

**Final report for the Rural Research Capacity Building
Programme (2010 intake)**

CLUBFOOT IN THE BUSH

The management and outcomes of infants who received their initial Ponseti casting for idiopathic congenital clubfoot in a regional centre compared to those treated exclusively at a tertiary Children's Hospital.



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List of Abbreviations

| | |
|---------|--|
| CHHC | Coffs Harbour Health Campus |
| CHW | The Children's Hospital at Westmead |
| CTEV | Congenital talipes equinovarus |
| DSI | Clubfoot Disease Specific Instrument |
| IPTAAS | Isolated Patients Travel and Accommodation Assistance Scheme |
| IQR | Inter quartile range |
| NSW | New South Wales |
| PROM DF | Post-operative passive range of motion of dorsiflexion |

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Abstract

Aim

This project aims to evaluate the outcomes of infants who receive initial Ponseti casting in Coffs Harbour, to a group of metropolitan infants who received their initial casting at The Children's Hospital, Westmead (CHW).

Methods

The study is a retrospective medical record audit. Fourteen infants have received initial Ponseti management at CHHC. A stratified recruitment strategy was used to identify 37 subjects from CHW; matching for age, gender, unilateral/bilateral presentation, and severity of initial presentation. Outcome measures were: the number of casts, age at Achilles tendon (TA) release, post-operative range of passive dorsiflexion (PROM DF), splinting prescription, compliance with splinting regime, the need for further casting or surgery, Dimeglio score, and the Clubfoot Disease Specific Instrument (DSI).

Results

Analysis of initial Pirani scores shows the groups were matched. Comparison of outcome measures between the two groups was unable to detect *any* statistically significant difference.

Median number of casts was six for both groups. Median age for TA release was 7.6 weeks at CHHC, 7.7 weeks at CHW. Median PROM DF was 18.5^o at CHHC, 25^o at CHW. Median Dimeglio score was 3 at both sites. Median DSI score was 11 at CHHC, 13 at CHW.

Splinting was prescribed in 100% of both groups. Both groups experienced some non- and partial compliance, but there was no significant difference detected between the two groups. There was higher need for further surgery in the CHHC group, and for further casting in the CHW group. However, these differences were not statistically significant.

Conclusions

This study was unable to detect a significant difference in clinical outcome between the CHHC and CHW groups. This result supports the shared care model of Ponseti management of clubfoot, as offered in Coffs Harbour. Clinical equivalence is difficult to prove, hence the study has been extended to a total of 5 years, to increase participants and gain stronger evidence.

Further work is necessary to identify which regional sites within NSW should be able to offer initial Ponseti management.

Executive Summary

Ponseti Management of Idiopathic Congenital Clubfoot

Ponseti management has become gold standard for the treatment of idiopathic congenital clubfoot world-wide. Initial Ponseti management involves weekly casting of a very young infant, usually followed by a percutaneous tenotomy of the Achilles tendon. The child is then required to wear splinting, decreasing over time, until four years of age.

The Clubfoot in the Bush study has shown that a shared-care model of Ponseti management can deliver clinical outcomes similar to that achieved at a tertiary children's hospital. Ponseti shared-care is currently offered at several regional sites throughout New South Wales (NSW), but clinical outcomes have not been formally evaluated.

The Shared-Care Model of Ponseti Management in NSW

In the shared care model of Ponseti management infants receive the initial casting phase of management locally, but travel to a tertiary children's hospital for tenotomy. The splinting phase can then also be managed regionally, with only occasional tertiary reviews necessary. This model of care saves families a significant amount of travel with a new baby. Such repetitive travel has inherent social, emotional and financial costs to families.

At many other regional NSW venues shared Ponseti management is not offered. Families are required to travel weekly to a tertiary children's hospital in a metropolitan centre for treatment. This is not equitable access to appropriate health care for regional families. There is also a significant cost to the health service for the travel under this model of care, as the Isolated Patients Travel and Accommodation Assistance Scheme (IPTAAS) subsidises regional families with the financial cost of travel for health care.

Compliance with the post-tenotomy splinting regime has been shown to be a major predictor of long term outcome for clubfoot. Continued long-distance travel to a tertiary children's hospital for review and monitoring is likely to have an increased drop-out rate for follow-up, with subsequent early cessation of splinting. Hence, clinically, there is a strong preference for local management. However, the regional clinician offering Ponseti management needs to be skilled in both the initial casting phase and the splint maintenance phase of management.

Not all regional venues have the skill base or experience to be offering Ponseti management for infants born with idiopathic clubfoot. In smaller health facilities that are likely to only see an occasional case of clubfoot, it is unlikely that this can

maintain an appropriate skill level. This level of incidence necessary to maintain skills has not been defined, and requires some investigation.

The Extent of the Problem

Utilising data from the most recent NSW Mothers and Babies Report and the NSW Admitted Patient Data Collection, it is possible to estimate there are approximately 70 infants with clubfoot born regionally in NSW each year. Some of these infants could receive Ponseti management under a shared care model.

Recommendations

This study indicates that the shared care model of Ponseti management could be effective throughout regional NSW. The model of care has been shown to have good results in one regional centre. It now needs to be investigated over several regional centres that offer Ponseti management. This will ensure that this is the best model of care for New South Wales Ministry of Health to pursue. The Ministry is then in a position to offer standardised care across the whole state.

If established that this is an appropriate model of care across several sites, then each of the regional sites that are capable of offering Ponseti management need to be identified. If there are large regional centres that are currently unable to offer Ponseti management, then this needs to be addressed. The NSW CTEV Network Working Party (chaired by Ms Alison Chivers, physiotherapist at the Children's Hospital, Westmead) has begun to address this issue with the development of an E-learning module. Unfortunately the working party has no ongoing funding for the implementation, evaluation and review of the module.

E-learning will not be able to provide all of the training necessary to begin Ponseti management of congenital clubfoot. Inherently there is a strong practical component to the casting, as well as the management of the splinting phase. Hence the training of potential regional clinicians still requires some planning and funding.

Smaller regional centres will not carry a clubfoot caseload that can sustain the skills necessary for gold standard management. How large a case load is necessary needs some investigation.

In these small centres, resources need to be allocated to assisting families, especially in the early intensive casting phase, to attend a neighbouring centre that is able to offer Ponseti management, or a tertiary children's hospital in a metropolitan area.

Fulfilling these recommendations will enable NSW to offer the gold standard of treatment for clubfoot, in a co-ordinated fashion that does not disadvantage regional families.

The Current Study

The Clubfoot in the Bush Study compared the outcomes of infants who received their initial Ponseti casting in Coffs Harbour (CHHC), to those who received their initial casting at the Children's Hospital at Westmead (CHW). The study was a retrospective medical record audit.

All 14 of the children who have been born with clubfoot in the Coffs Harbour region since the change to Ponseti management in 2006, were included in the study. A stratified recruitment strategy was used to select 37 infants from CHW that received initial Ponseti management over the same period.

A total of nine clinical outcome measures were used to compare the two groups. Statistical analyses were unable to detect a significant difference in **any one** of the outcome measures utilised, indicating that receiving the initial casting phase of Ponseti management in a regional centre can be as effective as receiving all treatment in a metropolitan tertiary centre.

The study has ethics approval to continue for a total of five years. However, these interim results indicate that it is probably already time to involve several regional centres, to begin to define which centres are offering initial Ponseti management, as well as which centres should be offering Ponseti management.

Background

Idiopathic congenital clubfoot, also known as congenital talipes equinovarus, is a relatively common congenital orthopaedic condition, affecting 1-2/1000 live newborns. The incidence varies between cultural groups, it is reported as high as 6.8/1000 in Polynesian populations (1).

Clubfoot has four components: equinus, heel varus, forefoot adduction and cavus. Severity of the deformity can vary considerably, from mild deformity to a foot that is stiff and resistant to correction. Left untreated clubfoot results in long term functional disability, deformity and pain (2). Children with untreated clubfoot walk on the lateral side of the foot. They are unable to wear usual footwear, and have significant limitations in mobility. The aetiology of clubfoot is thought to be multifactorial - involving both genetic and environmental factors. It is often identifiable in utero.

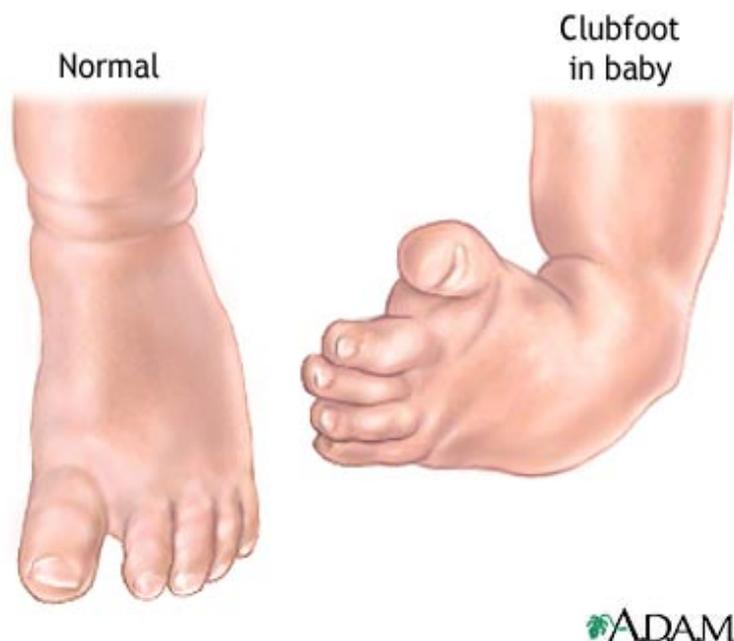


Figure 1: Comparison of clubfoot and a normal foot in an infant (3)

Treatment of Clubfoot

During the 20th century there was a gradual move away from joint invasive surgery in the initial treatment of idiopathic congenital clubfoot. Studies began to support the use of conservative management in preference to surgery (4-9). The three conservative methods of treatment most commonly reported are the Ponseti method (2), Kite's Method (10) and the French Functional (Physiotherapy) Method (7).

Kite's method of manipulation and casting gained popularity, but was found to have inferior results to that of the Ponseti Method (11, 12). The French Method is still used in some centres today. This method requires daily manipulations of the

newborn clubfoot, maintaining the new position with taping. Daily treatment is continued for the first two months, then three times a week until the six months of age. The taping allows some movement of the foot, the aim being to encourage peroneal muscle strengthening as a way of maintaining long-term correction. Once appropriate alignment is achieved, night splints are worn until walking age. The success of the French Method is reported as equivalent or marginally inferior to the Ponseti Method (13, 14). The French Method has the disadvantage of the large time commitment, from both families and the treating therapist, with the daily treatment regime.

Ponseti Management

Ignacio Ponseti first began treating clubfeet with joint-sparing techniques during the 1940's (15). However, it was not until a 30-year follow-up study was published in 1995 (16) that the method gained the attention of the wider orthopaedic community. The Ponseti technique has been shown to result in a more flexible, functional foot, with less pain reported when compared with extensive postero-medial release surgery that has been used in the past (12, 17). The gait characteristics of clubfeet that do not undergo joint invasive surgery have been found to be superior to that of operated feet (18).

The Ponseti technique for treatment of congenital clubfoot has several stages, described in detail by Staheli et al (2009) (2). The first involves weekly gentle manipulation of the foot, maintained with long leg plasters (groin to toe). This requires an average of five casts (Appendix One has a photograph of a series of casts). The majority of infants will then require a percutaneous release of the Achilles tendon to gain adequate dorsiflexion of the ankle, with a post-op cast for approximately three weeks. In New South Wales (NSW) this tenotomy is usually carried out in a specialist clubfoot clinic at a tertiary children's hospital. Correction is maintained by wearing splinting for 23 hours a day for a period of three months, and then for sleep times, until four years of age (Appendix One has a photograph of the most common form of splinting). Some children will later also require a transfer of the tibialis anterior tendon to combat the residual muscle imbalance during dorsiflexion (19).

The Ponseti method has become widely accepted as the gold standard for the treatment of congenital clubfoot, with many successful case series being reported from both first (17, 20-24) and third world countries (25-30). In 2006 the management of clubfoot in Coffs Harbour had not kept up with best practice. Children born with this condition in Coffs Harbour were offered what is considered inferior management, whereas each of the tertiary children's hospitals in NSW had moved to Ponseti management in approximately 2000.

Clubfoot in Coffs Harbour

It was obvious that Coffs Harbour needed to change to Ponseti management for infants born with clubfoot. This treatment could be offered in two models. The first would have been to ask families to travel to a tertiary centre weekly for six to eight weeks for the serial casting, and then every few months until the child is four years of age. This is a significant amount of travel with a new baby, with obvious emotional and financial costs to families. There is also a financial cost to the Health Service,

with the Isolated Patients Travel and Accommodation Scheme (IPTAAS) partially funding these trips.

It has been reported that the distance to treating facilities is one of the greatest barriers to the success of the Ponseti method (31). Hence, local treatment seemed to be the preferable model, and could be accomplished by sharing the care of these infants with a tertiary children's hospital. The initial casting phase could be offered in Coffs Harbour, families would then need to travel to a tertiary centre for the tenotomy.

Once in splinting, this phase could also be managed locally, with occasional review by the tertiary centre. Compliance with the splinting regime has previously been identified as a major predictor of the success of the Ponseti Method (31-35). It is possible that local monitoring of the splinting regime may enhance compliance, and hence outcomes.

The Shared-care Model of Ponseti Management

This model of shared care has previously been reported as successful (36, 37). However, both of these studies were in England, with the tertiary institution only an hour away, much easier for ongoing consult purposes.

There are several reports of physiotherapist-led Ponseti clinics achieving comparable results, if not better, than clinics led by orthopaedic surgeons (38-40). Hence it appeared Coffs Harbour Health Campus (CHHC) should be able offer much of the treatment necessary under the Ponseti Method without, compromising clinical outcome, with all of the advantages of decreased travel and stresses for families.

Shared-care Ponseti Management in Coffs Harbour

Coffs Harbour is approximately half way between Sydney and Brisbane. To begin a shared care model of Ponseti management of clubfeet it was necessary to establish relationships with Ponseti Clubfoot clinics in all 4 of the surrounding Children's hospitals:

- Children's Hospital at Westmead (CHW)
- Sydney Children's Hospital
- John Hunter Children's Hospital, Newcastle
- Royal Women's Hospital, Brisbane

This allowed families to choose the venue for the tertiary component of their care, dependent upon where they had support or felt most comfortable to travel.

There were other regional centres throughout NSW that were beginning to offer this shared care model, but none had established relationships with as many tertiary centres. Nor had outcomes been evaluated to ensure local service delivery had not compromised clinical outcome for these infants. If the shared care model proved to have inferior clinical outcomes for these infants, then resources needed to be allocated to managing and supporting the frequent travel to a tertiary centre necessary for families with an infant clubfoot.

This project aims to investigate the outcomes of infants receiving Ponseti management under a shared care model in Coffs Harbour, compared to the outcomes of infants receiving Ponseti management exclusively at a tertiary children's hospital.

The Research Question

Does the shared care model of Ponseti management compromise clinical outcomes for infants born with clubfoot in the Coffs Harbour region?

The Wider Impact of this Study

Investigating the outcomes of the shared care model has implications wider than Coffs Harbour. It is difficult to define how many infants are born with clubfoot outside of metropolitan centres in NSW each year. The most recent NSW Mothers and babies Report (41) detailed:

- 96, 343 live births in 2008
- 96, 439 live births in 2009.

Using the reported incidence of 1-2/1000 births, this is approximately 96-192 cases of clubfoot in NSW each year. However, many of these will reside in metropolitan centres. The NSW Admitted Patient Data Collection (42) identified 205 cases of clubfoot in the financial year 2008-2009, 180 in the financial year 2009-2010. Data is unavailable prior to this time, as there was a change in data collection procedures, making previous data not comparable.

The Admitted Patient Data Collection (42) identifies the area of residence of patients. In the 2008-09 financial year, there were 85 infants with clubfoot that reside outside a major city admitted to hospital; in the 2009-10 financial year this was 54 infants.

These figures equate to an incidence of 2.1/1000 in 2008-09, 1.8/1000 in 2009-2010, similar to the reported incidence. This data needs to be interpreted with some care, as it includes all infants who have been admitted into hospital with a diagnosis talipes equinovarus, talipes equinovarus unspecified, and structural talipes equinovarus (talipes equinovarus is another name for clubfoot). Hence the data may include some infants who have a positional talipes that would not require Ponseti management, so we see the figures being at the higher end of the reported incidence figures.

Methods

Study Design

This project was a retrospective medical record audit of 2 groups of children treated for idiopathic congenital clubfoot using the Ponseti method. One group received their initial management at CHHC, the other at CHW. The time line involved in the study

did not allow for a prospective trial, as the incidence of clubfoot in the Coffs Harbour region is only a few each year.

A trial with wider scope, involving several regional centres, was considered. However, it was found that clinical practice varied between centres, making it inappropriate to combine these infants into one “regional” group for analysis. A large state-wide design was also briefly considered. This was difficult as most infants from the northern, southern and far western regions of NSW cross state borders for the surgical component of their Ponseti management. A study that required human research ethics approval in four states was considered outside the timeline available.

Study Group Recruitment

All infants born in the Coffs Harbour region July 2006 until June 2012, with idiopathic congenital clubfoot, were included as the study group. July 2006 was the start point, as this is when Coffs Harbour Health Campus adopted Ponseti management. It was decided to exclude any cases of clubfoot that were not idiopathic, for example those associated with myelomeningocele. The co-morbidities associated with congenital syndromes would confound results in this study. There were 14 infants included in the study group.

Control Group Recruitment

In order to compare Ponseti management in a share care model in the Coffs Harbour region to Ponseti delivered at a tertiary metropolitan centre, the control group were selected from infants who received their initial Ponseti casting at CHW. CHW has a dedicated clubfoot clinic that has been utilising the Ponseti method for over a decade now. This clinic receives approximately 70 new referrals for clubfoot each year.

When comparing the outcomes of two groups, even group sizes are optimal for robust statistical analysis. However, due to the small number of cases of clubfoot born in the Coffs Harbour region it was not feasible to have two even groups of children. To increase the statistical power of the study the number of infants in the control group from CHW was increased in a case-control model. According to Berry (2002) (43) if one group contains m subjects, and the other rm , then the study is equal to a study with N in each group where

$$N = \frac{2rm}{r+1}$$

Power analysis indicated if two controls from CHW were included for each Coffs Harbour case, this would equate to a study with 19 participants in each group; 3 controls for each case equates to a study with 21 participants in each group; 4 controls for each case equal to 22 participants in each group.

It was decided to include three CHW control subjects for each Coffs Harbour case, as adding the fourth control for each case did not considerably enhance the statistical power of the study. This was also the level of recruitment where saturation of the number of suitable control subjects within the CHW cohort was reached.

To select the most suitable control subjects from the CHW cohort a stratified recruitment strategy was used. Firstly, infants were matched for:

1. **Age** (infants were considered age matched if they were born within the same financial year)
2. **Gender**
3. **Unilateral or bilateral** clubfoot presentation

It was then attempted to match on the following criteria, in this order, where possible:

4. **Initial Pirani Score** (44) The Pirani Score is used in both study venues as an indicator of severity of initial presentation. It is widely used in the evaluation of clubfoot during initial Ponseti management, and has been shown to be a valid measure, with strong inter-observer reliability (41, 42). Control group infants were chosen with a Pirani score within one point of their matched Coffs Harbour case. Hence, this measure ensured that one group should not have a significantly worse severity of clubfoot, creating a bias in results. Increasing Pirani score indicates increased severity of presentation. The Pirani Scoring system is included in Appendix Two.
5. **Left/right presentation** in unilateral cases
6. **Family history** of idiopathic congenital clubfoot

When considering which cases to include from CHW, it was decided to include infants who had received only one cast or splint at another venue. CHW is not a maternity hospital, hence infants often received some form of initial treatment before referral to CHW. A few of the infants in the CHHC group had also received a cast elsewhere, for example in Sydney when attending for a clubfoot clinic review. It was considered that one cast is unlikely to change outcome dramatically, and had occurred in both groups. Infants who received two or more casts at an alternate venue were excluded from the study.

There were 37 suitable control subjects from CHW identified. This fell just short of the 42 planned, but no further suitable matched controls were available within the timeline for this project.

Clinical Outcome Measures

The outcome measures collected were:

- **Number of casts required** until the Achilles tendon release, or until a decision that the release was not necessary
- **Age of the infant at the time of Achilles tendon release**, if this was necessary
- **Post-operative passive range of motion of dorsiflexion** at the ankle
- **If boots and bar, or alternate splinting, were prescribed** for the infant
- **An indication of compliance** with the splinting regime. This was a subjective rating by the researcher reviewing the record and was simply graded as not compliant, partially compliant or compliant.

- **Further surgery** that has been required to maintain adequate correction. In a few of the older children a transfer of the tibialis anterior tendon may have been performed. Tibialis anterior tendon transfer was not included as a need for further surgery here, as this is considered a part of routine Ponseti management when it is required. Further surgery was considered relevant where it indicated a relapse of the clubfoot.
- **Further casting** that has been required for relapse in foot posture or function.
- **Dimeglio Score** (45). This is a widely used score of clubfoot severity. Neither of the institutions involved in this study utilise the Dimeglio for the initial casting phase, but use it when assessing older infants. The Dimeglio has been found to be both a valid and reliable tool (46, 47). As Dimeglio score increases it indicates a poorer outcome. A copy of the Dimeglio score sheet that was utilised in the data collection phase of the study is included in Appendix Three.
- **The Clubfoot Disease Specific Instrument (DSI)** (48, 49). This is a patient-based questionnaire of clubfoot outcome that has been shown to be sensitive to the unique health issues of children born with clubfoot. It has been shown to have good reliability, validity and discriminatory power (50). Unfortunately, the DSI has not previously been reported when used with a parent proxy. As the children involved in this study were all under the age of 6, they lack the ability to interpret and answer the questions adequately; hence a parent proxy was necessary. Matsumoto et al (51) has investigated the use of parents as proxy when rating paediatric orthopaedic outcomes in children 5-18 years. They found that there are some inconsistencies in reporting between parents and children. Hence some caution needs to be used when interpreting these results. The DSI investigates longer-term outcomes. At both of the study sites it is only used once infants are two years of age. Hence DSI data is only included for infants who have reached two years of age at the time of writing. Increasing DSI score indicates a worse outcome. A copy of the Disease Specific Instrument is included in Appendix Four.

Some of the older study participants had more than one Dimeglio and DSI score documented within their record. Where this was the case, the most recent score was utilised for analyses, the thought being that this would reflect longer-term outcome.

Some form of pedobarographic or gait analysis would have provided an excellent additional measure of outcome, as it is a very functional measure. This data, however, is time consuming and expensive to collect, hence is not routinely collected within clinic at either of the study venues. The fact that the Dimeglio score has no weight-bearing component is a criticism of this tool as a long-term outcome measure. However, it was one of the best and only measures included routinely within the records at both of the study venues.

The Financial Cost of Not Sharing the Care

The financial costs associated with travel to metropolitan children's hospitals for regional families were examined. A preliminary cost analysis was generated to examine cost savings and/or cost shifting for Health Services utilising the shared care model of Ponseti management.

Data Analysis

Results were analysed using Stata v. 11.1 (52). Other than information not available because the study participant was not old enough for that particular assessment (for example the DSI), there was very little missing data. Table 1 shows the number of missing values from each of the data sets. Note that all other measures not mentioned in the table have complete data sets.

Table 1: Number of Missing Values in the Data Sets

| Measure | CHW | CHHC |
|----------------|-----|------|
| PROM DF | | 2 |
| Dimeglio score | 5 | 3 |
| Clubfoot DSI | 17 | 3 |

CHW = Children's Hospital Westmead, CHHC = Coffs Harbour Health Campus, PROM DF = post-operative passive range of motion of dorsiflexion, DSI = Clubfoot Disease Specific Instrument

A pairwise deletion was used in all analyses rather than a listwise approach, as the sample size was already small and it was not feasible to remove participants who did not have complete data sets.

Within the published clubfoot literature there is no consensus in how best to analyse results from a combination of unilateral and bilateral presentations. It is common to pool data from unilateral and bilateral presentations (11, 53-55). As the results of Ponseti management are greatly influenced by splinting compliance (32, 35), if both feet from a bilateral presentation are included in analyses and the family are not compliant with splinting, two feet are affected by the one person. If pooling of unilateral and bilateral cases occurs, then this leads to a unit of analysis error.

With this in mind, first the difference in outcome scores across the two feet of bilateral cases was calculated and tested for differences in measures across the rural and metro groups using independent t-tests. If any statistically significant differences were found the minimum and maximum values across the two feet were obtained for each bilateral case. These statistics were used in subsequent analyses related to that measure to ensure similar results are obtained regardless of the statistical method used to summarise the values of the two feet in bilateral cases.

It was not expected that the data for number of casts necessary, post-operative range of dorsiflexion, Dimeglio score or DSI would be normally distributed. Normal scores fall at an extreme point of the scale of each measure. Therefore, in order to analyse differences across groups Wilcoxon rank sum analyses were conducted rather than independent t-tests.

A t-test was performed to identify a statistically significant difference between the two groups in age of Achilles tendon release.

Cross tabulations and Fisher's Exact Chi-squares were used to calculate proportional differences in compliance with splinting, further casting and further surgery across the two groups.

To explore clinical equivalency the Dimeglio score means were calculated for the CHHC and CHW groups separately with 99.0% confidence intervals. Dimeglio defined a score of less than 5 as benign (45), stating that "all such feet are healed by conservative functional treatment, and normal conditions are easily restored". This definition for good clubfoot outcome has been utilised several times within the literature (11-13, 47). Therefore, mean scores and corresponding 99.0% confidence intervals less than 5 would provide evidence that both groups could be considered to be achieving good outcomes utilising the Ponseti method.

Ethics Approval

Human research ethics approval was gained from the Children's Hospital at Westmead lead ethics committee, approval number LNR-2011-05-01. Site specific authorisation was also gained from both the CHW and Mid North Coast Local Health Network.

Potential Conflicts of Interest

The principle researcher is a paediatric physiotherapist at CHHC, currently involved in the management of infants born with clubfoot in the Coffs Harbour region. The co-researcher is a physiotherapist at CHW, currently involved in the management of infants with clubfoot referred there. Neither researcher stands to make financial gain from the results of this study.

Results

Pirani Score

By nature Pirani scores are not normally distributed as a score of less than 2.6 would indicate that there is not a talipes equinovarus deformity present. Hence, non-parametric statistical analysis was used. The median initial Pirani score in the CHHC group was 5.1 with an interquartile range from 4.0 to 6.0; in the CHW group it was 5.5 with an interquartile range from 4.5 to 5.7. In a comparison using the Mann-Whitney U-test, there was no significant difference between the initial Pirani scores of the two groups ($z=-0.30$, $p=0.76$).

Non-parametric Analyses of Outcome Measures

Number of casts necessary, post-operative range of dorsiflexion, Dimeglio score and DSI were all also analysed using the Mann-Whitney U-test, as normal distribution could not be assumed. Table 2 gives a summary of these analyses.

Table 2: Results of the Wilcoxon-rank sum analyses of outcome measures across the two study facilities

| Measure | CHW median (IQR) | CHHC median (IQR) | <i>z</i> | <i>p</i> |
|-------------------|---------------------|----------------------|----------|----------|
| Number of casts | 6 (4 to 6.5) | 6 (5 to 7) | 0.68 | 0.50 |
| PROM DF (degrees) | 25.0 (15 to 30) | 18.5 (10 to 27.5) | -1.22 | 0.22 |
| Dimeglio score | 3.0 (2.5 to 4) | 3.0 (2 to 4) | -0.47 | 0.64 |
| Clubfoot DSI | 13.0 (10 to 15) | 11.0 (10 to 11.5) | -1.23 | 0.22 |

CHW = Children’s Hospital Westmead, CHHC = Coffs Harbour Health Campus, IQR = interquartile range, PROM DF = post-operative passive range of motion of dorsiflexion, DSI = Clubfoot Disease Specific Instrument

Age at Achilles Tendon Release

The mean age of Achilles tendon release in the CHHC group was 7.6 weeks with a standard deviation of 2.0; in the CHW group it was 7.7 weeks with a standard deviation of 1.9. The age at Achilles tendon release was assumed to have a normal distribution, hence the difference between the groups was analysed with the student’s t-test. Once again, there was no significant difference detected between the two groups for the age at release ($t=-0.11$, $p=0.91$, $df=33$).

Splinting

Boots and bar, or alternate splinting, were prescribed for 100% of the infants in both groups, as per the Ponseti method protocol. Chi square analysis found no statistical difference between the two groups in compliance with the splinting regime. Table 2 summarises the subjective assessment of compliance.

The Need for Further Casting or Surgery

The number of children requiring further casting or surgery to maintain correction differed between the two groups, but the difference was not statistically significant. See Table 3 for a summary of the analysis between the two groups.

Table 3: Fisher’s Exact Chi-square analyses of differences in proportion of participants for splinting compliance, need for further casting and need for further surgery across the two study facilities.

| Outcome | CHW | CHHC | <i>p</i> |
|---------------------------|-----|------|----------|
| Compliance | | | 0.72 |
| Non-compliant | 1 | 1 | |
| Partially compliant | 9 | 3 | |
| Fully compliant | 27 | 10 | |
| Casting | | | 0.47 |
| Further casting necessary | 10 | 2 | |
| No further casting | 27 | 12 | |
| Surgery | | | 0.67 |
| Further surgery necessary | 5 | 3 | |
| No further surgery | 32 | 11 | |

CHW = Children’s Hospital Westmead, CHHC = Coffs Harbour Health Campus

Clinical Significance of the Dimeglio Scores

There was no statistical difference detected between the two groups in any of the outcome measures chosen. However, in order to provide evidence of clinical equivalence it was decided to compare results to outcome measures that have established levels of clinical significance within the literature.

The Dimeglio score has defined categories. A grade I foot is defined as benign, with a score of 0-5 points. Dimeglio (1995) (45) stated that “all such feet are healed by conservative functional treatment, and normal conditions are easily restored”. As such, a foot with a score of less than five at the end of treatment can be considered a satisfactory outcome.

When analysing the most recent Dimeglio score for each infant (an indicator of medium term outcome), it was possible to create a 99.0% confidence interval for both groups. The mean Dimeglio score for participants in the CHHC group was 3.0, with a 99.0% confidence interval from 1.7 to 4.2. In the CHW group the mean Dimeglio score was 3.4, with a 99.0% confidence interval from 2.7 to 4.1. For both groups the whole confidence interval fell below a score of 5. This is an indicator that both the CHW and CHHC groups were experiencing good outcomes with the Ponseti method.

The Financial Cost of Not Sharing Care

The median number of casts in this initial Ponseti management was six, and the average distance to the tertiary centres involved in the surgical component for the CHHC group is an 888km round trip. This would equate to IPTAAS funding each of these families to an average of \$1374 for travel associated with initial Ponseti management, to the point of surgical intervention. The number of appointments post surgery is impossible to estimate, as the exact number of reviews necessary will vary

with each infant over the course of their management, which is potentially their whole lifetime.

Discussion

Matching the Two Groups

This study is the first in Australia to directly compare outcomes when using the Ponseti method in a rural shared care model to those treated exclusively at a tertiary referral centre. The stratified recruitment strategy allowed for matching on the basis of age, gender and unilateral or bilateral presentations. Analysis of the Pirani scores has also shown that the two groups were matched for severity of initial presentation. It was not possible to match for right/left in bilateral presentations; nor for a family history of clubfoot. To match for these criteria also would have severely limited the number of infants included in the control group from CHW.

The two groups appear to be appropriately matched for comparison.

Comparison of Clinical Outcome Measures

The results of the study indicate both CHHC and CHW are achieving good outcomes with initial Ponseti management for idiopathic congenital clubfoot. Comparison of **every one** of the clinical outcome measures was unable to detect a statistically significant difference between the two groups.

The number of casts required until Achilles tenotomy was six in both groups. The age at which infants underwent the Achilles tenotomy was also the same for both groups. The practical difficulties in organising an infant from Coffs Harbour to attend a tertiary children's hospital for tenotomy appears to not delay the surgical component of Ponseti management. If surgery is delayed, usual practice would be to continue weekly casting, increasing the number of casts necessary until surgery. Hence there is no evidence of delay in surgery for regional infants.

Post-operative range of dorsiflexion was functional in both of the study groups. Whilst these ranges are not as much as expected in the normal infant foot, both groups achieved appropriate functional range. Approximately 15° dorsiflexion is required for the end stage of stance phase in normal gait; and both groups have achieved at least 15° of dorsiflexion.

Normal passive range of dorsiflexion in the newborn has been reported to be an average of 58.9° (56). In comparison, case series of infants treated for clubfoot within the literature have reported mean dorsiflexion of $13^{\circ} \pm 8.7^{\circ}$ when followed for an average of 18.8 years (15); and 20° when followed for an average of 26 months (57).

The Dimeglio score was the most recent measure for each infant involved in the study, therefore reflective of longer-term outcome. Many methods of clubfoot treatment have been able to achieve reasonable short-term results, however a long term functional, pain free foot is the ultimate goal. The median Dimeglio score was three for both groups, well within the range of good outcome. Church et al (2012) (58) recently reported a series of 22 children with Ponseti-treated clubfoot, followed

for an average of six years. They found a median Dimeglio score of four, very similar to both groups in the present investigation.

The 99% confidence interval of the Dimeglio score also fell wholly within a good outcome range (less than five points) for both groups. Confidence intervals were calculated in recognition that differences in clinical outcome are always going to be difficult to detect with small numbers of participants. Being able to create the 99% confidence intervals adds strength to the argument that there is no clinical difference between the two study groups.

There has been a growing emphasis on patient-based measures of outcome within orthopaedic literature. The Clubfoot DSI was included in this study, despite the inherent problems in using parents as proxy reporters. This measure is only utilised once the infants are two years of age at both of the study venues; there was only data available if the infant has remained in follow-up for this length of time. There is some drop-out rate in any long term clinic. A few of the infants within the study also transferred management to another venue before two years of age. Hence DSI score was not available for all study infants.

The two group medians were not significantly different, being 11 (CHHC) and 13 (CHW). These scores indicate good parental satisfaction with the outcome of their child's clubfoot treatment (the score for a normal foot is 10). Church et al (2012) (58) is one of the few publications using the clubfoot DSI score. They found a median score of 12 in their group, comparable to the outcomes in this present study.

Splinting

The splinting regime is an integral component of Ponseti management, it needs to continue until the infant is 4 years of age. One hundred percent of the infants within both groups were prescribed splinting in keeping with the Ponseti protocol.

Compliance with splinting has been shown to be a major problem and has a direct effect on the success of treatment (32, 34, 57). It was important to gain some indication of compliance within this study. This was very difficult to do in this retrospective study. The grading system used in the study is a subjective measure whereby the investigators rated their perceived level of compliance with splinting from indications within the medical record. As both study venues understand the importance of splinting compliance, there is usually reliable documentation on which to make the rating. Both venues experienced some non-compliance and partial compliance, consistent with what is reported within the published clubfoot literature (32, 34). Increasing splinting compliance is a focus within current research in Ponseti management (35, 59).

The Need for Further Casting or Surgery

The number of infants who require further casting or further surgery is an indication of the rate of relapse. Although the rate of further casting and surgery varied between the two groups, it was not statistically significant indicating the rate of relapse was similar across both sites. The rate of recurrence varies considerably within the published literature. Initial data from Iowa reported a relapse rate of 56% (60). However, the recommendation for splinting was extended to four years after this study, improving the reported relapse. Recently, there has been work on earlier

identification of relapses, early casting for relapses and improving splinting compliance (32, 35, 39). This work has made comparison of relapse rates difficult.

Although not statistically significant, considerably more of the children in the CHW group received further casting. There is a physiotherapist within the CHW clubfoot clinic who has a specific interest in early casting for relapse. The higher rate of casting observed in the CHW group may well reflect this particular clinical interest.

Implications For Clinical Practice

This study has been unable to detect any significant difference in clinical outcome between the two groups, supporting the shared care model as a viable option in Ponseti management. This model has obvious advantages for families, as repeated travel with a new infant must carry social emotional and financial stresses. This is especially so if the infant with clubfoot is not the firstborn. Finding appropriate care for other children whilst attending appointments hundreds of kilometres away is very difficult for many families.

Appropriate initial Ponseti management is very dependent on the skill level of the clinician. The physiotherapist offering Ponseti management in Coffs Harbour was skilled in the technique prior to working there. However, there is currently no recognised clinical competency to identify clinicians with the skills to offer Ponseti management in NSW. It has also not been defined how large a case load is enough to maintain that skill level. For example, if a small regional centre sees only one case of clubfoot every few years, this may not be enough to maintain an adequate skill level of this expert casting technique. The number of cases necessary to maintain an appropriate clinical skill level is very difficult to define. It is also possible that repeated attendance at training can maintain clinical skills. However, this may not be a financially viable option for the health service.

As discussed earlier, there are approximately 70 infants with clubfoot born regionally in NSW each year. This study suggests that some, but not all, of these families could be offered shared Ponseti management, saving them the burden of repeated travel.

The Cost of Travel if Care is Not Shared

IPTAAS only partially funds the financial cost of travel. The remainder of the cost is left for the families. The amount of this cost was unable to be estimated. Nor was there any analysis of the social and emotional cost of the repeated travel, should it be necessary for Ponseti management to occur only in a tertiary centre.

Further Research

Ideally, this small study would be followed by a multicentre trial, involving several regional centres that currently offer the shared care model of Ponseti management. Initial work would need to be done to standardise practice across the centres. Then, by increasing the study participants substantially, stronger evidence of clinical equivalence could be gathered.

Further research is also necessary to allow clear decision making over which regional centres could offer shared Ponseti management. It is necessary to define some form of clinical competency, so that shared care is only initiated where an appropriate skill mix is present. Not all regional centres should be offering this model of care.

In order to begin to define the number of cases necessary to maintain an appropriate skill level, it might be possible to look at published literature in other relatively rare conditions. From there, it will be necessary to investigate clinical outcomes in Ponseti clubfoot management, from different regional centres, with a range of clubfoot incidence.

It is probably not possible to exactly define the necessary caseload to maintain clinical skills, but hopefully some guide could be estimated. This would allow planning in which regional centres should allocate spending to ensure they have a clinician with the relevant skills to offer Ponseti management. In regional centres without the caseload necessary, resources need to be allocated into either:

- establishing a relationship with a neighbouring regional centre that may have the required caseload, or
- Supporting families in the frequent travel necessary for initial Ponseti management at a tertiary children's hospital.

A qualitative analysis of shared care could also help in refining the model. These qualitative projects could investigate the model from both the perspective of the family and the regional therapist offering Ponseti management.

Limitations of this Study

Clearly, sample size is a major limitation of this study. This is a common problem in research involving relatively rare paediatric conditions. To this end, human research ethics approval has now been granted to continue the study for a total of five years. This will allow data to be collected on more infants at both study sites. The case control study design was the most appropriate design to use with such a small group.

The short follow-up time is another limitation. Longer term outcome measures are a better indication of the success of the Ponseti method. The continuation of this study will also address this issue to some degree.

Better evaluation of outcome of clubfoot treatment would also include some form of weight bearing measure. This is not routinely collected at either of the study sites, hence could not be included in this retrospective study. It is well worth consideration in a long term prospective study.

A true equivalence study is prospective by nature. This would provide a stronger level of evidence, but was simply not achievable within the timeframe of this programme.

One of the great strengths of this study is that it captures all of the infants born with clubfoot within the Coffs Harbour region over the study period – the entire population. The data sets are also very strong, with very little missing data. Hence an accurate picture of this population can be portrayed. This will assist the transfer of these findings to other comparable regional centres.

Conclusions and Recommendations

This study has been unable to detect a significant difference between the outcomes of infants who receive their initial Ponseti casting in Coffs Harbour and those who receive their initial casting at CHW. This result supports the shared care model of Ponseti management, as it is offered in Coffs Harbour. However, clinical equivalence is always a difficult issue to prove. Hence the extension of the project to a total of 5 years, to add participants and strength to the argument.

The scope of this investigation now needs to broaden, taking a state-wide perspective. The regional sites that have the ability to offer initial Ponseti management need to be identified and supported in doing so. The regional sites with an appropriate case load, but no clinician with the appropriate skills to offer Ponseti management, also need to be identified. Training in Ponseti management then needs to be offered to appropriate clinicians within these regional areas. The NSW CTEV Network Working Party is already working on an E-learning package to begin to address this need, but further work is still necessary.

Not all regional centres should be offering initial Ponseti management. It is likely there is a minimal caseload necessary to maintain reasonable clinical skill level, however this is yet to be defined. Smaller regional centres, that carry a minimal caseload of clubfoot, need to be supported in recognising idiopathic congenital clubfoot, then organising initial management at either an appropriate neighbouring regional centre, or a metropolitan children's hospital.

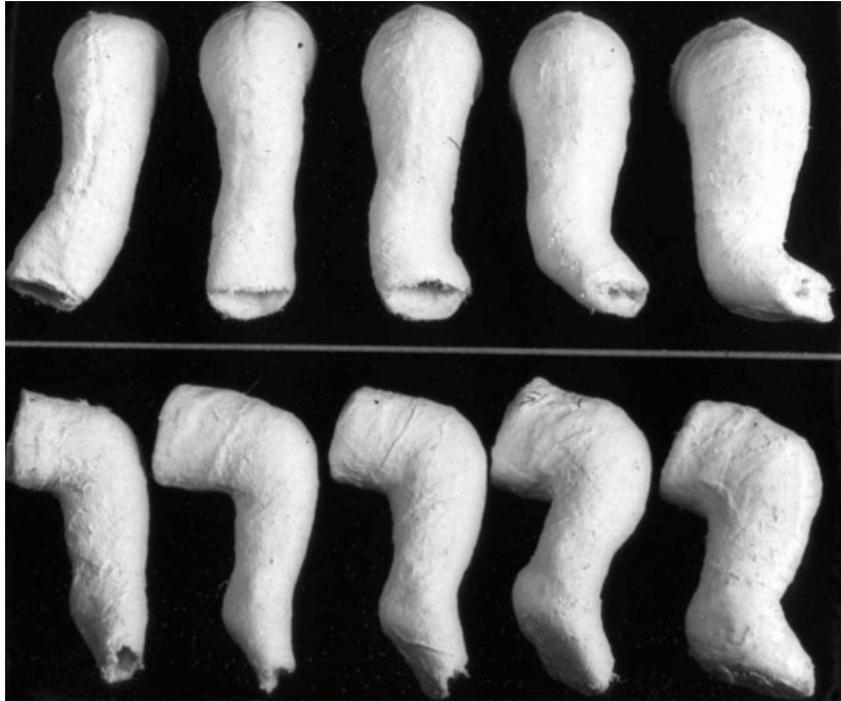
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61. Ponseti Cast Series. ehlaidlaw.blogspot.com [Accessed on 26 June 2012].
62. Mitchell Boots and Bar. wishuponahero.com [Accessed on 26 June 2012].

Appendix 1



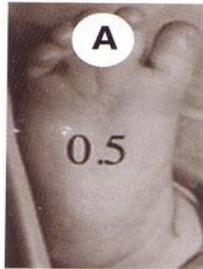
Initial Ponseti Cast Series. Week 1 on the left through to week 5 on the right.
Anterior view on top line, medial view bottom line. (61)



Mitchell Boots and Bar, one of the most common splinting in Ponseti management.
(62)

Appendix 2

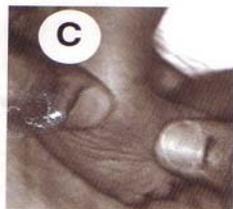
Pirani Score Sheet



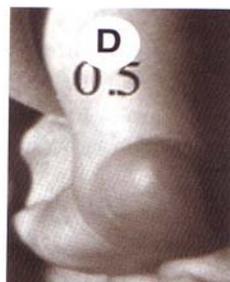
Curvature of the Lateral Border



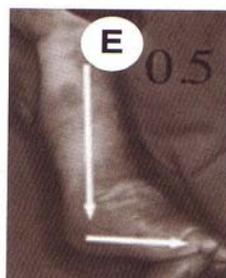
Medial Crease



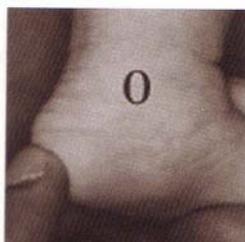
Lateral Head of Talus



Posterior Crease



Rigidity of Equinus



Emptiness of the Heel

Appendix 3

Dimeglio Score Sheet from Data Collection Sheets

| 7. Dimeglio Scale | | | | | |
|-------------------|------------|----------|--------------|----------------|----------------|
| Score | 0 | 1 | 2 | 3 | 4 |
| Equinus | > + 20 deg | 0-20 deg | -20 to 0 deg | -45 to -20 deg | -90 to -45 deg |
| Varus | > + 20 deg | 0-20 deg | -20 to 0 deg | -45 to -20 deg | -90 to -45 deg |
| Sup/Abd | > + 20 deg | 0-20 deg | -20 to 0 deg | -45 to -20 deg | -90 to -45 deg |
| Met Add | > + 20 deg | 0-20 deg | -20 to 0 deg | -45 to -20 deg | -90 to -45 deg |
| Post crease | No | Yes | | | |
| Med crease? | No | Yes | | | |
| Cavus | No | Yes | | | |
| Muscle | No | Yes | | | |

| Date | | | | | | | | |
|---------------------|-------|------|-------|------|-------|------|-------|------|
| | Right | Left | Right | Left | Right | Left | Right | Left |
| 1. D.F | | | | | | | | |
| 2. Heel Varus | | | | | | | | |
| 3. Abd (supin) | | | | | | | | |
| 4. Met Adductus | | | | | | | | |
| 5. P.C | | | | | | | | |
| 6. M.C | | | | | | | | |
| 7. Cavus | | | | | | | | |
| 8. Muscle deviation | | | | | | | | |
| Total | | | | | | | | |

**Refer to Dimeglio (1995) for diagrams to assist in evaluation

Appendix 4

Clubfoot Disease-Specific Instrument Parent Form

We are carefully evaluating the condition of your child's foot after treatment. Please circle the one best answer to each question. **All results will be kept strictly confidential.**

| | | | | | |
|----|--|-----------------|---|---|--------------------|
| 1. | How satisfied are you with the status of your foot? | 1 | 2 | 3 | 4 |
| | | Very satisfied | | | Very dissatisfied |
| 2. | How satisfied are you with the appearance of your foot? | 1 | 2 | 3 | 4 |
| | | Very satisfied | | | Very dissatisfied |
| 3. | Rate the amount of teasing you have related to the clubfoot. | 1 | 2 | 3 | 4 |
| | | Never teased | | | Always teased |
| 4. | Rate problems finding shoes that fit you. | 1 | 2 | 3 | 4 |
| | | Never a problem | | | Always a problem |
| 5. | Rate problems finding shoes that you like. | 1 | 2 | 3 | 4 |
| | | Never a problem | | | Always a problem |
| 6. | Do you complain of pain in the foot that was operated on? | 1 | | | 4 |
| | | No | | | Yes |
| 7. | Rate your limitations in the following activities. | | | | |
| | a. Walking | 1 | 2 | 3 | 4 |
| | | Not limited | | | Completely limited |
| | b. Running | 1 | 2 | 3 | 4 |
| | | Not limited | | | Completely limited |
| 8. | How much do you experience pain during heavy exercise? | 1 | 2 | 3 | 4 |
| | | Never | | | Always |
| 9. | How much do you complain of pain during moderate exercise? | 1 | 2 | 3 | 4 |
| | | Never | | | Always |