

CASE REPORT

Congenital bilateral agenesis of the tibialis anterior muscles: a rare case report

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Abstract

Congenital bilateral agenesis of the tibialis anterior muscles is a rare condition. We present a case of congenital absence of bilateral tibialis anterior muscles in a 6-year-old boy who presented with an abnormal gait. He was previously diagnosed to have bilateral congenital talipes equinovarus (CTEV) deformity for which he underwent corrective surgery two times. However, he still had a residual foot problem and claimed to have difficulty in walking. On examination, he walked with a high stepping gait and muscle power of both lower limbs was 5/5 on the medical research council scale (MRCS) except for both ankle dorsiflexors and long toe extensors. The sensation was intact. Magnetic Resonance Imaging (MRI) study of both legs revealed that tibialis anterior muscles were not visualized on both sides suggestive of agenesis of the tibialis anterior muscles. The rest of the muscles appeared mildly atrophied. The electrophysiological study showed normal motor and sensory conduction in both upper and lower limbs. Electromyographic (EMG) study of the vastus medialis was within normal limit and no response could be elicited for EMG of tibialis anterior muscles suggesting possible absence of tibialis anterior muscles, bilaterally. The patient underwent split tibialis posterior tendon transfer to achieve a balanced and functional foot and was well on discharge. The present case describes the normal anatomy and embryology of tibialis anterior muscles as well as possible causes of its agenesis along with its clinical implications.

Keywords: tibialis anterior, muscle, agenesis, anomaly, CTEV, foot drop.

Introduction

The tibialis anterior (TA) muscle originates from the lateral condyle and proximal two thirds of the lateral surface of the shaft of the tibia and the adjoining interosseous membrane [1]. The muscle descends with the distal portion in the form of a tendon in the lower part of the leg. It is inserted into the inferior and medial surface of the medial cuneiform and base of the 1st metatarsal bone [1]. Textbook of anatomy has reported the variation of insertion into the head of the 1st metatarsal, base of proximal phalanx of the hallux, talus and the extensor retinaculum [1]. The TA is innervated by the deep fibular nerve.

The TA overlaps the anterior tibial vessels and deep fibular nerve in the upper part of the leg [1]. Hence, any abnormality of the TA in the upper part of the leg may involve the neurovascular structures. Some of the abnormalities may be asymptomatic and remain undetected throughout life.

We hereby describe an interesting case of bilateral TA agenesis. It is a rare condition and the clinician should consider of such as one of the co-existing underlying etiology of CTEV and foot drop.

Patient, Methods and Results

We hereby report the case of 6-year-old boy who presented with the chief complaint of abnormal gait. His antenatal history was uneventful. The boy was born with emergency Lower Segment Caesarean Section (LSCS) due to transverse lie at 34 weeks of gestational age. His birth weight was 2.42 kg and he was diagnosed to have bilateral congenital talipes equinovarus (CTEV) for which he underwent first corrective surgery when he was 6-month-old and second corrective surgery was performed when he was 11-month-old. However, the deformities were not fully corrected and claimed to have difficulty in walking. His developmental milestones were normal.

On clinical examination, the patient walked with a high stepping gait. The anterolateral aspect of proximal part of the lower leg was small on both sides (Figures 1 and 2). The sensation (light touch, pinprick, temperature, proprioception, vibration) was intact in both the upper and lower limbs.

Power of ankle plantar flexor muscles was 5/5 on the medical research council scale (MRCS) bilaterally. Power of tibialis anterior muscles and toes extensors

was 0/5 and 3/5 respectively, on both sides. All reflexes were normal. Spine examination showed a dimple at the sacral region with no other spine deformities. Per-rectal examination was normal with no symptoms of bladder and bowel abnormalities. The initial provisional diagnosis was spina bifida occulta with L4–L5 motor neuropathy. Then, the patient proceeded with Magnetic Resonance Imaging (MRI) of lumbosacral spine and brain, which

revealed no abnormality. MRI of both legs revealed absence of tibialis anterior muscles (Figures 3–5). The electrophysiological study showed normal motor and sensory conduction on both upper limbs. Electromyographic (EMG) study of the vastus medialis muscles was within normal limit however, no response could be elicited for EMG of TA muscles suggesting possible absence of TA muscles bilaterally.

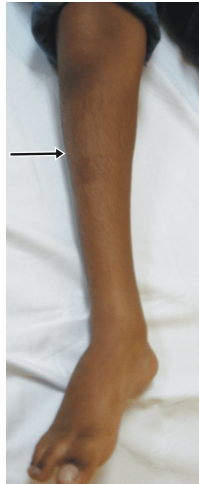


Figure 1 –
Photograph of right lower limb showing muscle wasting (arrow).



Figure 2 –
Photograph of left lower limb showing muscle wasting (arrow).

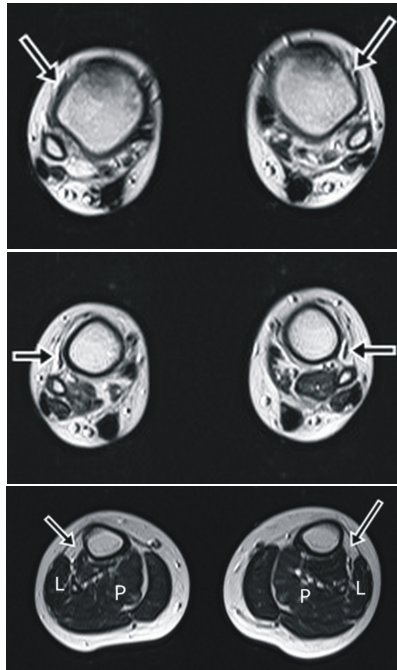


Figure 3 – Axial T2 weighted section of the ankle showing single anterior ankle tendons bilaterally (arrows).

Figure 4 – Axial T2 weighted section of the lower leg demonstrating atrophy of the anterior compartment muscles with a single thin common tendon bilaterally (arrows).

Figure 5 – Axial T2 weighted section of the mid calf demonstrating atrophic anterior compartment muscles (arrows) and normal lateral (L) and posterior (P) compartment muscles bilaterally.

Discussion

Variations of muscles have been reported in human beings. These variations include aberrant muscles, congenital absence or aplasia and anomalies regarding the origin or insertion. These anomalies are usually observed unilaterally and involve a single muscle or a related group of muscles [2]. One type of human muscle variation is the congenital absence or aplasia. Again, congenital absence may be total or partial [2]. According to the embryology textbooks, a single muscle may be wholly absent on one side of the body or there may be failure of development of only a part [3]. Considering such a fact, it is a rare variation that TA muscle agenesis may exist bilaterally.

The first indication of limb muscle development is observed during 7th week of intrauterine life [4]. The mesenchyme migrates into the limb buds to form the muscles. Consecutively, the connective tissue influences the pattern of muscle formation [4]. The limb buds elongate and the muscle splits into flexor and extensor components [4]. Thus, any defect during the 7th week of development may give rise to muscle anomalies.

Tesch NP *et al.* described the surgical implications and importance of tibialis anterior muscle anatomy in “Minimally invasive plate osteosynthesis of tibial fractures”. The process of percutaneous fixation for treatment of tibial fractures by using angular stable plates to be inserted between the TA and the periosteum is fast gaining popularity [5]. The TA muscle belly protects the neurovascular bundle. According to earlier researchers, the neurovascular bundle is protected in the

anterior compartment for a variable length [5]. The plates used for tibial fracture stabilization may be ranging in length from 15.6 cm (5 holes) – 31.6 cm (13 holes) which makes the neurovascular structures vulnerable to damage [5]. The 11-hole tibial fracture stabilization system, which extends for a length of 27.6 cm, may extend beyond the belly of the tibialis anterior muscle [2]. Thus, the location of origin and the length of fleshy component of the muscle may be anatomically important for the orthopedic surgeons.

An earlier study reported the case of bilateral agenesis of the tibialis anterior and unilateral aplasia of the extensor hallucis longus muscle in a 35-year-old woman who complained of pain in both legs while walking and difficulty in walking since childhood [6].

Abnormal variations regarding the insertion of TA muscle have been reported to be 21.7% [7]. Considering the fact that the TA muscle is a dorsiflexor and invertor of foot, any abnormality of TA may involve these two movements in the foot. Thus, congenital absence of TA muscles could contribute to the congenital talipes equinovarus (CTEV) deformity as seen in the present case. CTEV or clubfoot is a congenital deformity consisting of hindfoot equinus, hind foot varus, and forefoot varus. The deformity was described during the period of Hippocrates. The term talipes is derived from a contraction of the Latin words for ankle, talus, and foot, pes. The term refers to the gait of severely affected patients, who walked on their ankles [8, 9]. The exact cause of congenital clubfoot is unknown. Most infants who have clubfoot have no identifiable genetic, syndromal or extrinsic causes [10]. Extrinsic associations

include teratogenic agents (e.g., sodium aminopterin), oligohydramnios, and congenital constriction rings while the genetic associations include Mendelian inheritance (e.g., diastrophic dwarfism; autosomal recessive pattern of clubfoot inheritance) and cytogenetic abnormalities. It has been proposed that idiopathic CTEV in otherwise healthy infants is due to multifactorial system of inheritance [11].

Clubfoot associated with absence of TA muscle is a rare entity. Interestingly, earlier researchers found out the presence of abnormal arterial patterns in a limb with absence of the tibia and a contralateral limb with clubfoot and hence suggested that absent tibia and clubfoot may be etiologically related [12]. Histochemical examination of the muscle tissues in clubfoot showed no significant abnormalities but the researchers suggested that the muscle fibers in the affected calf were smaller in diameter [13]. Another subsequent histochemical study by Isaacs H *et al.* (1977) showed that hypotrophy, predominance of type-I fibers, and loss of direction and grouping of the fibers were common in patients with clubfoot. The same research also demonstrated electron-microscopic changes in the muscle and suggested that clubfoot may have a neurological basis [14].

The clinical symptoms and disabilities caused by aplastic muscles are influenced by their functions and physical activities. Small antero-lateral leg muscle bulk gives a clue of possible aplastic or dysplastic muscles, which could be due to neurological problems or primary muscle abnormality. Foot drop is one of the sequelae to absence of TA muscles. It can be associated with many conditions such as dorsiflexor muscle injuries, peripheral nerve injuries, stroke, neuropathies, drug toxicities or diabetes. Foot drop can be defined as a significant weakness of ankle and toe dorsiflexors, which include the tibialis anterior, extensor hallucis longus and extensor digitorum longus muscles. These muscles help the body clear the foot during swing phase and control plantar flexion of the foot on heel strike. Weakness in this group of muscles results in an equinovarus deformity.

Ultrasonography and especially magnetic resonance imaging (MRI) are helpful to understand and correlate symptoms and signs in patients with muscles agenesis.

Various anomalies of muscles have been reported causing clinical symptoms, or even being asymptomatic and often detected incidentally. In the present case, CTEV was corrected when the child was young. However, the patient still had the residual problem due to agenesis of the TA muscles bilaterally and walked with a high stepping gait for which he needed to wear bilateral hinged ankle foot orthoses. Patient underwent split tibialis posterior tendon transfer and achieved a balanced foot. He could ambulate well without orthoses

with energy efficient gait and rehabilitation outcome was satisfactory.

Conclusions

Bilateral congenital agenesis of the TA muscles is a very rare condition and it should be considered as one of the differential diagnoses for CTEV and foot drop. MRI of the lower limb muscles is strongly recommended in cases with similar presentation with normal brain and spine MRI so as to achieve prompt diagnosis and effective treatment.

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