**Abstract**

Background: Variations in pedal circulation in congenital talipesequinovarus (CTEV) are well documented. There is a reported risk of vascular injury to the posterior tibial artery (PTA) during operative procedures for CTEV, potentially leading to necrosis and amputation. The aim of this systematic review was to identify the most common anomalies in arterial pedal circulation in CTEV and to determine the relevance of these to clinical practice.

Methods: The systematic review was registered on PROSPERO and was carried out according to PRISMA guidelines by two independent reviewers. Studies that examined pedal circulation in idiopathic CTEV were included. Papers that studied non-idiopathic CTEV and those not published in English were excluded. Data extracted included patient demographics, imaging modalities and findings.

Results: A total of fourteen papers satisfied the inclusion criteria, including 192 patients (279 clubfeet), aged 0 – 13.5 years, at various stages in their treatment. Imaging modalities included arteriography (n=5), duplex ultrasound (n=5), magnetic resonance angiography (n=2), and direct visualisation intra-operatively (n=2). The dorsalis pedis (DP) was most frequently reported as absent (21.5%), and the anterior tibial artery (ATA) was most frequently reported as hypoplastic (18.3%). Where reported (n=36 feet), 61% of patients were noted to have a dominant supply from the PTA.

Conclusions: The most common variation in pedal circulation in CTEV is diminished supply from ATA and DP, although there are documented anomalies in all of the vessels supplying the foot. We therefore recommend routine Doppler ultrasound imaging prior to operative intervention in CTEV.

**Introduction**

Variation and insufficiency in the pedal circulation in CTEV has been acknowledged since 1910, with early arteriography demonstrating disparities in anatomy [1].

In health, pedal circulation is provided by three arteries – anterior tibial, posterior tibial, and the peroneal arteries, anastomosing to form the plantar arch. Vascular abnormalities occur frequently in the normal population [2], and a proportion of patients with CTEV have normal vascularity of the lower limb [3].

The most frequently performed procedure for CTEV is percutaneous Achilles Tenotomy (pAT) as part of the Ponseti method with serial casting. The technique for pAT has been described in detail, mostly by papers reporting complications of pAT, including bleeding, and pseudoaneurysm formation [4, 5].

Although the Ponseti correction is the most popular treatment method, corrective surgery is still performed in numerous centres around the world for primary CTEV and for relapsed cases. In a recent systematic review of the literature the reported surgery rate for CTEV relapse was up to 53% [6]. In the Turco approach, a posteromedial incision is made [6]. Dissection is carried down through subcutaneous tissues and the neurovascular bundle identified and dissected along its course and retracted with a vessel loop. The significant tissue dissection could pose a risk to unanticipated pedal vasculature. The Cincinnati approach for soft tissue release as well as the approaches used for corrective osteotomy carry a similar risk to this neurovascular bundle..

Research into variations in pedal circulation in CTEV have had contradictory findings [7, 8], and case reports have been published detailing anomalies in anterior tibial arteries, posterior tibial arteries and the dorsalis pedis. The largest study to date has encompassed 50 patients, with 74 clubfeet [9].

The aim of this review is to describe the pattern of pedal circulation, and most common aberrancies in CTEV, and to determine the relevance to clinical practice.

**Method**

***Search Strategy***

A systematic review was carried out according to Preferred Reporting Items for Systematic Reviews and Meta Analyses (PRISMA) guidelines. An online literature search was carried out on PubMed, Embase, Medline, and CINAHL from their inception to the search date of 1 May 2018. The systematic review was prospectively registered on PROSPERO (CRD42018103788).

All search strategy terms are outlined in Table 1 of the appendix. In addition, searches of the grey literature were carried out through Google Scholar, Web of Science and through clinical trials databases including clinicaltrials.gov, and WHO clinical trials. The reference list for each eligible text was reviewed for potentially eligible articles.

***Eligibility Criteria & Identification***

The systematic review included any paper reporting on arterial pedal circulation in patients with idiopathic CTEV. Any imaging or assessment modality, performed at any age, was included. Papers documenting non-idiopathic CTEV and papers written in languages other than English were excluded.

Two reviewers conducted screening of titles and abstracts independently (AP, KH), and any conflicts were resolved in consensus, or with a senior reviewer (BP) if necessary. Eligible texts were read and reviewed in full by both reviewers.

***Data extraction and appraisal***

Two reviewers (AP and KH) extracted data relevant for the systematic review with the use of a data collection table. Extracted data is demonstrated in Table 1.

***Bias assessment***

Bias assessment was carried out using a modified bias assessment tool developed by Murad *et al.* [10] *-* papers were scored according to domains of selection, ascertainment, and reporting. This is demonstrated in Table 2 of the appendix.

**Results**

The initial literature search identified 480 papers, after screening and assessment of full text eligibility, a total of fourteen papers were identified [3, 7-9, 11-20] (Figure 1). Patients ranged in age from newborn to 60 years old, the patient aged 60 [18] was excluded from the subsequent analysis resulting in 192 patients aged 0 – 13.5 years (Table 2).

Imaging modalities were described in all papers; five papers used arteriography [3, 7, 11, 14, 15], five utilised duplex ultrasounds [9, 13, 16, 19, 20], one via non-invasive 3D non-contrast magnetic resonance angiography (MRA) [18], one via contrast-enhanced MRA [17, 18], and two via direct intraoperative visualisation [8, 12]. 222 feet were imaged with duplex ultrasound, 39 via arteriography, and the remainder via MRA or intraoperative visualisation.

Pedal vessels were categorised as absent, hypoplastic or aberrant. There was a total of 198 variances noted in the 279 feet (70.2%), and ATA was most commonly reported to have variances (Table 3). When reported 61% of feet (n=36), were reported to have a dominant supply to the pedal circulation from the PTA, one foot was reported to have a dominant supply from the peroneal artery.

With both duplex ultrasound and arteriography (Figure 2 and 3), there were mostly variations in ATA (23% and 79.5% respectively), and DP (25.7 and 64.1%). Arteriography reported more variation in ATA (79.5%) compared with DP (64.1%), additionally 30.8% of feet (n=12) were reported to have variations in the plantar arch.

There were only two reports of patients experiencing complications secondary to arteriography – contrast allergy, and rash [11, 14].

**Discussion**

Clubfoot is a common congenital malformation. This systematic review suggests that up to 70% of the feet have anomalous vascular supplies. This is in line with previous papers which have estimated up to 89% of patients with CTEV may have absences or anomalies in the vascular supply to the foot [3].

Both angiography and duplex ultrasound demonstrate variances of the anterior tibial trunk, however there are fewer variations highlighted by duplex ultrasound compared with arteriography. This could be due to the greater sensitivity of angiography in visualising aberrant vessels. The ATA was reported as abnormal in 23% in Doppler vs 80% in arteriography. Limitations of duplex ultrasound include operator dependency, as well as the aforementioned false positive results attributed to collateral formation. Identification of arterial anomalies in the adult population is generally initially performed by duplex ultrasound, followed by arteriography and MRA if necessary. There are limited studies comparing imaging modalities when visualising peripheral arteries in the paediatric population, and studies focus on iatrogenic vascular injuries after cardiac catherisation [21]. In this emergency setting, duplex ultrasound has been deemed to be safe, and also effective at identifying vascular injury, and an appropriate alternative to more invasive arteriography [21]. However, Sodre *et al.* demonstrated no correlation between continuous wave-form Doppler and arteriography, and postulated that this may be because wave-form Doppler may not differentiate between major vessels and collateral vessels and thus produce a signal where arteriography demonstrated no vessel [3]. Creider *et al*. went on to suggest that colour duplex ultrasound would be more effective than wave-form Doppler, and supported this as the most acceptable method of perioperative evaluation of the clubfoot [22].

Where absence or diminished supply exists from the anterior tibial trunk, there is usually a univascular arterial supply of the foot. This is supported by evidence that suggest that in 61% of imaged patients the dominant supply is from the PTA.

For most patients with CTEV treated using the Ponseti method, tenotomy is the only procedure required in the course of treatment. **The Ponseti method is the effective and the preferred initial treatment and first line initial modality for relapsed deformity.** However, various surgical procedures are still being performed (6) carrying a risk of serious vascular injury and complications. Isolated case reports in the literature reflect this (Table 4) [4, 5, 23-25].

Dobbs et al. reported on four patients out of 219 feet (2%) having a serious bleeding complication following pAT with bleeding presumably due to injury of the peroneal vessels [4]. Another paper reported development of a pseudoaneurysm in one patient after undergoing pAT at the age of eight weeks [5] with damage to the neurovascular bundle necessitating open exploration and ligation of the artery.

The utmost care to preserve the neurovasculature during any operative procedure is taken, however the knowledge that necrosis could lead to amputation even following posteromedial release [23] affects the clinician’s perception and decision making. Where these complications have been reported it has not been possible to identify the vessel that has been damaged, although it has been postulated that extensive necrosis can occur in areas supplied by the dorsalis pedis and the anterior tibial artery and they have suggested a framework already for operative treatment of clubfoot [24].

This study has reinforced the knowledge that in CTEV, the foot has abnormal arterial anatomy in the majority of cases. **Surgical approach should be used only after rigorous scrutiny and when required, it should judiciously preserve the soft tissue from increased scarring and, preserve neurovascular bundle carefully.**

Therefore, careful pre-operative planning is needed when surgical intervention is planned. When planning for a soft tissue release and bony surgery, current practice guidelines should be reviewed, and we would suggest considering a colour duplex ultrasonography prior to operative procedure in conjunction with a thorough clinical examination (Table 5). CT angiogram or MRA imaging may be useful in delineating the anatomy if duplex is felt to be inadequate.

MRA confers the advantage of not subjecting the child to ionising radiation, as well as providing added information about soft tissue structures within the foot [18], which may be of benefit in preoperative planning. It has been demonstrated that CT angiography is an appropriate modality for identifying abnormalities where they exist. If CT angiography is to be employed, a discussion must be had with the patient’s family regarding the risks posed with CT angiography in terms of radiation and contrast allergy against the benefit conferred by delineating the anatomy and thereby mitigating against vascular injury. Where a child is unable to remain still then the risk of sedation also needs to be considered.

We acknowledge that routine preoperative imaging may not always be feasible in less economically developed countries where the Ponseti method has been readily adopted due to its low cost and ease of reproducibility. These finding further emphasise the importance of the Ponseti method in treating primary and relapsed CTEV before considering any surgical intervention.

Limitations for this systematic review include the relatively small number of studies that have been done in to arterial abnormalities in CTEV, and that all comprise either case reports, or cohort studies. Further research needs to be done on this topic, as this paper’s findings are based only on 14 eligible papers. There are only a few documented complications but they may be under-reported and when they occur, catastrophic.

Ideally prospective studies should be undertaken, to correlate different imaging modality findings, and in the CTEV population, although it must be acknowledged that such work would be challenging to perform. Further work needs to be done into evaluating the usefulness of MRA.

**Table Legend**

Table 1: Extracted Data

Table 2: Literature review of variations in pedal circulation in clubfoot

Table 3: Overall variations in pedal circulation (n=279 feet)

Table 4: Reported complications in the literature

Table 5: Suggested preoperative clinical evaluation of pedal circulation in CTEV

**Figure Legend**

Figure 1: Preferred Reporting Items for Systematic Reviews and Meta-Analyses ﬂow diagram of data selection process

Figure 2: Variations in pedal circulation when imaged by arteriography (n=57 feet)

Figure 3: Variations in pedal circulation when imaged by Doppler (n=222 feet)

**Appendix Legend**

Appendix Table 1: Search Terms used for MEDLINE (via OVID)

Appendix Table 2: Bias Assessment

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