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Research Article

Management of resistant congenital clubfeet by the new operative procedure (Hussain's procedure): An experience of a fellowship program

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ABSTRACT

Background and Objectives: Pakistan Society for Rehabilitation of the Disabled Orthopedic Hospital is renowned for the correction of orthopedic deformities including foot and ankle deformities. Consultant orthopedic surgeon Afzal Hussain, pioneers the treatment of orthopedic deformities and has developed a new operative technique for congenital clubfoot. This research was planned to report the success of Hussain's Procedure in detecting and managing the anomalous structures.

Material and Methods: This research was mixed retrospective and prospective research carried out during fellowship of first author in which, operative procedure of the resistant congenital clubfoot associated with anomalous structures viz. anomalous muscles and tarsal coalitions, were performed with the new operative procedure by Dr. Hussain, were assisted by the first author and a minimum of 5 years of follow up of the cases was done by the senior author (Afzal Hussain). Cumming's modification of Laavag and Ponseti score was applied for the calculation of the results at the follow-ups.

Results: During the fellowship, 10 cases of clubfeet with muscle anomalies were managed. Similarly 7 clubfeet with tarsal coalitions were managed. There was full correction of the clubfoot deformity. The cases were followed up by the senior author (Afzal Hussain) for at least 5 years. During the minimum of 5 years followup post completion of treatment, Cumming's modification of Laavag and Ponseti scores of the operated clubfeet were found to be excellent.

Conclusion: The resistance to management of congenital clubfoot by casting may be because of anomalous structures. New operative procedure by Consultant Afzal Hussain names as Hussain's Procedure was helpful in detecting and managing the anomalous structures.

Keywords: Anomalous muscles, congenital clubfoot, Ponseti score, Surgery, Tarsal coalition

INTRODUCTION

Pakistan Society for Rehabilitation of the Disabled Orthopedic Hospital is renowned for the correction of orthopedic deformities including foot and ankle deformities. Consultant orthopedic surgeon Afzal Hussain, pioneers the treatment of orthopedic deformities and has developed a new operative technique for congenital clubfoot. He gets referral of resistant and residual clubfoot from far and wide.

Working with the surgeon for 6 months, management and follow ups of rare anomalous structures in congenital clubfoot causing

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resistance to conservative management viz. anomalous muscles and tarsal coalitions was assisted by the author.

The anomalous muscles found in congenital clubfoot surgery are noteworthy. These structures may be the cause behind the rigidity of the deformity [1] and these also may effect on the further steps in surgery [2]. A tarsal coalition is a fibrous, cartilaginous or bony connection between two or more bones of the hind and mid foot. The tarsal coalition present in the clubfoot is usually not suspected preoperatively and can cause rigid deformity [3].

The operative management of the congenital clubfeet with anomalous structures viz. anomalous muscles and tarsal coalition was done by the new operative procedure and minimum of 5 years post-operative follow-ups were done.

This study could help in deciding for the management of resistant clubfoot which might have congenital anomalies in feet.

MATERIALS AND METHODS

A new method of defining the existing deformities in the clubfoot has been proposed by Dr. Hussain. This consists of recording the following existing deformities in the foot.

A dorsolateral hump
B1 half midfoot crease
B2 full midfoot crease
C equinus
D thin skin
E short first ray

Dorsolateral hump is the talar head which is palpable on dorsolateral aspect of clubfoot while midfoot crease is the crease on the sole. Record of whether the dorsolateral hump is reducible i.e. whether it disappears on abducting the foot with firm pressure over the bony prominence or it is rigid/irreducible is made. Reducibility of mid-foot crease i.e. whether it disappears on supinating first ray is also noted. Reducibility of equinus i. e. whether the foot can be brought into normal dorsiflexion by manipulation is also noted.

The Operative Procedure

The operation is tailored according to the existing/persisting deformities thus defined. Lateral release is done to reduce the rigid dorso-lateral hump; posterior and medial release is done in the presence of the rigid equinus, while abductor/planter release is done for rigid mid-foot crease. For example, when the clubfoot has rigid mid-foot crease and equinus, and reducible dorsolateral hump, the surgeon would opt for abductor-planter release and posterior-medial release Extreme caution is taken while elevating skin flaps in the presence of the thin skin.

The operation is done by 3 different incisions. The lateral release is done by Ollier's approach, posterior and medial release is done through a longitudinal incision and abductor/ planter release is done through a horizontal incision along the first metatarsal extending proximally to first metatarso-cuneiform joint.

Lateral release*:* A straight incision is given from a point 1cm below the lateral malleolus to the dorsolateral hump. Flaps of skin are elevated protecting the sural nerve. Release of the peroneal tendon sheath is done form the lateral border of foot to the superior peroneal retinaculum. This serves two other purposes: the peroneus longus tendon can then be mobilized and protected while releasing the inferior capsule of the calcaneocuboid joint; the lateral capsule of the subtalar joint lying beneath the peroneal tendon sheath can also be released.

Extensor digitorum brevis is elevated from its origin.

Lateral, superior, medial and inferior capsules of calcaneocuboid joint are released. Bifurcate and cubonavicular ligaments are also released.

The dorsal, lateral, medial and inferior capsules (along with the spring ligament) of the talonavicular joint are released under direct vision. Extreme care is taken to not to dissect in the neck of talus so as to preserve the vascularity of the bone. The extensor digitorum longus tendons and the dorsalis pedis artery and nerve are protected during the release.

Lateral capsule of the subtalar joint is released. Interosseous talocalcaneal ligaments are not released.

Postero-lateral part of the capsule of the subtalar joint along with the calcaneo-fibular ligament and the talofibular ligament are released.

Posterior and medial release: A longitudinal incision of 5-6 cm is given mid-way between medial malleolus and tendoachilles. The distal extent of the incision is curved medially just proximal to the insertion of tendoachilles.

Approach is made to the tendoachilles and the skin flap is elevated along with the sheath of the tendoachilles. This assures the adequate thickness of the skin flap- thus preventing flap necrosis. The sheath of the tendoachilles is sharply incised. Plantaris is released if present. Z-.lengthening of the tendoachilles is done.

Tibialis posterior and flexor digitorum longus tendon sheaths are exposed and sharply opened up. Release of the superficial deltoid ligament is done. Z-lengthening of tibialis posterior tendon is done. Z lengthening or tenotomy of the flexor digitorum longus tendon is done as required, depending upon the severity of the deformity. Flexor hallucis longus tendon sheath is opened up with a sharp incision. The sheath is opened up from the region above the ankle to the canal of flexor hallucis longus below the talus. The tendon is reflected along with the neurovascular bundle anteriorly. The dissection of the neurovascular bundle is not done. Ankle and subtalar joint capsulotomies are done. The location of the flexor hallucis longus tendon helps to identify the subtalar joint. In extreme equinus, the subtalar joint is released first, since the talus is wedged anteriorly due to equinus of calcaneus. The release of ankle joint is meticulously done. Z-lengthening of flexor hallucis is done. The deep part of deltoid is generally not released. The posterior one- third of the deep part of deltoid ligament is released in a most rigid clubfeet only.

Abductor and planter release: Abductor and planter release is done through a different medial longitudinal incision along the first metatarsal extending proximally to the metatarso-cuneiform joint. Structures released include the aponeurosis of abductor hallucis brevis, first metatarso-cuneiform joint and the planter fascia. It has been observed that the aponeurosis of abductor hallucis brevis lies at the planter aspect of the muscle in case of rigid cavus. Tibialis anterior tendon is identified and protected while releasing the first metatarso-cuneiform joint. The release of the planter fascia is done through the same medial longitudinal incision.

Reduction of the talonavicular joint is then done under direct vision. Talonavicular joint is fixed with a1.5mm K-wire if the reduction is unstable. Calcaneocuboid joint also needs fixation if the reduction is unstable.

The ends of flexor hallucis longus tendon is sutured with chromic catgut. The tendoachilles is sutured with the foot at 5 degrees dorsiflexion. Tibialis posterior and flexor digitorum longus tendons are realigned in the tendon sheaths and are not sutured.

The extensor digitorum brevis muscle origin is snugly repaired with chromic catgut.

The subcutaneous tissue and skin are meticulously closed with interrupted sutures.

Initial immobilization is done with an above knee back slab in cases with extensive release. If the release is not extensive, corrective above knee cast is applied.

Postoperative management:

Postoperative manipulation and casting is given importance, as this corrects the reducible deformities of the clubfoot which had not been corrected surgically. Removal of stitches along with manipulation is done under general anesthesia. Gentle manipulation was done in the presence of K-wire so that the joints which are not fixed by K-wire stretch out. K-wire is removed after 4 weeks. The cast is changed during these procedures

The next cast is applied after 4 weeks for non-rigid foot and after 2 weeks for the rigid one, which remains for 2 weeks. The final cast is then applied which remains for 2 more weeks. The foot remains in cast for a duration of 8-12 weeks after operation, depending upon its rigidity.

Ankle Foot Orthosis is then prescribed. The child wears AFO full time till he/she begins to walk. Then, the child wears the AFO at night or during afternoon naps till the age of 5-6 years. The manual exercises are also taught to the parents.

RESULTS

Operations of 10 cases of congenital clubfeet with anomalous muscles and 7 cases with tarsal coalition were assisted by the first author during the six-month period of the fellowship and a minimum of 5 years follow up was done by the senior author (Afzal Hussain). The cases had corrected well with the new operative procedure. Cumming's [4] modification of Laavag and Ponseti [5] scores of the operated clubfeet were calculated during the follow-ups and the results were found to be excellent in all of the cases.

The following cases of congenital clubfeet with anomalous muscles were managed by Hussain's procedure:

Master A, had accessory soleus in the left foot. He was operated for neglected congenital clubfoot.

Baby B had flexor digitorum accesorius longus muscle in his right foot. He was operated for resistant congenital clubfoot.

Baby C had flexor digitorum accesorius longus in his right foot. He had resistant congenital clubfoot.

Baby D had accessory soleus muscle along with the accessory head of abductor hallucis in his right foot. He had resistant congenital clubfoot.

Baby E had accessory soleus muscle in her right foot. She was operated for resistant congenital clubfoot.

Baby F, had accessory soleus muscle in her right foot. Other muscles were thick with short tendons. She also had talonavicular bar in the same foot, and also had a constriction ring in the right leg. Her foot was deemed syndromic

Baby G, who was operated for resistant congenital clubfoot and had bilateral clubfeet, had accessory soleus in his left foot.

Baby H with bilateral clubfeet, had accessory soleus muscle in his right foot. He was operated for resistant congenital clubfoot too.

Master I, who was operated for neglected congenital clubfoot, had accessory soleus muscle in his right foot.

Baby J, who was operated for resistant congenital clubfoot and had bilateral clubfoot deformity had agenesis of peronei in the right foot.

Thus, there were 7 cases of accessory soleus, 2 cases of flexor digitorum accessorius longus and one case of agenesis of peronei during clubfoot surgery. Among 10 cases, 3 had bilateral clubfeet

operations, and anomalies were present only in one side in the patients.

The anomalous muscles were released to correct the rigid deformities.

The following cases of congenital clubfeet with tarsal coalitions were managed during the 6 months period.

Baby A, female child, with bilateral congenital clubfoot, had the talonavicular bar on the right side. The right foot also had accessory hallucis muscle; muscles had thick bellies and short tendons. The child also had constriction rings.

Baby B, female child, with left congenital clubfoot had calcaneonavicular bar.

Baby C, female child, with multiple congenital contractures, had multiple and extensive tarsal coalitions on the right side.

Baby D, male child, with bilateral clubfoot was found to have the calcaneonavicular coalition on the left side.

Baby E, male child, with bilateral clubfoot deformity had the calcaneonavicular bar on the right side.

Baby F, male child, with bilateral congenital clubfeet, had the calcaneonavicular bar on the right side.

Baby G, male child, with left sided congenital clubfoot, found to have the calcaneonavicular bar during the surgery.

Among the above mentioned cases, two were deemed syndromic variety of clubfeet and rest were considered to be of idiopathic variety.

In all of the cases the cartilaginous coalitions were excised and interposition of fat at the site of resection was done.

The findings of the minimum of 5 years follow-ups of the cases were done by the senior author (Afzal Hussain).

Figure 1: Accessory soleus over the artery forceps. Distal cut end of tendoachilles is held with the Allis forceps

Figure 2: Accessory Soleus muscle

DISCUSSION

Wider use the Ponseti technique has improved the outcome of the non-operative treatment, but surgical treatment may be necessary in resistant or recurrent deformities [6]. While specific trends were reported and great variability exists in management of congenital clubfoot, certain principles are found to be universal: initial nonoperative management followed by surgery for persisting deformities [7]. Because there will probably always be patients with clubfoot deformity who are treated surgically, an operative plan that minimizes frequent or invasive surgical intervention may result in greater long-term success [8].

When considering surgery for CTEV, one must first determine what should be released. In the 1980s, McKay [9] and Simons [10] both reported success with aggressive, wide subtalar release.

More recent studies have indicated a return to a more limited release for CTEV [11].

Although some think that any surgery requires a comprehensive release of all soft tissues, Carroll [12], Bensahel et al [11], and Grant and Atar [13], among others, plan for and approach each case individually. In his classification scheme, Catterall [14] suggested what Grant and Atar [13] stated, "The surgeon should identify what failed in the conservative treatment" because these are the structures that need release."

Extensive surgical release may lead to decreased range of movement in the foot and ankle which compromises the functional result [15]. The essence of the new operation is the evaluation of the reducibility of the deformities in a congenital clubfoot. Only the rigid deformities are corrected by surgery and the reducible deformities are taken care by the post-operative serial casting.

By the new operative procedure, the structures causing resistance to conservative treatment could be pointed out. Sometimes the structures were anomalous ones, which are difficult to pick up with the classical Turco's Posteromedial release and the Cincinnati incision.

Clinically symptomatic muscle anomalies of the foot and ankle have been reported in the literature. Accessory soleus and accessory flexor digitorum longus muscles sometimes present with mass posterior to medial malleolus and have been implicated for tarsal tunnel syndrome [16, 17, 18]. Literature has documented the prevalence of the accessory flexor digitorum longus anywhere between 4 to 12 percent [16]. Accessory musculature is included in differential diagnosis of foot and ankle pain.16 The presence of accessory muscles can be diagnosed by MRI in symptomatic cases [3]. Excision of the accessory muscles has been reported to cause relief of symptoms of tarsal tunnel syndrome [16, 17, 18].

In very rare instances, anomalous muscles are present in a deformed foot. Some studies on clubfoot mention muscle anomalies as a possible cause of deformity and some do not [19]. Bonnell Cruess reported a case of bilateral accessory soleus muscles in a 9-year-old boy with a resultant fixed equinus deformity in the feet, which had been present since birth [20]. Ger and Sedlin suggested that the accessory soleus muscle can be responsible for producing a deformity, and their case had a tendency to inversion [21]. Grogono and Jowsey reported a case of bilateral clubfoot associated with a flexor digitorum accessorius longus. The anomaly was found only in the left foot. They warned, though, that this muscle may have been overlooked in the right foot. They believed that this anomalous muscle may have contributed to the persistence of the foot deformity [22].

M Del Sol et al. found two cases of accessory soleus muscles in 254 dissected legs (0.8%), one of which had an equinovarus deformity. No flexor digitorum accessorius longus was observed [23]. Turco identified anomalous muscles in about 15% of his patients with clubfoot. These included flexor accessories longus muscle in 16 feet (6.6% of the cases). The flexor accessories longus muscle attached to the calcaneus or to the intrinsic flexors of the toe. In three of the feet, the muscle was so well developed that it was transferred to the tendoachilles. He also noted a well developed plantaris muscle in two of the feet.Turco also noted absence of posterior tibial tendon in eight feet in five patients. In the three patients with bilateral club foot, the posterior tibial tendon was absent on both sides [2].

Sodre, et al observed muscle anomalies in 11 of 73 patients (15.3%) with talipes equinovarus treated by them. Among these anomalies, the authors found six patients (8.3%) with an accessory soleus muscle, four patients (5.6%) with a flexor digitorum accessorius longus and one patient (1.4%) with agenesis of the posterior

tibial muscle. In the patients with an accessory soleus muscle, a tenotomy of the anomalous muscle was performed and part of its tendon, about an inch, was resected. In the patients with a flexor digitorum accessorius longus the anomalous muscle was resected from its insertion on the medial aspect of the calcaneus [19].

In a study done by MB Dobbs et al, flexor digitorum accessorius longus muscle was identified in 55 (6.6%) of the 835 patients at the time of surgical correction of the clubfoot deformity. It was present in 4.5% of patients without a family history (33/741) and 23.4% of patients with a family history (22/94) ($P < 0.0001$). Children with first-degree relatives with clubfoot are 6.6 times more likely to have the anomalous flexor muscle than children without first-degree relatives with clubfoot [24]. Porter described an anomalous flexor muscle in calf of five children with clubfoot. He also observed that patients with this anomalous muscle had a greater frequency of first-degree relatives with clubfoot [25].

Chotigavanichaya et al. reported the case of a patient in whom clubfoot could be corrected only after release of an accessory soleus muscle [1]. Kishta et al published a report 16 case of accessory soleus muscle as a cause of persistent equinus in clubfeet treated by the Ponseti method [26]. Danielson et al. [27] and Chittarajan et al. [28] also published case reports about clubfeet with accessory soleus muscles.

Callahan reported an anomaly in a clubfoot of a 14-month old boy associated with an absent posterior tibial tendon and a tarsal coalition [29]. Occurrence of previously undescribed muscles found during clubfoot surgery have been noted in literature [30].

Dr. Afzal Hussain, who does high volume surgery for congenital clubfoot referred for resistant to casting and the new operative procedure was useful in picking the structures which caused resistance to conservative treatment and in these cases happened to be the anomalous muscles. The anomalous muscles would be difficult to recognize in clubfoot surgery done by typical Cincinnati incisions and also by minimal incisions.

Anomalous muscles may play an important role in equinovarus deformity, depending on their insertion and dynamic action. The muscle anomalies should be identified during clubfoot surgery and released to prevent residual deformities. If the diagnosis can be made by MRI prior to surgery, a small incision to release these muscles from the calcaneus may be enough to correct the deformity of the hindfoot and to prevent major surgery [19].

The cause of the tarsal coalition is almost irrefutably a failure of the primitive mesenchyme to segment by cleavage in a 27-72 mm fetus and thus produce the normal peritalar joint complex. Tarsal coalition generally presents as painful flatfoot in children aged 8 to 16 years [31]. Tarsal coalitions have been noted to occur with other disorders including fibular hemimelia, Apert's syndrome, Niert Pearlman syndrome and also with clubfoot [32].

Turco noted talocalcaneal coalition in eight feet in five children. In all the cases, the bar was in the region of the sustentaculum tali. In six of the feet, the coalition was cartilaginous and in the other two, it was osseous [2]. Khan reported about finding the tarsal coalition in a clubfeet which failed to correct by Ponseti technique of casting [33].

Spero et al. reported 18 cases of tarsal coalition in rigid equinocavovarus feet. 16 cases were found in surgery and 2 at morbid dissection. These 18 feet were of the 14 patients. 6 of them were deemed teratologic due to the presence of other pathological conditions and 8 were considered congenital.. 4 of the cases had bilateral clubfoot.

The majority of the cases showed cartilaginous subtalar coalition in the medial facet [34].

In 22% of the revised clubfoot, Atar et al found the talocalcaneal bars (bony or cartilaginous) that might contribute to the recurrence of the deformity. The bars might be iatrogenic (injury to the subtalar joint during a previous surgery) or might have been overlooked during the previous procedures [35].

Clubfeet with the tarsal coalition are rigid, and its presence of the coalition is difficult to determine preoperatively. Even during the operation it may be difficult to recognize the tarsal coalition.34 The tarsal coalitions are especially difficult to pick up by the typical PMR surgery by hockey-stick incision. The new operative procedure: Hussain's Procedure developed by Dr. Hussain would be of help. All of the joints are released under direct vision in this procedure. The tarsal coalitions would be recognized more easily and further management could be done at the same operation.

CONCLUSION

The resistance to management of congenital clubfoot by casting may be because of anomalous structures. New operative procedure named Hussain's Procedure by Dr. Afzal Hussain was helpful in detecting and managing anomalous structures in resistant, residual and neglected congenital clubfeet and the results were found to be excellent.

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