Epidemiology of clubfoot in Sweden from 2016 to 2019: A national register study

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Abstract

Background
This study aimed to estimate the birth prevalence of children born with isolated or non-isolated clubfoot in Sweden using a national clubfoot register. Secondarily we aimed to describe the clubfoot population with respect to sex, laterality, severity of deformity, comorbidity and geographic location.

Methods
A national register, the Swedish Pediatric Orthopedic Quality register, was used to extract data on newborn children with clubfoot. To calculate the birth prevalence of children with isolated or non-isolated clubfoot between 1st of January 2016 and 31st of December 2019, we used official reports of the total number of Swedish live births from the Swedish Board of Statistics. The Pirani score and predefined signs of atypical clubfoot were used to classify clubfoot severity at birth.

Results
In total 612 children with clubfoot were identified. Of these, 564 were children with isolated clubfoot, generating a birth prevalence of 1.24/1000 live births (95% confidence interval 1.15–1.35). About 8% were children with non-isolated clubfoot, increasing the birth prevalence to 1.35/1000 live births (95% confidence interval 1.25–1.46). Of the children with isolated clubfoot, 74% were boys and 47% had bilateral involvement. The children with non-isolated clubfoot had more severe foot deformities at birth and a greater proportion of clubfeet with atypical signs compared with children with isolated clubfoot.

Conclusion
We have established the birth prevalence of children born with isolated or non-isolated clubfoot in Sweden based on data from a national register. Moreover, we have estimated the number of children born with atypical clubfeet in instances of both isolated and non-isolated clubfoot. These numbers may serve as a baseline for expected birth prevalence when planning clubfoot treatment and when evaluating time trends of children born with clubfoot.
Introduction

Congenital talipes equinovarus, more commonly known as clubfoot, is a common congenital orthopedic pediatric foot deformity [1]. Clubfoot is characterized by equinus of the ankle, varus of the hindfoot, as well as cavus and forefoot adduction with associated atrophy of the calf muscles [2]. The most common presentation is isolated congenital, even though clubfoot may be associated with other conditions such as myelomeningocele or arthrogryposis [3]. The cause of clubfoot is not fully known, but a multifactorial model including both genetic and environmental factors is likely [3]. About two-thirds of the children with clubfoot are boys, and 50% have bilateral involvement. For those with unilateral involvement, there is according to some studies, a slight right-sided preponderance [4]. The Ponseti method is currently considered the gold standard for treatment of clubfoot and involves weekly strict manipulation and casting. An Achilles tenotomy is performed when there is rigid equinus and all other deformities of the foot have been corrected. This is followed by brace treatment for four to five years [5].

Global birth prevalence of children born with clubfoot varies but is often reported to be between 0.4 and 2.0/1000 live births, commonly averaged to around 1/1000 live births [1,6]. Higher rates, between 3 and 6/1000 live births are, among others, reported from Malaysia, the Polynesian population and the aboriginals in Australia [7–9]. In the Scandinavian countries during 1936–1996, the rate of children with clubfoot has been reported to be between 0.7 and 1.4/1000 live births [10–14]. The highest rate being reported from a Swedish national retrospective multicenter study from 1995 to 1996 [13].

Epidemiological studies are important to improve treatment and to ensure equal care despite residential location. The information can be used to plan and evaluate treatment procedures and to compare outcomes among different centers. Epidemiological data can be used to identify factors influencing and/or causing clubfoot. In 2015, the Swedish Pediatric Orthopedic Quality register (SPOQ) was designated as a national quality register covering five common pediatric diseases, of which clubfoot is one [15]. Of all 28 pediatric orthopedic centers treating clubfoot in Sweden, 27 (96%) agreed on reporting to SPOQ. The general aim of this national prospective total cohort register is to gain generalizable knowledge, to improve outcomes and to ensure equal care for all children born with clubfoot. In addition to the number of children with clubfoot, the severity of deformity including atypical signs and other diseases related to clubfoot have been registered from the start.

Aim

The primary aim of this study was to estimate the number of children born with isolated or non-isolated clubfoot in Sweden from 2016 to 2019 to create a baseline for future studies on birth prevalence trends. Secondarily, we aimed to describe the clubfoot population with respect to sex, laterality, severity of deformity, comorbidity and geographic location.

Materials and methods

At birth, upon suspicion of clubfoot, the child is referred to one of the 28 pediatric orthopedic centers treating clubfoot in Sweden. Twenty-seven of these centers report to SPOQ and the coverage of the register since 2017 is 96%. Since practically all children in Sweden are born in a hospital, there are rarely any undiagnosed cases. Register data is completed by an orthopedic surgeon or a specialized physiotherapist at diagnosis using a guided checklist. To validate the number of newborn children with clubfoot registered in SPOQ, numbers were compared with those in the Swedish national patient register (PAR). Healthcare providers in Sweden are required to report ICD-10-CM codes (Q66.0) to PAR when the children are diagnosed.
averaged national completeness, thus the agreement between PAR and SPOQ, from 2016 to 2019 was 84% [15]. Every individual living in Sweden has a personal ID number (birth date [6 digits] and a unique suffix [4 digits]). The number is exclusive to an individual and is not changed during a person’s life, making comparisons between registries possible.

**Ethics statement**

The study was approved by the Swedish Ethical Review Authority, Dnr: 2019–04989. The data we have used is manually registered in a specific national quality register separate from patients’ medical records. Use of data is regulated by the Swedish patient data act, in the part that applies to national quality registers. In order to be registered in a quality register, it is required that the patient is informed and given the opportunity to opt out. The information must include that the data may be used for research after approval from a research ethical board. The research ethical board decides if consent is required or not, and if data should be anonymized. In our case the decision was made that no further information or consent is required, and that data must be anonymized.

**Participants**

After a clubfoot diagnosis, the child is enrolled in SPOQ by the treating hospital. Inclusion criteria in SPOQ are diagnosis of clubfoot defined as ICD-10-CM code Q66.0; born in Sweden and having a Swedish personal ID or born abroad but not started clubfoot treatment abroad and have verified contact with the Swedish health care system within six weeks of age. Exclusion criteria in SPOQ are clubfoot treatment started abroad or born abroad and not having a Swedish personal ID number at first contact with the Swedish health care system. To be registered in SPOQ, clubfoot casting treatment must start before the age of 15 months and be completed before the age of 21 months. Children with clubfoot as part of a neurological or other disease are also registered. The gold standard for treatment of children with clubfoot in Sweden is the Ponseti method [16]. In this prospective cohort study, all 612 children with a total 905 clubfeet registered in SPOQ from the 1st of January 2016 to the 31st of December 2019 were included.

For our study, the following variables were extracted from SPOQ: number of children with isolated congenital clubfoot (from now on referred to as isolated clubfoot); number and description of children with clubfoot as part of another disease (from now on referred to as non-isolated clubfoot); time between birth and first assessment by an orthopedic surgeon or specialized physiotherapist; gender; uni- or bilateral involvement, if unilateral, side; Pirani score before start of treatment; and description of other diseases affecting the child’s development or treatment.

The Pirani score is a disease-specific foot deformity classification system, scoring the foot deformity from 0 (no foot deformity) to 6 (maximal foot deformity) [17–19]. In SPOQ, feet scoring 1 or above are defined as clubfeet and are possible to register. Feet scoring less than 1 (0 or 0.5) are classified as positional clubfeet or other minor foot deformity and are not possible to register in SPOQ [15].

In SPOQ, children with non-isolated clubfoot as part of another disease are reported upon entry in the register in any of the following categories: arthrogryposis multiplex congenita Q74.3, spina bifida Q05.9, congenital malformation syndromes predominantly involving limbs Q87.2, neurological diseases (not specified) or other (not specified). Numbers of non-isolated clubfoot were adjusted based on updated reports at the one-year follow-up. Orthopedic centers are also encouraged to register “other diseases potentially affecting the child’s development or treatment” and to describe the condition in their own words. This is an open
question, and no guidelines for reporting are provided from the register. For assessing the presence of atypical signs, SPOQ provides several illustrations to help clinicians to report this correctly. Presence of congenital atypical clubfoot is registered upon entry and is defined by two compulsory signs: distinct cavus and deep plantar creases crossing over to the lateral side, and five additional signs: short and stubby foot, extended greater toe, deep posterior crease, distinct equinus > 60˚, and short calf muscles < 1/3 of the calf’s length. In this study, atypical feet were defined as isolated clubfoot if no other diagnosis related to clubfoot was registered.

Data and statistical analyses
Statistical analyses were performed using the Statistical Package for Social Sciences, version 25 (SPSS Inc., Chicago, IL, USA) or the interactive calculator Epitools for Wilson’s score confidence intervals (CIs) [20]. To compare the annual number of newborn children with clubfoot with the total number of live births between 1st of January 2016 and 31st of December 2019, we used official reports concerning native data from Statistics Sweden [21]. The birth prevalence of children with clubfoot per thousand live births was calculated as the ratio between the number of children with clubfoot born between 2016 and 2019 and the number of live births during the same period. For calculating the prevalence of children born with clubfoot, Wilson’s score CIs were estimated both for isolated and non-isolated clubfoot for both genders and for each of the six official healthcare regions in Sweden, commonly used by Health Service Authorities for administrative purposes. The birth prevalence of children born with isolated clubfoot was estimated for each of the four included years separately and summarized. Only live births are registered in SPOQ; hence, abortive cases and stillborn children were not included. Norrbotten county in Sweden was not reporting children with clubfoot to the national register between 2016 and 2019; therefore, the annual numbers of live-born children from that county, in total 9784, were subtracted from the total number of live-born children in Sweden during those years.

Demographics and disease characteristics are described using median and minimum and maximum, frequency or percent. A one-sample t-test was used to evaluate differences between the proportion of boys/ girls, uni-/ bilateral involvement, and in children with unilateral involvement, between the right/ left sides. Differences in Pirani score between children with isolated clubfoot and those with non-isolated clubfoot were evaluated using the Mann–Whitney U test. Differences in the proportion of feet with atypical signs between children with isolated clubfoot and those with non-isolated clubfoot were evaluated using a t-test for independent samples. To account for the effect of bilateral disease when evaluating the difference in Pirani score and proportion of feet with atypical signs between children with isolated and those with non-isolated clubfoot, we performed a sensitivity analysis. In this sensitivity analysis, we randomly included one clubfoot from each of the children with bilateral disease. The results from the two different approaches (one or both randomly selected bilateral clubfeet) did not differ. The results presented in this article include both clubfeet in bilateral cases. Differences were considered statistically significant for P-values < 0.05.

Results
From the 1st of January 2016 to the 31st of December 2019, 564 children with isolated clubfoot were registered generating a birth prevalence of 1.24/1000 (95% CI 1.15–1.35) live births. Forty-eight children with non-isolated clubfoot were registered over the same time period generating a birth prevalence of 0.11/1000 (95% CI 0.08–0.14). In total, 612 children with isolated or non-isolated clubfoot were registered, and the birth prevalence was estimated as 1.35/1000 (95% CI 1.25–1.46). As shown in Table 1, there were slight variations between years.
Isolated clubfoot

Of the 564 children with isolated clubfoot, 74% were boys (mean diff. 0.24 [95% CI 0.21–0.28]) and 53% of the children had a unilateral involvement (mean diff. 0.03 [95% CI –0.01 to 0.07]), indicating no significant statistical difference between numbers of children with uni- or bilateral involvement. There was no statistically significant difference between the numbers of children with left- (47%) or right- (53%) sided unilateral involvement (mean diff. 0.03 [95% CI –0.03 to 0.09]) (Table 2).

Of the 564 children diagnosed with isolated clubfoot, clinicians reported other disease or condition with possible effect on development in 25 children (4.4%): prematurity (n = 9), postnatal sepsis or infection (n = 4), hip dysplasia (n = 3), congenital heart defect (n = 3), single kidney (n = 1), skin disease (n = 1), contralateral vertical talus (n = 1), contralateral metatarsus varus adducts (n = 1), abnormal pelvis (n = 1) and missing distal phalanges toes (n = 1).

Non-isolated clubfoot

The 48 children (with a total of 77 clubfeet) with non-isolated clubfoot were reported in combination with Arthrogryposis multiplex congenita (Q74.3), n = 8; Spina bifida (Q05.9), n = 5; congenital malformation syndromes predominantly involving limbs (Q87.2), n = 18; neurological diseases (not specified), n = 7; and other (not specified), n = 10. For one child, the disease was not classified. Of the 48 children, 52% were boys and 60% had bilateral involvement.

Table 1. Newborn children with clubfoot in Sweden from 2016 to 2019.

| Isolated clubfoot | Children with clubfoot, n | Live births in Sweden, n | Birth prevalence per thousand live births (95% CI) |
|-------------------|--------------------------|--------------------------|-----------------------------------------------|
| 2016              | 144                      | 114 884                  | 1.25 (1.06–1.48)                              |
| 2017              | 121                      | 113 003                  | 1.07 (0.90–1.28)                              |
| 2018              | 159                      | 113 459                  | 1.40 (1.20–1.64)                              |
| 2019              | 140                      | 112 066                  | 1.25 (1.06–1.47)                              |
| Total 2016–2019   | 564                      | 453 412                  | 1.24 (1.15–1.35)                              |
| Boys, isolated clubfoot, 2016–2019 | 420                      | 233 020                  | 1.80 (1.64–1.98)                              |
| Girls, isolated clubfoot, 2016–2019 | 144                      | 220 392                  | 0.65 (0.56–0.77)                              |
| Total children non-isolated clubfoot 2016–2019 | 48                      | 453 412                  | 0.11 (0.08–0.14)                              |
| Total registered children with clubfoot 2016–2019 | 612                      | 453 412                  | 1.35 (1.25–1.46)                              |

SPOQ, Swedish Pediatric Orthopedic Quality register; n, number of children; CI, confidence intervals. Years refers to birth years.

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Table 2. Description of children with isolated clubfoot.

| Year | Total n of children | Boys n (%) | Girls n (%) | Bilateral n (%) | Unilateral n (%) | Uni right n (%) | Uni left n (%) |
|------|---------------------|------------|-------------|-----------------|-----------------|----------------|---------------|
| 2016 | 144                 | 104 (72)   | 40 (28)     | 64 (44)         | 80 (56)         | 46 (58)        | 34 (42)       |
| 2017 | 121                 | 94 (78)    | 27 (22)     | 55 (46)         | 66 (55)         | 25 (38)        | 41 (62)       |
| 2018 | 159                 | 119 (75)   | 40 (25)     | 79 (50)         | 80 (50)         | 48 (60)        | 32 (40)       |
| 2019 | 140                 | 103 (74)   | 37 (26)     | 66 (47)         | 74 (53)         | 40 (54)        | 34 (46)       |
| Total| 564                 | 420 (74) a | 144 (26) a  | 264 (47) b      | 300 (53) b      | 159 (53) c     | 141 (47) c    |

n, numbers of children.

a There were significantly more boys than girls.

b No statistical difference between numbers of children with uni- or bilateral involvement.

c In children with unilateral clubfoot, no statistical difference between numbers of right or left involvement was observed.

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Severity of clubfoot deformity

The clubfeet (n = 828) in children with isolated clubfoot had a median Pirani score of 4.5 (range 1–6). The Pirani score in children with non-isolated clubfoot (n = 77) was significantly higher (median 5.5, range 1–6) (P < 0.001; Table 3, Fig 1). Of the 828 isolated clubfeet, 6% (46 feet) were classified as atypical at birth. A statistically significantly higher proportion of the 77 non-isolated clubfeet, 36% (28 feet), was classified as atypical at birth (mean diff. 0.31 (95% CI 0.25–0.37) (Table 3).

Table 3. Pirani score and presence of atypical signs at birth in clubfeet.

| Year | Total n of clubfeet | Total score median (min, max) | Atypical n (%) | Total n clubfeet | Total score median (min, max) | Atypical n (%) |
|------|---------------------|-------------------------------|---------------|----------------|-------------------------------|---------------|
| 2016 | 208                 | 4.5 (1, 6)                    | 18 (9)        | 31             | 5.5 (1.5, 6)                  | 6 (19)        |
| 2017 | 176                 | 4.5 (1, 6)                    | 13 (7)        | 20             | 5.25 (3.5, 6)                 | 9 (45)        |
| 2018 | 238                 | 4.5 (1, 6)                    | 8 (3)         | 10             | 5.0 (3.5, 6)                  | 3 (30)        |
| 2019 | 206                 | 4.5 (1.5, 6)                  | 7 (3)         | 16             | 6.0 (3.6)                     | 10 (63)       |
| Total| 828                 | 4.5 (1, 6)*                   | 46 (6)*       | 77             | 5.5 (1.5, 6)*                 | 28 (36)*      |

a The Pirani score of the clubfeet in children with non-isolated clubfoot was significantly higher compared with children with isolated clubfoot.

b The proportion of clubfeet with atypical signs was significantly higher in children with non-isolated clubfoot compared with children with isolated clubfoot.

The numbers of children with isolated clubfoot are shown in light blue, and children with clubfoot associated with other disease are shown in black.

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with 67% of the children diagnosed within two weeks. There was a national variation between the six Swedish health care regions (Fig 2).

Geographical distribution

The estimated prevalence of children with isolated or non-isolated clubfoot distributed among the six healthcare regions in Sweden is descriptively shown in Table 4. The lowest prevalence was noted in the North region and the highest in the South-East region.

Discussion

This national Swedish register study, covering almost half a million births, found 564 children with isolated clubfoot corresponding to a birth prevalence of 1.24/1000 live births. Including the children with non-isolated clubfoot, the prevalence increased to 1.35/1000 live births. The children with non-isolated clubfoot had more severe foot deformities at birth and a greater proportion of clubfeet with atypical signs.

Table 4. Children with clubfeet from 2016 to 2019 in the six healthcare regions in Sweden.

| Healthcare region | Children with isolated clubfoot, n | Children with non-isolated clubfoot, n | Total n of children with clubfoot | Live births in Sweden, n | Birth prevalence per thousand live births (95% CI) |
|-------------------|-----------------------------------|----------------------------------------|---------------------------------|--------------------------|-----------------------------------------------|
| North             | 26                                | 3                                      | 29                              | 27 400                   | 1.06 (0.74–1.52)                              |
| Middle            | 120                               | 8                                      | 128                             | 90 742                   | 1.41 (1.19–1.68)                              |
| Stockholm         | 122                               | 10                                     | 132                             | 117 444                  | 1.12 (0.95–1.33)                              |
| South-East        | 76                                | 10                                     | 86                              | 46 809                   | 1.84 (1.49–2.27)                              |
| West              | 108                               | 10                                     | 118                             | 86 282                   | 1.37 (1.14–1.64)                              |
| South             | 112                               | 7                                      | 119                             | 84 735                   | 1.40 (1.17–1.68)                              |

n, number of children; CI, confidence Interval. Live births refers to all children live born in Sweden from 2016 to 2109. The birth prevalence of clubfeet is based on the total number of children with clubfeet, including both isolated and non-isolated.
The estimated birth prevalence of clubfoot in children, both isolated and non-isolated, is in line with the prevalence reported by Wallander et al from 1995 to 1996 of 1.4/1000 live births [13]. Wang et al. reported a summarized birth prevalence of 1.08/1000 children with clubfoot from several countries in Europe between 1995 and 2011. In their report, our Scandinavian neighbors, Denmark (1.30/1000) and Norway (1.40/1000) reported birth prevalences of clubfoot in line with ours [1]. Comparing birth prevalence in our study with those in earlier studies covering the Swedish population from 1936 to 1990 (0.74–0.93/1000 live births), an increasing trend can be seen [10,11]. This increasing trend has also been described by Krogsgaard et al. evaluating the birth prevalence of Danish children born with clubfoot between 1978 and 1993. They showed a small increase in prevalence over time and an increased risk of being born with clubfoot in counties with higher population density [14]. However, others have described both stable and declining trends of clubfoot over time. Wang et al. reported declining trends in the proportion of children born with clubfoot in Europe between 1999 and 2011 [1], while Morris et al. described a stable trend in Europe between 2003 and 2012 [22]. Nevertheless, caution should be taken when comparing prevalences and trends of children born with clubfoot over time. Given different study protocols, lack of reporting and different inclusion and exclusion criteria, efforts should be taken to standardize these parameters [1,22–24].

Of the children with isolated clubfoot, 74% were boys, which is in line with earlier reports from Sweden [10,13] but slightly higher than in other studies from Europe reporting numbers between 65% and 72% [1,25]. Of the 48 children with non-isolated clubfoot, 52% were boys. Hence, in this group we saw an even distribution between genders. Our results are in line with other studies that also found gender differences are less pronounced in children with non-isolated clubfoot [7]. Of the children with isolated clubfoot, 47% had bilateral involvement, which harmonized with earlier reports from Sweden (46%) but was lower than reported numbers from Wang et al. (57%) [1,13]. While it is commonly described that the right side is slightly more often involved in children with unilateral clubfoot [7], this could not be statistically confirmed in our cohort. In accordance with others, we also found that bilateral involvement is more common in children with non-isolated clubfoot compared with those with isolated clubfoot [7]. Children with non-isolated clubfoot had significantly higher Pirani scores, indicating more severe deformities. We also saw a higher proportion of feet with atypical signs at birth in this group compared with children with isolated clubfoot. Syndromic clubfeet and feet with atypical signs are known to be more difficult to correct and might require more casting sessions and a slightly modified casting technique [26]. It is imperative to identify these feet early, adapt treatment and refer to a more specialized center if necessary. By including these parameters in the national register SPOQ, weaknesses in the treatment of a severely deformed clubfoot might be identified and corrected early. However, since atypical signs are registered upon diagnosis, and not after initial casting treatment, there might be missing cases. On the other hand, reporting atypical signs before start of treatment excludes those feet made complex/ atypical by improper casting.

A commonly presented rate of non-isolated clubfoot is between 10% and 13% [1,12,27]. In our cohort, about 8% of the children were diagnosed with clubfoot as part of another disease. Of the children reported as having isolated clubfoot, 4.4% had other conditions affecting development, but classified by the children’s healthcare providers as not directly associated with clubfoot. To validate the diagnosis of non-isolated clubfoot, reports upon entry were compared with reports from one-year follow-up, which classified seven additional children as having non-isolated clubfoot. This suggests to us that the 8% level of non-isolated clubfoot is low, and we expect this number to rise as the register matures. Moreover, the diagnoses that are included in non-isolated clubfoot vary between studies, a complication that is compounded by promising genetic research that may add further associated diagnoses in the future [28,29]. To
make valid comparisons between studies possible, the consensus in defining isolated clubfoot as well as the subgroups of non-isolated clubfoot is essential.

Striving for an early start of clubfoot treatment after birth is generally advised even though several studies have shown positive results of Ponseti treatment in older children [30]. In our study, 67% of the children were assessed at a pediatric orthopedic center within two weeks after birth, but national variations were seen. Nowadays, clubfoot in many children is already discovered during prenatal ultrasound screening, and treatment planning can start during pregnancy. Slight variations in the birth prevalence of clubfoot were seen among the different healthcare regions with the highest prevalence reported for the South-East region. This result differs from earlier reported variations in Sweden from 1995 to 1996 where the South region had the highest prevalence [13]. The lower prevalence in the North region can be explained by low record completeness found when cases of children with clubfoot in SPOQ were compared with the national patient register for this region. In the future, when SPOQ has matured to a critical number of clubfeet, these national variations can be scrutinized statistically. In addition, the distribution of children born with non-isolated clubfoot or clubfeet with atypical signs can be further assessed.

A strength of the current study is the national prospective approach of the SPOQ register with well-predefined prerequisites for registration, excluding other subtypes of foot abnormalities or postural clubfoot. Associated diseases as well as signs of atypical clubfeet are reported upon first registration, increasing the likelihood for caregivers to address these aspects. Even so, estimated prevalence is often an underestimation of the true prevalence as there are always several missing cases. Thus, our study is not an exception. In this study only live births, and not still births or interrupted gestations, were included. Comparing numbers of children with clubfoot in SPOQ with the national patient register showed an average completeness (2016–2019) of 84%. This number is influenced by missing cases of clubfoot in SPOQ but also by wrongly diagnosed and reported cases in the national patient register. Therefore, we believe this completeness is underestimated, and further validation of the data within the register is warranted.

**Conclusion**

We have established the birth prevalence of children with isolated or non-isolated clubfoot in Sweden based on data from a total population register. These numbers may serve as a baseline expected birth prevalence when planning clubfoot treatment and when evaluating time trends of children born with clubfoot.

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References

1. Wang H, Barisic I, Loane M, Addor MC, Bailey LM, Gatt M, et al. Congenital clubfoot in Europe: A population-based study. Am J Med Genet A. 2019; 179(4):595–601. https://doi.org/10.1002/ajmg.a.61067 PMID: 30740879

2. Moon DK, Gurnett CA, Aferol H, Siegel MJ, Commean PK, Dobbs MB. Soft-tissue abnormalities associated with treatment-resistant and treatment-responsive clubfoot: Findings of MRI analysis. J Bone Joint Surg Am. 2014; 96(15):1249–1256. https://doi.org/10.2106/JBJS.M.01257 PMID: 25100771

3. Dobbs MB, Gurnett CA. Genetics of clubfoot. J Pediatr Orthop B. 2012; 21(1):7–9. https://doi.org/10.1097/BPB.0b013e328349927c PMID: 21817922

4. Nguyen MC, Nhi HM, Nam VQ, Thanh do V, Romitti P, Morcuende JA. Descriptive epidemiology of clubfoot in Vietnam: A clinic-based study. Iowa Orthop J. 2012; 32:120–124. PMID: 23576932

5. Ponseti IV. Congenital clubfoot. Fundamentals of treatment. Oxford University Press; 1996. PMID: 8981905

6. Smythe H, Kuper H, Macleod D, Foster A, Lavy C. Birth prevalence of congenital talipes equinovarus in low- and middle-income countries: A systematic review and meta-analysis. Trop Med Int Health. 2017; 22(3):269–285. https://doi.org/10.1111/tmi.12833 PMID: 28000394

7. Carey M, Bower C, Mylvaganam A, Rouse I. Talipes equinovarus in Western Australia. Paediatr Perinat Epidemiol. 2003; 17(2):187–194. https://doi.org/10.1046/j.1365-3016.2003.00477.x PMID: 12675786

8. Boo NY, Ong LC. Congenital talipes in Malaysian neonates: Incidence, pattern and associated factors. Singapore Med J. 1990; 31(6):539–542. PMID: 2281348

9. Ching GH, Chung CS, Nemeck RW. Genetic and epidemiological studies of clubfoot in Hawaii: Ascertainment and incidence. Am J Hum Genet. 1969; 21(6):566–580. PMID: 5365759

10. Danielsson LG. Incidence of congenital clubfoot in Sweden. 128 cases in 138,000 infants 1946–1990 in Malmö. Acta Orthop. 2006; 77(6):847–852. https://doi.org/10.1080/17453670610013114 PMID: 17260191

11. Severin E. The frequency of congenital hip dislocation and congenital equinovarus in Sweden. Nord Med. 1956; 55(7):221–223. PMID: 13309785

12. Clubfoot Somppi E. Review of the literature and an analysis of a series of 135 treated clubfeet. Acta Orthop Scand Suppl. 1984; 209:1–109. PMID: 6377807

13. Wallander H, Hovelius L, Michaelsson K. Incidence of congenital clubfoot in Sweden. Acta Orthop. 2006; 77(6):847–852. https://doi.org/10.1080/17453670610013125 PMID: 17260191

14. Krogsgaard MR, Jensen PK, Kjaer I, Hustad H, Lorentzen J, Hvass-Christensen B, et al. Increasing incidence of club foot with higher population density: Incidence and geographical variation in Denmark over a 16-year period—A epidemiological study of 936,525 births. Acta Orthop. 2006; 77(6):839–846. https://doi.org/10.1080/17453670610013114 PMID: 17260190

15. spoq.registercentrum.se [Internet]. Svenskt Pediatrisk Ortopediskt Qualitetsregister; c2021 [cited 2021 June 7]. Available from: http://www.spoq.registercentrum.se/.

16. Ponseti IV. The treatment of congenital clubfoot. J Orthop Sports Phys Ther. 1994; 20(1):1. https://doi.org/10.2519/jospt.1994.20.1.1 PMID: 8081402

17. Shaheen S, Jaiballa H, Pirani S. Interobserver reliability in Pirani clubfoot severity scoring between a paediatric orthopaedic surgeon and a physiotherapy assistant. J Pediatr Orthop B. 2012; 21(4):366–368. https://doi.org/10.1097/BPB.0b013e3283514183 PMID: 22343939

18. Jain S, Ajmera A, Solanki M, Verma A. Interobserver variability in Pirani clubfoot severity scoring system between the orthopedic surgeons. Indian J Orthop. 2017; 51(1):81–85. https://doi.org/10.4103/0019-5413.19755 PMID: 28216755

19. Bettuzzi C, Abati CN, Salvatori G, Zanardi A, Lampasi M. Interobserver reliability of Dimeglio and Pirani score and their subcomponents in the evaluation of idiopathic clubfoot in a clinical setting: a need for improved scoring systems. J Child Orthop. 2019; 13(5):467–485. https://doi.org/10.1007/s11833-019-00645-y PMID: 31695815

20. Epitools (Summarise categorical or continuous data). Calculate confidence limits for a sample proportion [software]; c2021 [cited 2021 June 7]. Available from: https://epitools.ausvet.com.au/cipropportion? page=CIPropportion

21. scb.se/en [Internet]. Statistics Sweden; c2021 [cited 2021 June 7]. Available from: http://www.scb.se/en/
22. Morris JK, Springett AL, Greenlees R, Loane M, Addor MC, Arriola L, et al. Trends in congenital anomalies in Europe from 1980 to 2012. PLoS One. 2018; 13(4):e0194986. https://doi.org/10.1371/journal.pone.0194986 PMID: 29621304

23. Kancherla V, Romitti PA, Caspers KM, Puzhankara S, Morcuende JA. Epidemiology of congenital idiopathic talipes equinovarus in Iowa, 1997–2005. Am J Med Genet A. 2010; 152A(7):1695–1700. https://doi.org/10.1002/ajmg.a.33481 PMID: 20583169

24. Mai CT, Isenburg JL, Canfield MA, Meyer RE, Correa A, Alverson CJ, et al. National population-based estimates for major birth defects, 2010–2014. Birth Defects Res. 2019; 111(18):1420–1435. https://doi.org/10.1002/bdr2.1589 PMID: 31580536

25. Cardy AH, Sharp L, Torrance N, Hennekam RC, Miedzybrodzka Z. Is there evidence for aetologically distinct subgroups of idiopathic congenital talipes equinovarus? A case-only study and pedigree analysis. PLoS One. 2011; 6(4):e17895. https://doi.org/10.1371/journal.pone.0017895 PMID: 21533128

26. Ponseti IV, Zhivkov M, Davis N, Sinclair M, Dobbs MB, Morcuende JA. Treatment of the complex idiopathic clubfoot. Clin Orthop Relat Res. 2006; 451:171–176. https://doi.org/10.1097/01.blo.0000224062.39990.48 PMID: 16788408

27. Todd M, Kodovaru SS, Antoniou G, Cundy PJ. Clubfoot deformity in the Solomon Islands: Melanesian versus Polynesian ethnicity, a retrospective cohort study. J Child Orthop. 2020; 14(4):281–285. https://doi.org/10.1302/1863-2548.14.190172 PMID: 32874360

28. Dobbs MB, Gurnett CA. Update on clubfoot: Etiology and treatment. Clin Orthop Relat Res. 2009; 467(5):1146–1153. https://doi.org/10.1007/s11999-009-0734-9 PMID: 19224303

29. Sadler B, Gurnett CA, Dobbs MB. The genetics of isolated and syndromic clubfoot. J Child Orthop. 2019; 13(3):238–244. https://doi.org/10.1007/s11999-018-3458-0 PMID: 31312262

30. Liu YB, Li SJ, Zhao L, Yu B, Zhao DH. Timing for Ponseti clubfoot management: Does the age matter? 90 children (131 feet) with a mean follow-up of 5 years. Acta Orthop. 2018; 89(6):662–667. https://doi.org/10.1080/17453674.2018.1526534 PMID: 30334643