

ORIGINAL ARTICLE

Medicine Science 2020;9(3):556-9

The frequency and causes of pes equinavarus in the neonatal intensive care unit in a tertiary care center from eastern in anatolia**^{ID}Sezai Ozkan¹, ^{ID}Cihan Adanas¹, ^{ID}Murat Basaranoglu²**¹*Van Yuzuncu Yil University Faculty of Medicine Department of Orthopedics and Traumatology, Van, Turkey*²*Yuzuncu Yil University Faculty of Medicine, Department of Neonatology, Van, Turkey*

Received 24 February 2020; Accepted 08 April 2020

Available online 16.05.2020 with doi: 10.5455/medscience.2020.09.9234

Abstract

PEV is one of the most common deformities in the foot. The incidence of PEV varies between communities and many theories about its etiology have been proposed. In this study, we aimed to share our experience with the incidence and etiology of PEV in the neonatal intensive care unit of a tertiary hospital. 3658 patients who were followed up in a tertiary neonatal intensive care unit from 2014 to 2017 were included in the study. Data on the number and etiology of newborns with PEV deformity were obtained from the automation records of our hospital. PEV deformity was diagnosed in 32 (0.87%) of 3658 babies treated in the neonatal intensive care unit over a period of 3 years. Of these 32 patients, 18 (56.25%) were bilateral, and 14 (43.75%) unilateral PEV. Two (6.25%) of our patients had a positive family history. While 23 (71.87%) of our patients were delivered by cesarean, 9 (28.1%) of our patients were born normally. Considering birth weights, the number of babies born under 2500 grams was 7 (21.8%), and the number of babies born above 2500 grams was 25 (78.1%). Considering the data we obtained at the end of the study, the incidence of PEV was lower than in the literature. In fact, our hypothesis before starting the study was that the incidence of PEV was higher in the neonatal intensive care unit. Although the frequency of PEV is known to vary between populations, the etiology of PEV is not fully known. We believe that future studies (such as genetic studies) will be more beneficial to science than classical knowledge about PEV etiology.

Keywords: Pes equinavarus, deformity, newborn, anatolia**Introduction**

Pes equinovarus (PEV) is the most common congenital deformity in the foot. PEV; it is a disease characterized by an equinus of the ankle, varus of the calcaneus, adductus and inversion of the forefoot. Although many factors related to PEV etiology have been suggested, it is not fully known [1,2]. Congenital talipes equinovarus, also called clubfoot, is the most common congenital foot defect of the musculoskeletal system and is one of the most important causes of disability in children. Although PEV and developmental hip dislocation (DDH) are the most common musculoskeletal disorders in children, they are a preventable disease with early diagnosis. [3-6]. PEV deformity may appear unilateral or bilateral and its association with other congenital anomalies is common. Approximately half of the PEV cases are bilateral. In unilateral deformities, the right foot is more frequently

affected than the left foot. It is observed in boys 2 times more often than girls [7]. The theory of mechanical compression within the uterus of Hippocrates was considered the only reason for PEV etiology until the early twentieth century. However, studies have not found a significant relationship between PEV and diseases that increase intrauterine pressure, such as oligohydroamnios, multiple pregnancies, and overweight babies [8]. Although many factors such as genetic disorders, defects in embryological development, plasma germ cell defect in talus cartilage have been suggested in the etiology of PEV, its etiology is not known exactly today [9]. When we look at the literature, it was reported that the incidence of PEV varies between races and ethnic communities [10]. In this study, we aimed to share our knowledge about the incidence and etiology of PEV in our region in the light of the literature.

Materials and Methods

3658 patients hospitalized in the Neonatal Intensive Care Unit of our hospital between 2014-2017 were examined. Maternal age, birth weight, birth week, number of pregnancies and delivery type of patients with PEV deformity were obtained from hospital automation records. Mothers were divided into three categories

*Corresponding Author: Sezai Ozkan, Van Yuzuncu Yil University Faculty of Medicine Department of Orthopedics and Traumatology, Van, Turkey, E-mail: doktorsezai@hotmail.com

according to age: under 23, 23-34 and over 35. We divided the babies into two groups as 2500 grams or less depending on their birth weight. Mothers of newborns with PEV were classified into 3 groups according to the number of pregnancies. Ethics committee approval was received for the study by the Van YYÜ Medical School Non-Interventional Ethics Committee with the date and number 2019 / 19-12.

Statistical analysis

SPSS 19.0 software was used for statistical analysis. The demographic data of the patients were done by Fischer exact test and nonparametric Mann-Whitney U-test. Frequency tables were given for categorical variables and numerical variables were given as mean. $P < 0.05$ value was considered statistically significant.

Results

PEV deformity was diagnosed in 32 (0.87%) of 3658 babies treated in the neonatal intensive care unit over a period of 3 years. Of these 32 patients, 18 (56.25%) were bilateral, and 14 (43.75%) unilateral PEV. PEV deformity was diagnosed in 32 (0.87%) of 3658 babies treated in the neonatal intensive care unit over a period of 3 years. 19 (59.4%) of our patients with PEV deformity were male and 13 (40.6%) were female. Two (6.25%) of our patients had a positive family history. While 23 (71.87%) of our patients were delivered by cesarean, 9 (28.1%) of our patients were born normally. Considering birth weights, the number of babies born under 2500 grams was 7 (21.8%), and the number of babies born above 2500 grams was 25 (78.1%). Demographic data of our patients are given in Table 1 and descriptive statistical analyzes are given in Table 2. The image of a patient with bilateral pes equinovarus deformity is given in Figure 1.

Table 1. Demographic data of our patients

Phenotypic properties	Count (32)
Gender	
Male	19(%59,4)
Woman	13(%40,6)
Side	
Bilateral	18
Unilateral	14
Left	8
Right	6
Family history	
Yes	2
No	30



Figure 1. A patient with bilateral PEV

Table 1. Descriptive information of our patients

Demographic feature	Count (32)
Birth week	
<37	20
>37	12
Birth weight (gram)	
>2500	25
≤2500	7
Form of Delivery	
Caesarean	23
Normal birth	9
Maternal age	
<23	20
24-34	8
>35	
Number of pregnancy	6
<3	20
3-5	6
>6	

Discussion

Although the frequency of foot deformities in newborns is not fully known, 4.2% were reported in one study. While the most common foot deformities are metatarsus adductus; Other common foot deformities are PEV, calcaneo valgus, vertical talus [11]. PEV may be accompanied by deformity myelodysplasia, arthrogryposis or congenital anomalies, but it is commonly seen as idiopathic and isolated birth defects [12]. Multifactorial causes such as vascular disorders, environmental factors, intrauterine position, muscle insertion anomalies and genetic factors were considered in the

occurrence of PEV defect [13-16]. However, considering that the incidence of PEV is observed in 33% of twins and 25% of patients are familial, it is reported that genetic origin is more accused in PEV etiology [17]. While the probability of PEV deformity occurring in maternal twins is 32.5%, it has been reported as 2.9% in dizygotic twins [18,19]. Although foot deformity related to PEV disease is well defined, its etiology is still not fully elucidated. The basis of its etiology is heterogeneous and has been linked to genetic and environmental factors [20]. The incidence of PEV varies between populations and has been reported as 1 in 1000 live births in the literature [21]. In a study conducted in Chinese society, the incidence of PEV was reported as 0.39 per 1000 live births, 0.76 in the Spanish population, 1.12 in the white population, and 6.8 in the Polynesian population [22-24]. The incidence of PEV is 1 in every 1000 live births in the Vietnam population [20]. In addition, in a study conducted in the Netherlands [25], this rate was reported as 1.09 per 1000 live births, 1.4 in Sweden [26] and 1 in Romania [27]. In a study reported in Uganda, PEV deformity was reported as 1.2 per 1000 live births [28]. In a large-scale study in the Tuscany community between 1992 and 2011, the frequency of PEV was reported to be 1.56 per 1000 live births [29]. Haowang et al. Reported the incidence of PEV deformity in the European community as 1.13 per 1000 live births [30]. The incidence of PEV in our study was 0.87 per 1000 live births. The difference of our study from other studies was that it was performed in the neonatal intensive care unit. Since the additional congenital anomalies were more common in the neonatal intensive care unit and PEV was accompanied by these diseases, we expected our rates higher than the studies reported in the literature. In addition, in the neonatal intensive care unit where patients with extremity anomalies were examined and recorded by the orthopedic doctor immediately, this rate would be expected to be higher than the rates in the literature.

In our study, it was seen that PEV deformity was 1.4 times higher in males (59.4%) than females (40.6%). In previously reported studies, PEV genetic predisposition was found to be higher in men than in women [31,32]. In the study in Peru [33] the frequency of PEV was reported in men (61%) and women (39%). In a study conducted in the Netherlands this rate was found in men (66%) and women (34%), in Sweden, in men (72%) and women (28%) [25,26]. In Vietnam, the incidence of PEV was 64% in men and 33% in women [20]. When the data we obtained from our study are examined, the frequency of PEV is higher in men and these results are consistent with the literature. More than half of the patients with pes equinovarus are bilateral. Some studies have reported that bilateral involvement is around 50% and right foot and left foot involvement is equal [33]. In some studies, right foot involvement was found to be more than the left foot [34]. In a study it was observed that bilateral involvement (53%) was more than unilateral (47%), the right foot (49%) was more involved than the left foot (44%) [20]. In our study, it was observed that PEV was mostly bilateral deformity and right foot (57.1%) involvement was higher than left foot (42.9%). Although previous studies [35,36] reported that the incidence of PEV was higher in those with a birth weight below <2500 grams and in preterm deliveries (before 37 weeks), some studies have not reported this relationship [33].

Most studies have found an increased risk of CTEV for male sex. Relative risks of between 1.6 and 3.7 for males compared with females have been reported [37]. For isolated PEV, as in previous

authors, they have been found to have no relationship with increased birth weight or post-maturity, but the relationship with oligohydroamnios has been reported. Both maternal anemia and hyperemesis were found to be significantly associated with PEV [38,39].

Conclusion

In our study, no relation was found between birth weight, preterm delivery, and PEV incidence. It has been reported that there is a higher probability of developing PEV in maternal births under the age of 23 [33,34,40]. In our study, 4 PEV deformities were seen in those whose mothers were younger than 23, while there were 20 PEV deformities between the ages of 24-34 and contrary to some previous studies, no relation was established with the age of the mother. In this article, we examined the frequency of PEV in the neonatal intensive care unit in a tertiary care institution in the Eastern Anatolia region. Considering the data, we obtained at the end of the study, the incidence of PEV was lower than in the literature. In fact, our hypothesis before starting the study was that the incidence of PEV was higher in the neonatal intensive care unit. However, the results we obtained in our study were generally lower than the literature. We could not explain these results. Although the frequency of PEV is known to vary between populations and ethnic groups, our knowledge of the etiology of PEV is still insufficient and contradictory. We believe that future studies (such as genetic studies) will be more beneficial to science than classical knowledge about PEV etiology.

Conflict of interest

The authors declare that they have no competing interest.

Financial Disclosure

All authors declare no financial support.

Ethical approval

Van YYÜ Medical Faculty Non-Interventional Ethics Committee with the date and number 2019 / 19-12.

References

1. Waschak K, Radler C, Grill F. Der kongenitale Klumpfuß Congenital club foot. Z Orthop Unfall. 2009;147:241-62.
2. Minkowitz B, Finkelstein BI, Bleicher M. Percutaneous tendo-Achilles lengthening with a large-gauge needle: a modification of the Ponseti technique for correction of idiopathic clubfoot. J Foot Ankle Surg. 2004;43:263-5.
3. Göksan S, Bora: Doğuştan çarpık ayağın ponseti yöntemi ile tedavisi Acta ortopodica et Traumatol Turcica. 2002;36:281-7.
4. Dyer PJ, Davis N. The role of the Pirani scoring system in the management of club foot by the Ponseti method. J Bone Joint Surg Br. 2006;88:1082-4.
5. Penny JN. The neglected clubfoot. Techniques Orthop. 2005;20:153-66.
6. Sevimli R, Ceylan MF, Yıldırım E, et al. Is reassessment of radiographs taken from pediatric patients useful for detecting unrecognised hip dysplasia? East J Med. 2017;22:180-3.
7. Werler MM, Yazdy MM, Mitchell AA, et al. Descriptive epidemiology of idiopathic clubfoot. Am J Med Genet A. 2013;161A:1569-78.

8. Tachdjian's pediatric orthopaedics 3 ed, Editor: R. Lampert. W.B. Saunders Company 2002.
9. Staheli LT. Foot. In: Staheli LT, editor. Practice of pediatric orthopedics. Philadelphia: Lippincott Williams & Wilkins.2001;89–114.
10. Byron-Scott R, Sharpe P, Hasler C, et al, A South Australian population-based study of congenital talipes equinovarus. Paediatr Perinat Epidemiol 2005;19:227-37.
11. Widhe T. Foot deformities at birth: a longitudinal prospective study over a 16-year period. J Pediatr Orthop. 1997;17:20–4.
12. Wynne-Davies R. Family studies and the cause of congenital clubfoot, talipes equinovarus, talipes calcaneo-valgus and metatarsus varus. J Bone Joint Surg Br. 1964;46:445–63.
13. Hootnick DR, Levinsohn EM, Crider RJ, et al. Congenital arterialmalformations associated with clubfoot. A report of two cases. Clin Orthop Relat Res. 1982;167:160–3.
14. Dunn PM. Congenital postural deformities: perinatal associations. Proc R SocMed. 1972;65:735–8.
15. Bonnell J, Cruess RL. Anomalous insertion of the soleus muscle as a cause of fixed equinus deformity. A case report. J Bone Joint Surg Am. 1969;51:999–1000.
16. Gurnett CA, Boehm S, Connolly A, et al. Impact of congenital talipes equinovarus etiology on treatment outcomes. Dev Med Child Neurol. 2008;50:498–502.
17. Lochmiller C, Johnston D, Scott A, et al. Genetic epidemiology study of idiopathic talipes equinovarus. Am J Med Genet. 1998;79:90–6.
18. Cummings RJ, Davidson RS, Armstrong PF, et al. Congenital clubfoot. J Bone Joint Surg Am. 2002;84:290–308.
19. Tarraf YN, Carroll NC. Analysis of the components of residual deformity in clubfeet presenting for reoperation. J Pediatr Orthop. 1992;12:207–16.
20. Nguyen MC, Nhi HM, Nam VQ, et all. Descriptive epidemiology of clubfoot in Vietnam: a clinic-basedstudy. Iowa Orthop J. 2012;32:120-4.
21. Balasankar G, Luximon A, Al-Jumaily A. Current conservative management and classification of club foot: A review. J Pediatr Rehabil Med. 2016;9:257–64.
22. Ching GH, Chung CS, Nemecek RW. Genetic and epidemiological studies of clubfoot in Hawaii: ascertainment and incidence. Am J Hum Genet. 1969;21:566-80.
23. Chung CS, Nemecek RW, Larsen IJ, et al. Genetic and epidemiological studies of clubfoot in Hawaii. General and medical considerations. Hum Hered. 1969;19:321-42.
24. Moorthi RN, Hashmi SS, Langois P, et al. Idiopathic talipes equinovarus (ITEV) (clubfeet) in Texas. Am J MedGenet A. 2005;132:376-80.
25. Besselaar AT, Kamp MC, Reijman M, et al. Incidence of congenital idiopathic clubfoot in the Netherlands. J Pediatr Orthop B. 2018;27:563-7.
26. Wallander H, Hovelius L, Michaelsson K. Incidence of congenital clubfoot in Sweden. Acta Orthop. 2006 ;77:847-52.
27. McConnell L, Cosma D, Vasilescu D, et al. Descriptive epidemiology of clubfoot in Romania: a clinic-based study. Eur Rev Med Pharmacol Sci. 2016;20:220–4.
28. Mathias RG, Lule JK, Waiswa G, et all. Incidence of clubfoot in Uganda. Can J Public Health. 2010;101:341–4.
29. Seravalli V, Pierini A, Bianchi F, et al. Prevalence and prenatal ultrasound detection of clubfoot in a non-selected population: an analysis of 549, 931 births in Tuscany. J Matern Fetal Neonatal Med. 2015;28:2066–9.
30. Wang H, Barisic I, Loane M, et al. Congenital clubfoot in Europe: A population-based study. Am J Med Genet A. 2019;179:595–601.
31. Kancherla V, Romitti PA, Caspers KM, et al. Epidemiology of congenital idiopathic talipes equinovarus in Iowa. Am J Med Genet A. 2010;152A:1695–700.
32. Alderman BW, Takahashi ER, LeMier MK. Risk indicators for talipes equinovarus in Washington State. Epidemiology. 1991;2:289–92.
33. Palma M, Cook T, Segura J, et all. Descriptive epidemiology of clubfoot in Peru: a clinic-based study. Iowa Orthop J. 2013;33:167–71.
34. Parker SE, Mai CT, Strickland MJ, et al. Multistate study of the epidemiology of clubfoot. Birth Defects Res A Clin Mol Teratol. 2009;85:897–904.
35. Barker SL, Macnicol MF. Seasonal distribution of idiopathic congenital talipes equinovarus in Scotland. J Pediatr Orthop B. 2002;11:129-33.
36. Robertson WW Jr, Corbett D. Congenital clubfoot. Month of conception. Clin. Orthop Relat Res. 1997;:14-8.
37. Pryor G, Villar R, Ronen A, Scott P. Seasonal variation in the incidence of congenital talipes equinovarus. J Bone Joint Surg. 1991;73:632–4.
38. Limpaphayom M, Jirachaiprasit P. Factors related with the incidence of congenital clubfoot in Thai children. J Med Associati Thailand. 1983;68:1–5
39. Boneva R, Moore C, Botto L, et al. Nausea during pregnancy and congenital heart defects: a populationbased case-control study. Am J Epidemiol. 1999;149:717–25.
40. Loder RT, Drvaric DM, Carney B, et al. Lack of seasonal variation in idiopathic talipes equinovarus. J Bone Joint Surg Am. 2006;88:496-502.