

Etiology and Treatment of Congenital Vertical Talus: A Clinical Review

Seema Sehmi

ABSTRACT

Congenital vertical talus is a rare rigid flat foot deformity. Although the cause of the congenital vertical talus is heterogeneous, recent researches strongly support a genetic cause linking the genes expressed during early limb development. If remain untreated, it causes a lot of disability like pain and functional limitations. Traditional treatment for vertical talus involves extensive surgeries, which are associated with short and long complications. A minimally invasive approach involving serial manipulation and casting will produce excellent short-term results with regard to clinical and radiographic correction. To achieve correction without extensive surgery leading to more flexible and functional foot, a long-term research study is required.

Keywords: Clinical, Congenital, Limb, Talus.

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INTRODUCTION

The ankle joint complex involves articulation of talus with tibia and fibula.¹ The movements of the ankle joint are plantar flexion and dorsiflexion in sagittal plane and abduction and adduction in the coronal plane.² Talus also articulates with plantar calcaneonavicular ligament and calcaneus to form talocalcaneonavicular joint.³ Congenital vertical talus is a rare foot deformity, which is characterized by hindfoot valgus and equinus, with associated midfoot dorsiflexion and forefoot abduction caused by a fixed dorsal dislocation of the navicular bone on the head of the talus.⁴ It has rigid flatfoot or rocker bottom foot deformity with forefoot dorsiflexed and hindfoot plantar flexed.⁵⁻⁷ Congenital vertical talus was named first by Rocher⁸ in 1913 as foot in piole. Henken in 1914⁹ published the first clinical case of congenital vertical talus. Other terminologies used for this condition were pied plat valgus congenital, reversed club foot, vertical talus, and congenital convex pes valgus⁹⁻¹¹ and also named as congenital valgus flat foot and rocker bottom foot.^{12,13} Congenital convex pes valgus is the term, which is currently most commonly used.¹⁴ Its incidence is 1 in 100,000 live births with no sex predilection,^{15,16} but according to Marcinko and Schwartz,¹⁷ it is more common in males than females. About 50% of the children showed bilateral involvement.¹⁶ The exact cause of congenital vertical talus remains unidentified. It occurs either as an isolated deformity in approximately half of the cases and is associated with neuromuscular and genetic disorders in the remaining cases.¹⁸ The other causes may be due to arrest in prenatal development of foot, or sometimes, paralysis and contracture of the soft tissue may be the cause.¹⁹ The other parameters associated with congenital vertical talus are hypoxic birth injury, microcephaly, and cryptorchidism.²⁰ Without treatment, it leads to significant disability, including foot and ankle pain and medial plantar callus around prominent talar head.²¹ In early times, surgical treatment of congenital vertical talus was intensive with a large number of short- and long-term complications.²² Development of minimally invasive alternative approach has proved successful in providing correction and at the same time avoiding extensive soft tissue release.²³ The aim of the present study is to discuss various

Department of Anatomy, Sri Guru Ram Das Institute of Medical Sciences and Research, Vallah (Amritsar), Punjab, India

Corresponding Author: Seema Sehmi, Department of Anatomy, Sri Guru Ram Das Institute of Medical Sciences and Research, Vallah (Amritsar), Punjab, India, Phone: +91 9914754354, e-mail: drseema16@gmail.com

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causative factors, associated congenital anomalies, and various treatment approaches of congenital vertical talus after reviewing the available literature.

Epidemiology

Although the exact incidence of congenital vertical talus is unknown, the prevalence is estimated as 1 in 10,000 live births.¹⁶ The estimated number is low because of difficulty in recognition of this condition in newborn. Furthermore, the condition is rare and large studies are lacking in various populations to determine its laterality or gender predilection.

Pathoanatomy

Congenital vertical talus is a foot deformity in which calcaneus is in equinus and the talus is plantar flexed associated with rigid irreducible dislocation of talonavicular joint with navicular bone articulating on the dorsolateral aspect of talar neck. It is associated with systemic involvement. The head of the talus produces a prominence on the medial side, and a rocker bottom on the sole of the foot is seen. Congenital vertical talus should be differentiated from other deformities of the foot like club foot in which displacement of the talonavicular joint is directed downward and medially, whereas in congenital vertical talus, it is directed upward and laterally. Another important difference is that

the elevated heel is not fixed in congenital vertical talus, while it is fixed in club foot.²⁴ Coleman et al.⁷ gave the most widely used anatomical classification and described two types of congenital vertical talus. Type 1 deformity involves rigid dorsal dislocation of the talonavicular joint. In type II deformity, in addition to rigid dorsal dislocation of talonavicular joint, there occurs dislocation or subluxation of calcaneocuboid joint. Oblique talus is considered as milder form of congenital vertical talus based on clinical and radiographic examination. This condition requires treatment only in some cases despite its milder nature.⁶ The muscles and soft tissues involved in marked equinus and valgus deformity seen in congenital vertical talus are contracture of the Achilles tendon and involvement of posterolateral part of ankle and subtalar joint capsules.¹⁵ The midfoot and forefoot are dorsiflexed and abducted due to the contracture of tibialis anterior, extensor digitorum longus, extensor hallucis brevis, peroneus tertius, extensor hallucis longus tendon, and dorsal part of talonavicular capsules.¹³ The ankle joint remains in dorsiflexed position due to anterior subluxation of tibialis posterior and peroneus longus. The navicular bone lying on the dorsal and lateral aspects of the head of talus becomes hypoplastic and wedge shaped.²⁵ Talar head and neck are now medially directed and are abnormal in shape. Due to the vertical position of talus bone, plantar soft tissues including calcaneonavicular or spring ligament become weak causing rocker bottom appearance. The calcaneus bone is in extreme equinus position causing dorsolateral subluxation or dorsal dislocation of calcaneocuboid joint.¹⁵ The forced plantar flexion and forced dorsiflexion lateral radiographs will confirm the diagnosis and will differentiate congenital vertical talus from the oblique talus. The forced dorsiflexion lateral radiograph shows decreased tibioacalcaneal angle showing fixed hindfoot with persistent malalignment of the long axis of the talus in relation to the navicular bone. The forced plantar flexion lateral view depicts persistent malalignment of the long axis of the talus and first metatarsal bone¹⁵ and is known as talus axis and metatarsal bone axis (TAMBA).⁶ Also, associated underlying neuromuscular disease⁴ or flexion contracture or ulnar deviation of hand or fingers, multiple malformations, and CNS defects or chromosomal defects should be ruled out.⁷ It should be differentiated from the most common calcaneovalgus foot, posterior medial bowing of the tibia, and flexible flatfoot.¹⁵

Etiology and Associated Anomalies

The exact cause of congenital vertical talus is unknown. It is difficult to differentiate congenital vertical talus from other more common benign positional foot anomalies at the time of birth.⁴ To make definite diagnosis, the navicular bone should be dislocated dorsally on the neck of talus when the foot is maintained in extreme plantar flexion.^{6,7} Approximately half of the cases are idiopathic and half are associated with neuromuscular or genetic disorders. Congenital vertical talus occurs in both isolated and systemic forms. Therefore, in patients with apparently isolated forms of congenital vertical talus, it is important to rule out congenital vertical talus associated clinical features.^{26,27} Rarely, congenital vertical talus is seen as a nonsyndromic condition.⁴ Rather, it is associated with various syndromes like Cri du chat syndrome,²⁸ multiple pterygium syndrome,²⁹ muscle abnormalities like ischiocalcaneal band. The genetic conditions associated are Marfan syndrome and aneuploidy of chromosomes 13,15,18,^{30,31} De Barys Syndrome,¹⁵ fetal brain abnormalities like microcephaly and migration anomalies, spinal abnormalities, such as scoliosis, myelomeningocele, and tethered

cord,³² and Cri du Chat syndrome.²⁸ Stern et al.³³; Levinsohn et al.³⁴ gave transmission of congenital vertical talus by autosomal dominant trait with variable expression and incomplete penetrance. Genetic etiology of the congenital vertical talus is suspected in a child if any of these features are present. The features are various congenital anomalies, developmental delay and developmental regress, undeveloped secondary sexual characters, and ambiguous genitalia.³⁵ Other possible causes given are arrest in the rotational development of the foot in the second or third month of fetal life³⁶ or muscle imbalance³⁷ or increased intrauterine pressure and resultant tendon contractures or arrest in fetal development occurring 7th and 12th week of gestation¹⁵ or intrauterine growth retardation.³⁸ Incidence of IUGR in the general population was given between 3 and 7% by Romeo et al.³⁹ and was 80% by Muhsin.²⁰

Gene Sequencing

Cartilage-derived morphogenetic protein I (CDMP1)—Binding of CDMP1 with transforming growth factor-beta plays a role in the development of cartilage, joint, and growth of neuronal axons.^{40,41} Mutations in CDMP1 cause severe upper and lower skeletal malformation and ultimately congenital vertical talus in some families.^{41,42}

HOXD-II Gene

HOXD-II gene encodes a homeobox DNA-binding domain containing pro-HOXD II protein, which is involved in differentiation and limb development. Mutation in HOXD II gene is associated with isolated congenital vertical talus^{4,34,43,44} as well as bilateral congenital vertical talus and Marie–Tooth disease.^{45,46}

Gene Controlling Contractile Muscle Fibers

Muscle biopsy specimen from patients of congenital vertical talus showed small size of muscle fibers and abnormal fiber type.⁴⁷ Mutation in skeletal muscle contractile gene is common in distal limb defects like distal arthrogyposis⁴⁷ and congenital vertical talus.⁴⁸

18q Deletion Syndrome

The deletion of the distal part of the long arm of chromosome 18 is known to cause the 18q deletion syndrome. Variation in the clinical features of the patients with distal 18q deletion is due to differences in the size of the deletion.^{49,50}

TREATMENT

The main aim of the treatment is to resume the normal function of foot by correcting anatomical articulations between talus, navicular, and calcaneum bone. Commonly noninvasive treatment involves conservative treatment like manipulation and serial casting.⁴ Early casting for congenital vertical talus is sporadic with unexpected mixed results unlike in club foot where early caste treatment proves a success.⁵¹ But, this method is not successful in all the cases.^{52,53} Invasive procedures mostly involve single-stage surgery for required correction.^{54,55} Some surgeons initially do conservative plaster cast treatment to slowly reduce the talonavicular joint, followed by minimal surgical interventions giving early good results.^{4,56} Surgical treatment is always preferred before the age of 2 years.⁵⁷ Most surgeons follow open reduction of the talonavicular joint with tendon lengthening and release procedures. Others involve tendon transfers or total excision of navicular or talus.⁵⁸

If treatment gets delayed beyond 2 years of age, more aggressive procedures like navicular excision are required. Even older children require subtalar or triple arthrodesis as treatment.^{59,60} Most children before 2 years of age operated by extensive release with tendon lengthening and fixation procedures showed the average success rate as 71%.^{54,58,55} Extensive release with tendon transfer was the second most common surgical procedure, and the success rate was 74%.^{61,58} The other procedures like naviculectomy are not used in this age-group.^{12,62} The children older than 2 years of age showed best success rate of 73% with extensive release and tendon transfer,^{63,58} while extensive release with tendon lengthening and fixation procedures showed success rate of 46%.^{55,64} Naviculectomy with release procedures and tendon transfer was 73% at a mean age of 43.6 months at the time of surgery⁶⁵ and is 71% for subtalar/triple arthrodesis at a mean age of 75 months at surgery.⁷ Excision of the navicular bone was only done in five feet.⁶⁶

CONCLUSION

Congenital vertical talus is a rare foot deformity, which is misdiagnosed in newborn due to rigidity of foot deformity. A careful physical examination is needed as early recognition of the deformity leads to early treatment. Currently, the treatment involves serial manipulation and casting with minimally invasive approach. Additional research studies are needed to elucidate the etiology of congenital vertical talus, which will help the clinician to identify the congenital vertical talus at the time of presentation and to predict patient response to the treatment.

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