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Clinical Profile and Factors Associated With Syndromic Clubfoot: A Hospital-Based Comparative Study From Northeast India

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Abstract

Introduction: About one in five children with clubfoot has an associated syndrome or congenital anomaly, yet data on syndromic clubfoot from Northeast India are scarce. We estimated the share of syndromic clubfoot at a tertiary centre in Assam, described its clinical profile, and identified factors that distinguish it from idiopathic clubfoot.

Methodology: This hospital-based comparative study (November 2024 to May 2026) enrolled all 73 children with syndromic clubfoot, 73 children with idiopathic clubfoot randomly selected from 283 seen in the same period, and 73 children without clubfoot attending the same orthopaedic outpatient department for trauma. Parents were interviewed with a structured schedule, and socioeconomic class was assigned with the modified Kuppuswamy scale (2026 update). Groups were compared with Pearson's chi-square test (exact tests for sparse tables), the Kruskal-Wallis H test and the Mann-Whitney U test. Binary logistic regression identified factors independently associated with syndromic versus idiopathic clubfoot.

Results: Of 360 children with clubfoot, 73 (20.3%; 95% confidence interval (CI) 16.5-24.7) had syndromic clubfoot. Arthrogryposis multiplex congenita was the commonest diagnosis (22/73, 30.1%), followed by associated limb anomalies (10/73, 13.7%) and Down syndrome (9/73, 12.3%). Bilateral involvement was similar in syndromic and idiopathic clubfoot (51/73, 69.9% vs. 44/73, 60.3%; $\chi^2 = 2.85$, $P = 0.241$), but deformity was more severe (median worst-foot Pirani score 3.5 vs. 2.0; U = 3682.5, $P < 0.001$). Previous foetal loss (36/73, 49.3% vs. 11/73, 15.1% vs. 1/73, 1.4%; $\chi^2 = 52.03$, $P < 0.001$), abnormal foetal presentation (27/73, 37.0% vs. 11/73, 15.1% vs. 10/73, 13.7%; $\chi^2 = 14.57$, $P < 0.001$) and neonatal intensive care admission (27/73, 37.0% vs. 15/73, 20.5% vs. 9/73, 12.3%; $\chi^2 = 12.88$, $P = 0.002$) were most frequent in the syndromic group. On multivariable analysis, syndromic clubfoot was independently associated with previous foetal loss (adjusted odds ratio (AOR) 11.45; 95% CI 4.08-32.15), abnormal foetal presentation (AOR 5.87; 95% CI 1.96-17.55), a previous preterm or post-term delivery (AOR 11.39; 95% CI 1.94-66.93) and maternal education up to primary level (AOR 4.11; 95% CI 1.10-15.40). The model discriminated well (area under the curve 0.876).

Conclusions: One in five children with clubfoot at this centre had syndromic clubfoot, with more severe deformity than idiopathic clubfoot. A history of foetal loss, abnormal presentation or an earlier preterm or post-term birth should prompt a full search for associated anomalies and referral for genetic evaluation.

Categories: Orthopedics
Keywords: arthrogryposis multiplex congenita, clubfoot, congenital talipes equinovarus, idiopathic clubfoot, northeast india, pirani score, risk factors, syndromic clubfoot

Introduction

Congenital talipes equinovarus (clubfoot) is one of the commonest structural birth defects. A recent global meta-analysis estimated its birth prevalence at 1.18 per 1,000 births, with the highest pooled estimate, 1.80 per 1,000, in the World Health Organization South-East Asia Region [1]. In India, talipes is among the two most frequently reported congenital anomalies [2]. Hospital-based series from Northeast India likewise identify the musculoskeletal system as the system most often affected by gross congenital malformations [3,4].

About 80% of clubfeet are isolated (idiopathic). The remainder occur with neuromuscular disorders, chromosomal abnormalities or recognised syndromes and are termed non-idiopathic or syndromic clubfoot [5,6]. The distinction matters clinically. Syndromic feet are stiffer, need more casts, relapse more often and more often need surgery after Ponseti treatment [7-11], whereas relapse in idiopathic clubfoot ranges from 1.9% to 45% [12]. Arthrogryposis multiplex congenita (AMC), defined by congenital contractures in two or

more body areas, is the commonest syndromic association and itself covers a heterogeneous group of conditions [6,13].

Most epidemiological work has examined idiopathic clubfoot. Family history and maternal smoking are the most consistent risk factors, and markers of foetal constraint such as oligohydramnios and breech presentation have also been implicated [14-18]. Far less is known about the parental, obstetric and perinatal background of children with syndromic clubfoot, and almost nothing is known about this in Northeast India, a region with distinct ethnic composition, tribal populations and health-care access. The Ponseti method is now the standard first-line treatment [19,20] and, in India, is delivered largely through public clubfoot clinics, so knowing which children are likely to have an associated condition would help clinicians plan examination, counselling and follow-up.

This study therefore aimed to (1) estimate the proportion of syndromic clubfoot among children with clubfoot attending a tertiary centre in Assam; (2) describe the spectrum of associated diagnoses and the laterality and severity of syndromic clubfoot compared with idiopathic clubfoot; and (3) identify sociodemographic, maternal and perinatal factors independently associated with syndromic clubfoot, using idiopathic clubfoot as the primary comparison and children without clubfoot as a reference group.

Materials And Methods

Study design, setting and period

This hospital-based comparative study was conducted at the Clubfoot Clinic of the Department of Orthopaedics of a tertiary care teaching hospital in Guwahati, Assam, from November 2024 to May 2026. The hospital is the main public referral centre for clubfoot in Assam and receives children from neighbouring northeastern states. Reporting follows the Strengthening the Reporting of Observational Studies in Epidemiology (STROBE) guidance [21].

Participants and selection

During the study period, 15,799 children aged under 18 years attended the orthopaedic outpatient department, of whom 360 had clubfoot. Three groups of 73 (33.3%) children each were enrolled.

Syndromic Clubfoot

Every child with clubfoot and at least one associated syndrome, congenital anomaly or neuromuscular disorder identified during the study period (n = 73) was included. Associated conditions were identified by clinical examination by the treating orthopaedic surgeon, supplemented by paediatric and genetic consultation and investigations where available.

Idiopathic Clubfoot

Children with isolated clubfoot and no other congenital anomaly on examination. Of the 287 children without an identified associated syndrome, four could not be enrolled because their parents or guardians were unavailable for interview. Thus, 283 eligible children constituted the sampling frame, from which 73 were selected by simple random sampling.

Non-clubfoot Reference Group

Seventy-three children without clubfoot or any other congenital anomaly attending the same outpatient department for trauma were selected at random on clinic days.

Children with postural (flexible) foot deformity and those whose parent or guardian was unavailable for interview were excluded. No formal sample size was calculated; the size was fixed by the number of syndromic cases available, and equal-sized comparison groups were drawn.

Data collection and variables

A pretested, structured interview schedule was administered to the mother or to the primary caregiver when the mother was unavailable and was cross-checked against antenatal cards and discharge summaries. Variables were grouped as follows.

Child and Family

Age, sex, district, type of family, consanguineous marriage, family history of clubfoot, and parental age at conception, religion, ethnicity (tribal or non-tribal), education and occupation.

Socioeconomic Class

The modified Kuppuswamy scale, which combines the education and occupation of the head of the family with total monthly family income, using the income thresholds updated for 2026 with the Consumer Price Index for Industrial Workers [22]. Classes were grouped as upper/upper middle, lower middle, and upper lower/lower.

Perinatal

Season of birth, gestation at birth, place of birth, birth order, birth weight, cry at birth, birth trauma or asphyxia, foetal presentation, mode of delivery and neonatal intensive care unit (NICU) admission.

Maternal Obstetric and Antenatal History

Gravidity, parity, previous foetal loss (miscarriage or stillbirth), previous preterm or post-term delivery, other previous or current obstetric events, infection, hyperemesis, oligohydramnios, anaemia, hypertensive disorder, gestational diabetes and twin pregnancy in the index pregnancy, tobacco (smoked or smokeless, including areca nut) and alcohol use, passive smoke exposure, and completeness of iron-folic acid supplementation.

Clinical

Laterality and the Pirani score of each affected foot (0-6, from six components scored 0, 0.5 or 1) [23]. The worst-foot score was used per child and classified as mild (0-2), moderate (2.5-4) or severe (4.5-6). Pirani scores were recorded at the time of study assessment. Pre-treatment Pirani scores were not consistently available for children who had initiated treatment before enrolment; therefore, baseline severity could not be uniformly assessed across all participants.

Use of medicines other than routine antenatal supplements was recorded only for the two clubfoot groups and is therefore compared between them alone.

Statistical analysis

Categorical variables are presented as *n* (%) and continuous variables as median (interquartile range, IQR), because none was normally distributed (Shapiro-Wilk $P \leq 0.003$ in every group). Sparse categories were merged before testing (birth order, gravidity and parity ⩾ 3; birth weight as reported). Each variable was compared across the three groups and, as the pre-specified primary contrast, between the syndromic and idiopathic groups with Pearson's chi-square (χ^2) test; χ^2 and degrees of freedom (df) are reported for every comparison. Where more than 20% of expected cell counts were below five, the exact *P*-value is reported alongside the χ^2 statistic (Fisher's exact test for 2 × 2 tables; Fisher-Freeman-Halton test, Monte Carlo estimate from 20,000 resamples, for larger tables). Age was compared with the Kruskal-Wallis H test and the Mann-Whitney U test, and Pirani scores with the Mann-Whitney U test per child and per affected foot.

Factors associated with syndromic versus idiopathic clubfoot were examined with binary logistic regression. Crude odds ratios (OR) were estimated for candidate pre-natal and background factors. Factors with crude $P < 0.05$ that preceded the index birth entered the multivariable model; infection during pregnancy was added a priori as a plausible teratogenic exposure. Place of birth, mode of delivery and NICU admission were not entered because they may follow from an antenatally recognised anomaly. The eight resulting predictors gave about nine events per variable [24]. Results are expressed as adjusted odds ratios (AOR) with 95% confidence intervals (CIs) and Wald χ^2 values. Collinearity was checked with variance inflation factors (VIFs). Model fit was assessed with the likelihood-ratio χ^2 test, Nagelkerke R^2 and the Hosmer-Lemeshow test, and discrimination with the area under the receiver operating characteristic curve (AUC), with a bootstrap 95% CI from 2,000 resamples. Two-sided $P < 0.05$ was considered significant. Analyses were performed using Python version 3.13, SciPy version 1.18, statsmodels version 0.15 and scikit-learn version 1.9.

Ethical considerations

The study was approved by the Institutional Ethics Committee of Gauhati Medical College and Hospital. Written informed consent was obtained from a parent or legal guardian of every child. The study followed the Declaration of Helsinki, and participation did not affect clinical care.

Results

Of the 15,799 children who attended the orthopaedic outpatient department during the study period, 360 (2.28%) had clubfoot. Syndromic clubfoot was present in 73 children: 20.3% (73/360, 95% CI 16.5-24.7) of all children with clubfoot and 0.46% (73/15,799, 95% CI 0.37-0.58), or 4.62 per 1,000, of all outpatient attendees. A total of 219 (100%) children (73, 33.3% per group) were analysed.

Sociodemographic and family characteristics

Table 1 compares the three groups. Sex distribution did not differ (males: 47/73, 64.4%; 42/73, 57.5%; 44/73, 60.3%; $\chi^2 = 0.73$, df = 2, $P = 0.695$). Children with syndromic and idiopathic clubfoot were of similar age (median 6 vs. 8 months; $U = 2202.5$, $P = 0.070$), whereas the non-clubfoot children were older (median 48 months; $H = 51.40$, df = 2, $P < 0.001$).

Characteristic	Syndromic (*n* = 73), *n* (%)	Idiopathic (*n* = 73), *n* (%)	Non-clubfoot (*n* = 73), *n* (%)	Three groups: χ^2 (df), *P*	Syndromic vs. idiopathic: χ^2 (df), *P*
Age at assessment (months), median (IQR)	6 (2-14)	8 (4-18)	48 (12-72)	*H* = 51.40 (2), <0.001§	*U* = 2202.5, 0.070
Sex					
Male	47 (64.4)	42 (57.5)	44 (60.3)	0.73 (2), 0.695	0.72 (1), 0.396
Female	26 (35.6)	31 (42.5)	29 (39.7)		
Maternal age at conception					
<20 years	15 (20.5)	9 (12.3)	0 (0.0)	16.88 (4), 0.002	2.14 (2), 0.427†
20-35 years	55 (75.3)	59 (80.8)	66 (90.4)		
>35 years	3 (4.1)	5 (6.8)	7 (9.6)		
Paternal age at conception					
≤35 years	54 (74.0)	59 (80.8)	39 (53.4)	13.98 (2), <0.001	0.98 (1), 0.322
>35 years	19 (26.0)	14 (19.2)	34 (46.6)		
Religion					
Hindu	32 (43.8)	25 (34.2)	35 (47.9)	4.27 (4), 0.431†	2.59 (2), 0.236†
Muslim	40 (54.8)	48 (65.8)	37 (50.7)		
Christian	1 (1.4)	0 (0.0)	1 (1.4)		
Maternal ethnicity: tribal	11 (15.1)	4 (5.5)	1 (1.4)	10.65 (2), 0.005	3.64 (1), 0.056
Paternal ethnicity: tribal	14 (19.2)	4 (5.5)	1 (1.4)	16.02 (2), <0.001	6.34 (1), 0.012
Maternal education					
Illiterate/primary	20 (27.4)	6 (8.2)	10 (13.7)	73.64 (4), <0.001	11.38 (2), 0.002†
Middle/high school	48 (65.8)	65 (89.0)	27 (37.0)		
Intermediate or above	5 (6.8)	2 (2.7)	36 (49.3)		
Paternal education					
Illiterate/primary	22 (30.1)	18 (24.7)	9 (12.3)	93.45 (4), <0.001	0.74 (2), 0.690
Middle/high school	43 (58.9)	48 (65.8)	10 (13.7)		
Intermediate or above	8 (11.0)	7 (9.6)	54 (74.0)		
Mother employed outside the home	0 (0.0)	5 (6.8)	5 (6.8)	5.24 (2), 0.083†	5.18 (1), 0.058†
Joint family	52 (71.2)	55 (75.3)	23 (31.5)	35.47 (2), <0.001	0.31 (1), 0.575
Socioeconomic class*					
Upper/upper middle	9 (12.3)	3 (4.1)	57 (78.1)	125.14 (4), <0.001	11.11 (2), 0.004
Lower middle	42 (57.5)	29 (39.7)	14 (19.2)		

Upper lower/lower	22 (30.1)	41 (56.2)	2 (2.7)		
Consanguineous marriage	5 (6.8)	6 (8.2)	1 (1.4)	3.70 (2), 0.249†	0.10 (1), 0.754
Family history of clubfoot	6 (8.2)	9 (12.3)	0 (0.0)	9.02 (2), 0.011	0.67 (1), 0.414

TABLE 1: Sociodemographic and family characteristics of the study groups.

*Modified Kuppuswamy scale, 2026 income update [22].

†Exact *P*-value (Fisher's exact or Fisher-Freeman-Halton test) reported with the Pearson χ^2 statistic.

§Kruskal-Wallis H test.

¶Mann-Whitney U test.

IQR, interquartile range

Compared with the idiopathic group, the syndromic group had more fathers of tribal ethnicity (14/73, 19.2% vs. 4/73, 5.5%; $\chi^2 = 6.34$, df = 1, $P = 0.012$) and more mothers educated only to primary level or below (20/73, 27.4% vs. 6/73, 8.2%; $\chi^2 = 11.38$, df = 2, exact $P = 0.002$). Socioeconomic class differed in the opposite direction: upper-lower or lower class was recorded for 22/73 (30.1%) syndromic and 41/73 (56.2%) idiopathic families ($\chi^2 = 11.11$, df = 2, $P = 0.004$). Consanguinity (5/73, 6.8% vs. 6/73, 8.2%; $\chi^2 = 0.10$, df = 1, $P = 0.754$) and a family history of clubfoot (6/73, 8.2% vs. 9/73, 12.3%; $\chi^2 = 0.67$, df = 1, $P = 0.414$) did not differ between the two clubfoot groups; neither was reported in more than one non-clubfoot family.

Perinatal characteristics

Perinatal characteristics

Perinatal characteristics are shown in Table 2. Abnormal foetal presentation was reported in 27/73 (37.0%) syndromic children, compared with 11/73 (15.1%) idiopathic and 10/73 (13.7%) non-clubfoot children ($\chi^2 = 14.57$, df = 2, $P < 0.001$); breech presentation accounted for 24 of the 27 syndromic cases, with one shoulder presentation and two unspecified presentations. NICU admission was also more common in the syndromic group (27/73, 37.0% vs. 15/73, 20.5%; $\chi^2 = 4.81$, df = 1, $P = 0.028$). Caesarean delivery differed across the three groups (32/73, 43.8% vs. 22/73, 30.1% vs. 12/73, 16.4%; $\chi^2 = 13.01$, df = 2, $P = 0.001$), but not significantly between the two clubfoot groups ($P = 0.086$). No idiopathic child was born in autumn, compared with 13/73 (17.8%) in each of the other groups (syndromic vs. idiopathic: $\chi^2 = 14.67$, df = 4, $P = 0.005$). Gestation at birth, birth order, birth weight, cry at birth and birth trauma did not differ.

Characteristic	Syndromic (*n* = 73), *n* (%)	Idiopathic (*n* = 73), *n* (%)	Non-clubfoot (*n* = 73), *n* (%)	Three groups: χ^2 (df), *P*	Syndromic vs. idiopathic: χ^2 (df), *P*
Season of birth					
Monsoon	27 (37.0)	32 (43.8)	14 (19.2)	23.84 (8), 0.002	14.67 (4), 0.005
Autumn	13 (17.8)	0 (0.0)	13 (17.8)		
Winter	17 (23.3)	20 (27.4)	20 (27.4)		
Spring	10 (13.7)	15 (20.5)	15 (20.5)		
Summer	6 (8.2)	6 (8.2)	11 (15.1)		
Gestation at birth					
Term	58 (79.5)	57 (78.1)	53 (72.6)	1.37 (4), 0.850	0.31 (2), 0.857
Preterm	8 (11.0)	10 (13.7)	11 (15.1)		
Post-term	7 (9.6)	6 (8.2)	9 (12.3)		
Place of birth					
Government hospital	58 (79.5)	63 (86.3)	69 (94.5)	13.87 (4), 0.006†	5.21 (2), 0.076†
Private hospital	12 (16.4)	4 (5.5)	1 (1.4)		
Home	3 (4.1)	6 (8.2)	3 (4.1)		
Birth order					
1	35 (47.9)	30 (41.1)	40 (54.8)	8.19 (4), 0.085	2.84 (2), 0.242
2	28 (38.4)	25 (34.2)	27 (37.0)		
≥3	10 (13.7)	18 (24.7)	6 (8.2)		
Birth weight					
<1.5 kg	5 (6.8)	6 (8.2)	0 (0.0)	10.84 (6), 0.089†	0.66 (3), 0.923†
1.5 to <2.5 kg	19 (26.0)	15 (20.5)	26 (35.6)		
2.5-4 kg	45 (61.6)	48 (65.8)	46 (63.0)		
>4 kg	4 (5.5)	4 (5.5)	1 (1.4)		
Did not cry immediately at birth	8 (11.0)	9 (12.3)	9 (12.3)	0.09 (2), 0.957	0.07 (1), 0.796
Birth trauma or asphyxia	20 (27.4)	15 (20.5)	15 (20.5)	1.30 (2), 0.523	0.94 (1), 0.332
Abnormal foetal presentation	27 (37.0)	11 (15.1)	10 (13.7)	14.57 (2), <0.001	9.11 (1), 0.003
Caesarean delivery	32 (43.8)	22 (30.1)	12 (16.4)	13.01 (2), 0.001	2.94 (1), 0.086
NICU admission	27 (37.0)	15 (20.5)	9 (12.3)	12.88 (2), 0.002	4.81 (1), 0.028

TABLE 2: Perinatal characteristics of the index child.

†Exact *P*-value reported with the Pearson χ^2 statistic.

NICU, neonatal intensive care unit

Maternal obstetric and antenatal history

Table *3* summarises the maternal history. Previous foetal loss showed the steepest gradient: 36/73 (49.3%) mothers of syndromic children, 11/73 (15.1%) of idiopathic children and 1/73 (1.4%) of non-clubfoot

children (χ^2 = 52.03, df = 2, P < 0.001; syndromic vs. idiopathic χ^2 = 19.61, df = 1, P < 0.001). A previous preterm or post-term delivery was also commoner among syndromic than idiopathic mothers (10/73, 13.7% vs. 2/73, 2.7%; χ^2 = 5.81, df = 1, P = 0.016). Infection during pregnancy was reported by 15/73 (20.5%), 9/73 (12.3%) and 1/73 (1.4%) mothers, respectively (χ^2 = 13.37, df = 2, P = 0.001), without a significant difference between the two clubfoot groups (P = 0.180). Hyperemesis, oligohydramnios and anaemia were, by contrast, most frequent in the non-clubfoot group (all P < 0.001) and similar in the two clubfoot groups. No mother reported cigarette or beedi smoking; smokeless tobacco or areca nut use was common and evenly spread (36/73, 49.3%; 35/73, 47.9%; 30/73, 41.1%; χ^2 = 1.14, df = 2, P = 0.566). Use of medicines beyond routine supplements, recorded for the clubfoot groups only, was less frequent in syndromic than idiopathic mothers (9/73, 12.3% vs. 19/73, 26.0%; χ^2 = 4.42, df = 1, P = 0.036).

Characteristic	Syndromic (*n* = 73), *n* (%)	Idiopathic (*n* = 73), *n* (%)	Non-clubfoot (*n* = 73), *n* (%)	Three groups: χ^2 (df), *P*	Syndromic vs. idiopathic: χ^2 (df), *P*
Gravidity					
1	26 (35.6)	22 (30.1)	30 (41.1)	8.56 (4), 0.073	2.14 (2), 0.343
2	30 (41.1)	26 (35.6)	33 (45.2)		
≥3	17 (23.3)	25 (34.2)	10 (13.7)		
Parity					
1	34 (46.6)	26 (35.6)	31 (42.5)	6.02 (4), 0.197	2.04 (2), 0.361
2	26 (35.6)	29 (39.7)	34 (46.6)		
≥3	13 (17.8)	18 (24.7)	8 (11.0)		
Previous foetal loss	36 (49.3)	11 (15.1)	1 (1.4)	52.03 (2), <0.001	19.61 (1), <0.001
Previous preterm or post-term delivery	10 (13.7)	2 (2.7)	13 (17.8)	8.76 (2), 0.013	5.81 (1), 0.016
Other previous obstetric event	10 (13.7)	7 (9.6)	6 (8.2)	1.26 (2), 0.532	0.60 (1), 0.439
Obstetric event in index pregnancy	14 (19.2)	8 (11.0)	8 (11.0)	2.78 (2), 0.249	1.93 (1), 0.165
Infection during pregnancy	15 (20.5)	9 (12.3)	1 (1.4)	13.37 (2), 0.001	1.80 (1), 0.180
Hyperemesis gravidarum	15 (20.5)	16 (21.9)	43 (58.9)	30.90 (2), <0.001	0.04 (1), 0.840
Oligohydramnios	12 (16.4)	9 (12.3)	31 (42.5)	21.54 (2), <0.001	0.50 (1), 0.479
Anaemia	27 (37.0)	18 (24.7)	45 (61.6)	21.39 (2), <0.001	2.60 (1), 0.107
Hypertensive disorder of pregnancy	9 (12.3)	3 (4.1)	0 (0.0)	11.11 (2), 0.004†	3.27 (1), 0.071
Gestational diabetes	5 (6.8)	1 (1.4)	1 (1.4)	4.72 (2), 0.129†	2.78 (1), 0.209†
Twin pregnancy	1 (1.4)	2 (2.7)	2 (2.7)	0.41 (2), 1.000†	0.34 (1), 1.000†
Passive smoke exposure	10 (13.7)	15 (20.5)	8 (11.0)	2.78 (2), 0.249	1.21 (1), 0.272
Smokeless tobacco or areca nut use	36 (49.3)	35 (47.9)	30 (41.1)	1.14 (2), 0.566	0.03 (1), 0.868
Alcohol use	1 (1.4)	3 (4.1)	1 (1.4)	1.64 (2), 0.624†	1.03 (1), 0.620†
Incomplete iron-folic acid supplementation	8 (11.0)	4 (5.5)	1 (1.4)	6.05 (2), 0.055†	1.45 (1), 0.228
Medicines other than routine supplements‡	9 (12.3)	19 (26.0)	Not recorded	-	4.42 (1), 0.036

TABLE 3: Maternal obstetric and antenatal history.

†Exact *P*-value reported with the Pearson χ^2 statistic.

‡Recorded for the two clubfoot groups only.

Clinical profile of clubfoot

Table 4 compares the clubfoot itself. Bilateral involvement was frequent in both groups and did not differ

significantly (51/73, 69.9% vs. 44/73, 60.3%; $\chi^2 = 2.85$, df = 2, $P = 0.241$). Deformity was more severe in syndromic clubfoot: the median worst-foot Pirani score was 3.5 (IQR 2.5-5.0) against 2.0 (IQR 0.5-3.75) in idiopathic clubfoot ($U = 3682.5$, $P < 0.001$), and severe deformity (score 4.5-6) was recorded in 28/73 (38.4%) against 16/71 (22.5%) children ($\chi^2 = 19.83$, df = 2, $P < 0.001$). The difference held per affected foot ($U = 10101.5$, $P < 0.001$). Pirani scores were missing for two idiopathic children.

Characteristic	Syndromic (*n* = 73)	Idiopathic (*n* = 73)	Test statistic	*P*-value
Bilateral, *n* (%)	51 (69.9)	44 (60.3)	$\chi^2 = 2.85$ (df = 2)	0.241
Unilateral right, *n* (%)	15 (20.5)	15 (20.5)		
Unilateral left, *n* (%)	7 (9.6)	14 (19.2)		
Worst-foot Pirani score, median (IQR)*	3.5 (2.5-5.0)	2.0 (0.5-3.75)	*U* = 3682.5	<0.001
Pirani score per affected foot, median (IQR)†	3.5 (2.5-5.1)	1.5 (0.5-3.5)	*U* = 10101.5	<0.001
Severity*				
Mild (0-2), *n* (%)	14 (19.2)	39 (54.9)	$\chi^2 = 19.83$ (df = 2)	<0.001
Moderate (2.5-4), *n* (%)	31 (42.5)	16 (22.5)		
Severe (4.5-6), *n* (%)	28 (38.4)	16 (22.5)		

TABLE 4: Laterality and severity of syndromic and idiopathic clubfoot.

*Idiopathic *n* = 71 (Pirani score missing for two children).

†Syndromic 124 feet, idiopathic 113 feet.

U, Mann-Whitney U test; IQR, interquartile range

Spectrum of syndromic diagnoses

The associated conditions are grouped in Table 5, each child counted once under the leading diagnosis. AMC was the commonest (22/73, 30.1%), and seven of these children had a further anomaly (cerebral palsy 2, syndactyly 2, developmental dysplasia of the hip 1, amniotic bands 1, achondroplasia 1). Associated limb anomalies (10/73, 13.7%), Down syndrome (9/73, 12.3%) and other systemic anomalies (8/73, 11.0%) followed. In 9/73 (12.3%) children, syndromic features were recorded but no unifying diagnosis had been established at the time of the study. Children with AMC had the most severe deformity (median worst-foot Pirani 4.5), and those with neural axis disorders the least (median 2.25).

Leading diagnosis	Conditions included (number of children)	*n* (%)	Bilateral, *n* (%)	Worst-foot Pirani, median (IQR)
Arthrogryposis multiplex congenita	AMC alone (15); with cerebral palsy (2), syndactyly (2), DDH (1), amniotic bands (1), achondroplasia (1)	22 (30.1)	18 (81.8)	4.5 (4.0-5.9)
Associated limb anomalies	Hand defects or club hand (4), syndactyly (2), congenital ray absence (1), toe aplasia (1), elbow flexion deformity (1), DDH with stiff knee (1)	10 (13.7)	6 (60.0)	3.5 (2.5-4.9)
Down syndrome (trisomy 21)	Down syndrome alone (7); with DDH (1); with amniotic bands (1)	9 (12.3)	7 (77.8)	3.5 (2.5-4.0)
Syndromic features, diagnosis not established	Dysmorphic or multisystem features under evaluation	9 (12.3)	6 (66.7)	3.0 (2.0-4.5)
Other systemic congenital anomalies	Congenital heart disease (3, one with cleft lip), anorectal malformation (1), hypospadias (1), cryptorchidism (1), incomitant squint (1), ectropion with micrognathia (1)	8 (11.0)	5 (62.5)	3.5 (3.0-4.1)
Amniotic band sequence	Bands alone (4); with syndactyly (2); with limb amputation (1)	7 (9.6)	3 (42.9)	4.0 (3.5-4.5)
Neural axis disorders	Cerebral palsy (2), hydrocephalus (2), spina bifida with hydrocephalus (1), sacral agenesis (1)	6 (8.2)	4 (66.7)	2.25 (2.0-2.5)
Other named genetic syndromes	Apert syndrome (1), Marfan syndrome (1)	2 (2.7)	2 (100.0)	3.75 (2.6-4.9)

TABLE 5: Spectrum of associated diagnoses among children with syndromic clubfoot (n = 73).

AMC, arthrogryposis multiplex congenita; DDH, developmental dysplasia of the hip; IQR, interquartile range

Factors independently associated with syndromic clubfoot

Table *6* presents the logistic regression comparing syndromic with idiopathic clubfoot. After adjustment, four factors were independently associated with syndromic clubfoot: previous foetal loss (AOR 11.45; 95% CI 4.08-32.15; $P < 0.001$), abnormal foetal presentation (AOR 5.87; 1.96-17.55; $P = 0.002$), a previous preterm or post-term delivery (AOR 11.39; 1.94-66.93; $P = 0.007$) and maternal education up to primary level (AOR 4.11; 1.10-15.40; $P = 0.036$). Upper-lower or lower socioeconomic class (AOR 0.19; 0.07-0.49; $P < 0.001$) and use of medicines beyond routine supplements (AOR 0.17; 0.04-0.69; $P = 0.013$) were inversely associated. Paternal tribal ethnicity and infection during pregnancy were not significant after adjustment. The model was significant (likelihood-ratio $\chi^2 = 74.78$, df = 8, $P < 0.001$), fitted well (Hosmer-Lemeshow $\chi^2 = 8.32$, df = 8, $P = 0.403$), explained 53.4% of the variation (Nagelkerke $R^2 = 0.534$), and discriminated well (AUC 0.876; 95% CI 0.816-0.928), classifying 118/146 (80.8%) children correctly. The largest VIF was 1.28.

Factor	Syndromic (*n* = 73), *n* (%)	Idiopathic (*n* = 73), *n* (%)	Crude OR (95% CI)	Wald χ^2, *P*	AOR (95% CI)	Wald χ^2, *P*
Previous foetal loss	36 (49.3)	11 (15.1)	5.48 (2.49-12.07)	17.90, <0.001	11.45 (4.08-32.15)	21.41, <0.001
Abnormal foetal presentation	27 (37.0)	11 (15.1)	3.31 (1.49-7.35)	8.63, 0.003	5.87 (1.96-17.55)	10.04, 0.002
Previous preterm or post-term delivery	10 (13.7)	2 (2.7)	5.63 (1.19-26.70)	4.75, 0.029	11.39 (1.94-66.93)	7.25, 0.007
Maternal education illiterate/primary	20 (27.4)	6 (8.2)	4.21 (1.58-11.24)	8.26, 0.004	4.11 (1.10-15.40)	4.40, 0.036
Paternal tribal ethnicity	14 (19.2)	4 (5.5)	4.09 (1.28-13.11)	5.63, 0.018	3.47 (0.86-14.07)	3.05, 0.081
Upper-lower/lower socioeconomic class	22 (30.1)	41 (56.2)	0.34 (0.17-0.67)	9.82, 0.002	0.19 (0.07-0.49)	11.82, <0.001
Infection during pregnancy	15 (20.5)	9 (12.3)	1.84 (0.75-4.52)	1.76, 0.184	3.56 (0.87-14.62)	3.10, 0.078
Medicines other than routine supplements	9 (12.3)	19 (26.0)	0.40 (0.17-0.96)	4.25, 0.039	0.17 (0.04-0.69)	6.17, 0.013

TABLE 6: Binary logistic regression of factors associated with syndromic versus idiopathic clubfoot (n = 146).

Reference category for each factor, absence of the factor (for socioeconomic class, upper/upper middle and lower middle combined). Model, likelihood-ratio χ^2 = 74.78, df = 8, *P* < 0.001; Nagelkerke R^2 = 0.534; Hosmer-Lemeshow χ^2 = 8.32, df = 8, *P* = 0.403; AUC 0.876 (95% CI 0.816-0.928).

OR, odds ratio; AOR, adjusted odds ratio; CI, confidence interval; AUC, area under the receiver operating characteristic curve

Discussion

In this tertiary-centre study from Assam, one in five children with clubfoot (73/360, 20.3%) had syndromic clubfoot. Their deformity was more severe than in idiopathic clubfoot, though bilateral involvement was not significantly commoner. Previous foetal loss, abnormal foetal presentation, a previous preterm or post-term delivery and low maternal education were independently associated with syndromic clubfoot.

The proportion we observed is consistent with the roughly 20% of clubfeet that are non-isolated in large series [5]. The figure of 0.46% (73/15,799) among outpatient attendees reflects referral patterns and case mix at a tertiary centre and should not be interpreted as a birth prevalence; population estimates for South-East Asia are of the order of 1.8 per 1,000 births for all clubfoot [1]. AMC was the leading diagnosis, as reported in previous reviews of syndromic and non-idiopathic clubfoot [6,7]. Children with AMC also had the stiffest feet (median worst-foot Pirani score 4.5). This matters for planning: arthrogrypotic clubfeet can usually be corrected with the Ponseti method, but they relapse more often, carry residual deformity and functional limitation at 5 and 10 years, and frequently need further surgery [8-10]. Clubfeet associated with amniotic band sequence also respond less well than idiopathic feet [11]. The higher Pirani scores in our syndromic group, recorded on a scale that predicts the number of casts needed [23], point to the same longer treatment course. The higher Pirani scores observed in the syndromic group indicate greater deformity at the time of study assessment. However, as pre-treatment scores were not consistently available and previous treatment exposure may have influenced the recorded scores, these findings should not be interpreted as evidence of a longer treatment course in our study.

Bilateral involvement was frequent in both groups (69.9% in syndromic vs. 60.3% in idiopathic clubfoot), with no statistically significant difference between them. This suggests that laterality alone may not be a reliable clinical feature for distinguishing syndromic from idiopathic clubfoot.

Previous foetal loss was the strongest factor. Nearly half the mothers of syndromic children (36/73, 49.3%) reported a previous miscarriage or stillbirth, against 11/73 (15.1%) mothers of idiopathic children and 1/73 (1.4%) in the reference group. Several mechanisms are plausible: chromosomal abnormalities, of which Down syndrome was represented here; heritable syndromes such as distal arthrogryposis, which can recur within families [6]; and uterine or placental factors shared across pregnancies. A large Japanese birth cohort

found that women with recurrent pregnancy loss had more obstetric complications in later pregnancies but no excess of congenital anomalies among live births [25]. Our association may therefore be specific to syndromic conditions, or it may be inflated by recall, since mothers of children with multiple anomalies may search their histories more thoroughly. In either case, a history of foetal loss in the mother of a child with clubfoot is cheap to elicit and should prompt a careful search for associated anomalies and referral for genetic evaluation.

Abnormal foetal presentation, mostly breech, was associated with syndromic clubfoot independently of other factors. Breech presentation is a recognised correlate of idiopathic clubfoot through foetal constraint [15], but in syndromic clubfoot it is more likely a consequence of reduced foetal movement, as occurs in AMC and neuromuscular disorders, than a cause. The higher NICU admission rate (27/73, 37.0% vs. 15/73, 20.5%) is consistent with that interpretation. A previous preterm or post-term delivery was also associated, but it rests on 10 and 2 exposed mothers, and its wide CI (1.94-66.93) calls for confirmation.

The sociodemographic findings need careful reading. Low maternal education was independently associated with syndromic clubfoot, and paternal tribal ethnicity was associated before but not after adjustment. These may reflect less antenatal ultrasound and later recognition of anomalies in disadvantaged and tribal communities, rather than any biological effect. Consanguinity was uncommon and did not differ between the clubfoot groups, consistent with the low prevalence of consanguineous marriage reported in Assam [4]. Conversely, idiopathic families were more often in the lowest socioeconomic classes. We interpret this as a referral effect: idiopathic clubfoot is identified at birth and referred through India's free Ponseti services, whereas a larger share of syndromic children were born in private hospitals (12/73, 16.4% vs. 4/73, 5.5%) and reached the clinic through other routes. The inverse association with use of medicines in pregnancy probably reflects reporting differences, and our data provide no basis for inferences about teratogenic or protective effects.

Family history of clubfoot, the most consistent risk factor for idiopathic clubfoot [14,17], did not distinguish the two clubfoot groups and was absent in the reference group. The reference group differed from both clubfoot groups in age, education and socioeconomic class, and reported more hyperemesis, oligohydramnios and anaemia. These differences show that trauma attendees are not exchangeable with children born with clubfoot, which is why the syndromic-idiopathic contrast was pre-specified as the primary analysis. The reference group remains useful as a benchmark: previous foetal loss in 1/73 (1.4%) of its mothers shows how unusual the 36/73 (49.3%) in the syndromic group is.

These findings have practical implications for Northeast India. Every newborn screened for clubfoot under the national child health screening programme should have a systematic head-to-toe examination for associated anomalies, and the obstetric history should include previous pregnancy losses. Children with syndromic clubfoot need multidisciplinary review, genetic counselling where possible, and a treatment plan that anticipates more casts, more relapse and longer follow-up than for idiopathic clubfoot [7,12,19,20].

Limitations

This was a single-centre, hospital-based study, so referral patterns shape the case-mix and limit generalisability. The reference group consisted of children attending for trauma; they were older and socioeconomically different, so comparisons with them are descriptive only. Obstetric and antenatal histories were largely recalled and may be subject to differential recall. Associated diagnoses were made clinically, without uniform genetic testing, and 9/73 (12.3%) syndromic children had no established unifying diagnosis. Pre-treatment Pirani scores were not consistently available for all participants, particularly those who had initiated treatment before enrolment. Consequently, the severity comparisons were based on Pirani scores recorded at the time of study assessment, which may not accurately reflect the initial severity of deformity. Previous treatment may have influenced the recorded scores. Use of medicines was not recorded in the reference group. Furthermore, treatment-related outcomes, including the number of casts required, duration of treatment and relapse rates, were not assessed in this study. Therefore, no direct conclusions regarding differences in treatment requirements between the two groups can be drawn from our findings. The modified Kuppuswamy scale was developed for urban families and may misclassify rural households. Several estimates rest on small numbers and have wide CIs, and the many bivariate comparisons raise the chance of false-positive findings. Finally, the design cannot establish the temporal order of all exposures, and treatment outcomes were not assessed.

Conclusions

Syndromic clubfoot made up one-fifth of all clubfoot cases seen at this tertiary centre in Assam. Arthrogryposis multiplex congenita was the leading association, and syndromic feet were more severely deformed than idiopathic feet, although bilateral involvement was similar. Previous foetal loss, abnormal foetal presentation, a previous preterm or post-term delivery and low maternal education were independently associated with syndromic clubfoot.

These markers are simple to obtain at the first visit. Their presence in a child with clubfoot should prompt a systematic search for associated anomalies, referral for genetic evaluation and counselling, and a treatment

plan that anticipates more casts, relapse and longer follow-up. Multicentre studies with standardised genetic evaluation and prospective follow-up are needed to confirm these associations across Northeast India.

Additional Information

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All authors have reviewed the final version to be published and agreed to be accountable for all aspects of the work.

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