³⁵ Nevertheless, during a recent survey of chronic F poisoning in school children in the F endemic villages of the Dungarpur district of southern Rajasthan, India, we detected this deformity (genu-valgum, knock-knee) in a tribal individual who was a permanent resident of Katisor village, located in the Aspur tehsil of the Dungarpur district of Rajasthan (Figure 1). In the present communication we present and discuss this case of a person with genu-valgum and critically review the current status of this skeletal deformity in other F endemic states in India.



Figure 1. Map of the Dungarpur district of Rajasthan (India) showing all five tehsils (blocks), Aspur, Bichhivara, Dungarpur, Sagwara, and Simalwara as well as the location of Katisor village in which a case genu-valgum (knock-knee) was detected. Map available from: <https://www.downtoearth.org.in/coverage/science-and-technology/font>

MATERIALS AND METHODS

Study area: The Dungarpur district is the third smallest district in Rajasthan, India, and is situated in the southern part of the State. It lies between latitude 23°20' and 24°01' N and longitude 73°22' and 74°23' E. It has five tehsils (blocks) namely Aspur, Bichhiwara, Dungarpur, Sagwara, and Simalwara (Figure 1). The principle population in this district is the Bhil tribe.

The village in which a case of genu-valgum detected: According to the 2011 census Katisor village is number 097851 and is located in the Aspur tehsil of Dungarpur district in Rajasthan (Figure 1). It is situated 16 km away from the sub-district headquarters, Aspur, and 41 km away from the district headquarters, Dungarpur. The total geographical area of village is 853.93 hectares and the total population is 3,338 (1,577 males and 1,761 females). There are about 771 houses in the village. The occupations of inhabitants are mainly agriculture and hard labour. Economically the residents are very poor.

Nutritional status: The nutritional status of the tribal individuals in Katisor village is very poor. In general, their diet comprises maize, wheat, rice, onion, and garlic, with or without pulses and vegetables. Other food stuffs like milk, curd, ghee, vegetable oil, and fruit are very rare in their diets. Many residents smoke and have a habitual daily intake of tea and locally produced wine.³⁶

Status of F in the drinking water: In the villages of all five tehsils of the Dungarpur district, the drinking water sources are mainly hand-pumps and wells with a F range of 0.1 to 10.0 ppm.²⁵ The highest drinking water F concentration (10.0 ppm) was reported in the villages of Durgapur tehsil, followed by Sagwara (6.7 ppm), Aspur (4.55 ppm), Simalwara (3.25 ppm), and Bichhiwara (3.25 ppm). In the Katisor village, the drinking waters contained F in the range of 1.75 to 3.75 ppm.²⁶

In the present report, only the observational findings are presented and no radiological investigations or biochemical measurements were made, e.g., the F concentrations in the urine and blood and the blood vitamin D level.

OBSERVATIONS AND DISCUSSION

During a recent survey of osteo-dental fluorosis in school children in Katisor village of Dungarpur district of Rajasthan, we detected a male tribal subject, aged 54 years, with a genu-valgum (knock-knee) deformity (Figure 2). When he was standing upright, his thighs (upper legs) showed a medial deviation so that his knees touched each other while his ankles were spread further apart (Figures 4 and 5). He was a permanent resident of Katisor village having lived there since childhood and having never stayed outside of the village for any lengthy period of time. On examination of his teeth, he was found to have a moderate degree of dental fluorosis. Physically, he was very weak and thin and had the complaints of pain in the neck, back, and joints, abdominal pain, constipation, bloating, frequency of urination (polyuria), excessive thirst (polydipsia), and extreme muscle weakness. From his health complaints and the presence of dental fluorosis, a diagnosis was made of chronic F intoxication.¹⁻³



Figure 2. A case of skeletal fluorosis, with a genu-valgum (knock-knee) deformity, from Katisor village of the Dungarpur district of Rajasthan, India.



Figure 3. A case of skeletal fluorosis, with a genu-varum (bow legs) deformity, from Katisor village of the Dungarpur district of Rajasthan, India.



Figure 4. Appearance of the legs (anterior view) in the subject with genu-valgum (knock-knee). In the standing position. The thighs (upper legs) show a medial deviation so that the knees touch each other and the ankles are spread further apart.



Figure 5. Appearance of legs (posterior view) the subject with genu-valgum (knock-knee). In the standing position. The thighs (upper legs) show a medial deviation so that the knees touch each other and the ankles are spread further apart.

In 1973, cases of genu-valgum syndrome were detected and reported in India for the first time from the villages of the Nalgonda district of Andhra Pradesh⁹ where the drinking waters contained relatively high levels of F (up to 6.0 ppm). Subsequently, similar cases were also detected in the villages of the Prukasam and Guntur districts of Andhra Pradesh where the drinking water F range was 1.5–10.8 ppm.^{10,11} At a later date, cases of genu-valgum, along with dental fluorosis, were also detected and reported from F endemic villages in the states of Bihar,¹² Chhattisgarh,¹³⁻¹⁶ Karnataka,^{11,17-20} Madhya Pradesh,²¹⁻²² and Tamil Nadu¹¹ (Table). However, none of these studies revealed the exact cause for the development of the genu-valgum (knock-knee) deformity in the F endemic areas or the mechanism involved. It is still not clear why the genu-valgum deformity is rare in its occurrence in the fluorosed population compared to the prevalence of the genu-varum syndrome (Figure 3).

Table. Drinking water (DW) fluoride (F) levels (ppm) and prevalence (%) or number (No) of cases of genu-valgum (GV) detected and reported so far from fluorosis endemic villages of various states in India (Ref=reference, NS=not specified)

State	District/Taluka	Village or No of villages	F in DW (ppm)	Sample size	GV prevalence (%) or No of cases	Ref
Andhra Pradesh	Nalgonda	3	3.5–6.0	NS	24 cases	9
Andhra Pradesh	Nalgonda+Prukasam+Guntur	76	>3.0	>15,250	3.4–3.8%	10
Andhra Pradesh	Nalgonda+Prukasam+Guntur	110	1.5–10.8	NS	9.0–17.1%	11
Bihar	Nawada	Kachariadih	3.5–14.5	240	13.5%	12
Chhattisgarh	Durg	Gureda	0.2–7.8	805	6.3%	13
Chhattisgarh	Konker	Domarpali	0.5–2.5	1,359	0.7%	14
Chhattisgarh	Ambikapur+Balod+Balrampur+Bastar+Kankar+Korba+Surajpur			1,414	0.7%	15
Chhattisgarh	Ambikapur	NS	0.4–6.8			
Chhattisgarh	Balod	NS	0.5–2.2			
Chhattisgarh	Balrampur	NS	0.2–4.0			
Chhattisgarh	Bastar	NS	0.1–7.3			
Chhattisgarh	Kankar	NS	0.5–2.8			
Chhattisgarh	Korba	NS	0.1–3.5			
Chhattisgarh	Surajpur	NS	0.2–9.0			
Chhattisgarh	Korba	Phulsar	>2.5	1,087	1.0%	16
Karnataka	Mallapuram	26	0.6–2.2	NS	A few cases	11
Karnataka	Gulbarga	Kheru, Naga, Thanda,	0.6–13.4	46	1 case	17
Karnataka	Chickballapur	Kaiwara	1.0–2.68	416	11.4%	18
Karnataka	Chickballapur	Kaiwara, Hobli	>1.5	1,544	8.4%	19
Karnataka	Koppal	15	–	1,722	11.2%	20
Madhya Pradesh	Mandala	Tilapani	9.22–10.83	542	51.1%	21
Madhya Pradesh	Mandala	Hirapur	<1.0–13.5	620	6.25%	22
Tamil Nadu	Pollachi	14	0.5–4.0	NS	0.2–1.6%	11

In India, 23 of the 36 states and union territories have an elevated F content in their drinking waters. However, the genu-valgum deformity is endemic in only in a few states (Table). Earlier, it was postulated that genu-valgum was restricted to those F endemic areas where the consumption of jowar (sorghum) as a food was prevalent.^{1,9-12} This proposition, however, is not supported by studies from several F endemic states in India, e.g., Gujarat, Maharashtra, western Rajasthan, Punjab, and Haryana, etc., where the staple diet of the people is jowar-bajra (sorghum-pearl

millet) but in which, so far, genu-valgum has not been found and reported. The way in which F causes these genu-valgum and genu-varum deformities is still mysterious and uncertain. In order to know and understand the possible causes or mechanisms involved in the development of these two different and opposite skeletal deformities in the individuals of the same community, inhabiting the same F or fluorosis endemic area, more epidemiological studies in different geographical provinces are highly suggested. The significance of this case of genu-valgum is that it reports genu-valgum (knock-knee) for the first time in Rajasthan where hydrofluorosis is hyperendemic.

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