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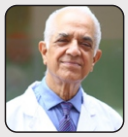
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Ponseti Casting in Obtaining Correction of Non-idiopathic Clubfoot: Where do we Stand? A Tertiary Care Institution-Based Prospective Observational Study

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Abstract

Introduction: Background: Clubfoot is a common congenital deformity, with idiopathic cases well managed by the Ponseti method. However, non-idiopathic clubfoot (NICF), often associated with syndromic or neuromuscular conditions, remains challenging. Evidence on Ponseti casting in NICF is limited, and outcomes vary across etiologies.

Methods: This prospective observational study was conducted at a tertiary care center between September 2011 and September 2013. Out of 739 patients with clubfoot, 38 patients (59 feet) were diagnosed with NICF. Severity was assessed using Pirani and Diméglio scores. Ponseti casting was performed with weekly or accelerated cast changes, followed by Denis-Browne splinting. Relapse and non-compliance were documented, and surgical interventions were tailored when casting failed.

Results: The majority of cases were due to Streeter's dysplasia (36.9%) and arthrogryposis multiplex congenita (23.7%). All feet were rigid (Diméglio >5). Significant improvement in Pirani scores was observed across age groups (mean reduction from 5.2 ±1 to 2.9 ±1.4, $P < 0.001$). The mean number of casts required was 6.84, with Charcot-Marie-Tooth disease requiring the highest (13 casts). Correction was achieved in 61.7% of feet with casting ± heel cord release, while 38.3% required surgery. Relapse occurred in 18.9% of feet, mostly linked to splint non-compliance (31.6%). Parent-reported satisfaction was high (78.9%).

Conclusion: Ponseti casting provides a reasonable success rate (61%) in NICF, particularly in younger age groups, and should be considered a first-line treatment before surgical intervention. Despite higher rigidity and relapse rates compared to idiopathic cases, functional and cosmetic outcomes were satisfactory, with minimal complications.

Keywords: Dimeglio grading, Non-idiopathic clubfoot, Ponseti casting, Pirani score, Secondary clubfoot.

Introduction

Clubfoot has been an enigma for medical professionals for a long time. Idiopathic cases with unknown cause has various hypothesis has been postulated regarding its pathogenesis and causation [1]. Non-idiopathic clubfoot (NICF) constitutes the cases, where other pathology is a reason behind its causation and is also known as secondary clubfoot or syndromic clubfoot [2, 3]. NICF and its management have been an dilemmatous topic to orthopedic surgeons. Ponseti casting is the standard of care for management of idiopathic clubfoot, which has shown fabulous results [4]. Literature search shows limited studies regarding Ponseti casting and soft tissue

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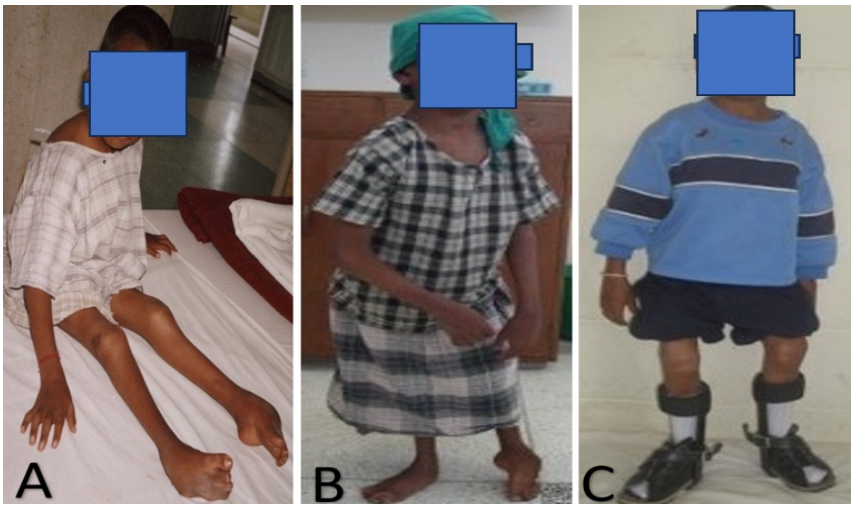

Figure 1: Illustrative photo of a kid with Arthrogryposis Multiplex Congenita (a), who after casting failure underwent bilateral talectomy, initially right sided (b), followed by left sided (c) talectomy with regaining of motility.

release protocols in NICF [5]. Studies with an individualistic etiologic approachhave been conducted on arthrogryposis multiplex congenita (AMC) [6-9], spina bifida [10, 11, 12], or tethered cord syndrome [13]. Some reports have addressed the efficacy of casting, emphasizing the need for additional studies to be needed to better assess its effectiveness [14, 15]. In our study, we provide a more comprehensive causes of NICF and its management. Being a tertiary level center catering to the large number of idiopathic and NICF cases at our facility, we decided to conduct this prospective study to shed some light on this topic. The main aim of this study is to evaluate the outcomes of the treatment of NICF with the Ponseti method of casting.

Materials and Methods

This observational prospective study was conducted between September 2011 and September 2013. Institutional Review Board approval was obtained after presenting the research protocol to the board of expertise. Among 739 patients with clubfoot, 38 patients (59 feet) were diagnosed with NICF. The consecutive cases of NICF between 3 months and 60 months of age were included in the study. Informed consent was obtained from family members. Pro forma was designed for the uniform evaluation of all patients. The severity of the deformity was assessed by the Pirani scoring system [16] on the initial visit, and subsequent follow-up. At the initial evaluation, the foot was also evaluated by the Diméglio grading system [17]. Casting was performed using the Ponseti protocol [18]. The patients were managed on both an outpatient and an inpatient basis. Accordingly, two protocols were followed for the cast change. Those patients managed as outpatients underwent cast change every 7th day, while those admitted were managed on an accelerated Ponseti program with the cast change on every 5th day [19].

A Denis-Browne (DB) splint [20] was used to maintain the correction. Parental self-reports regarding splint use were utilized to evaluate compliance. Non-compliance was defined when the DB splint was not used for at least 10 h a day. A relapse was defined as the appearance of any of the components of the deformity, either cavus, adductus, varus, and/or equinus, after multiple casting being offered. Relapse after initial correction was treated with additional manipulation and serial casting in marked foot abduction. When the Pirani Score showed no improvement after multiple castings, the surgical procedure was considered. The type of surgery required was dictated by the suppleness of the foot. Heel cord release (HCR) was performed for the supple foot, while open tendo-achilles lengthening or posterior release was offered for the rigid foot. Posterior release was performed only when the adduction of the forefoot and varus angulation of the hindfoot were corrected. Complications that occurred during casting were also recorded.

Statistical tools

Data were entered in Microsoft Excel with coding of the variables. SPSS software version 16 was used for the data analysis. Frequencies, percentages, and means with standard deviations were calculated to describe the quantitative data. A paired sample t-test was used to compare the means between

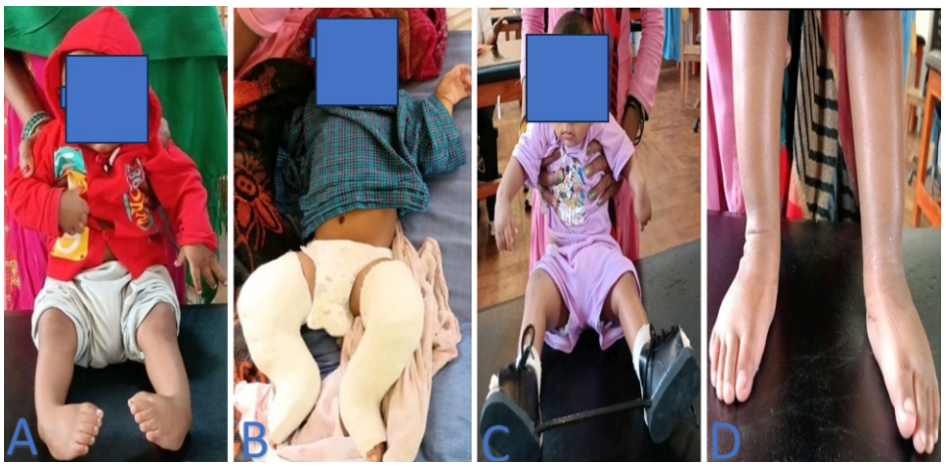

Figure 2: Illustrative photo of a kid with Arthrogryposis Multiplex Congenita (a), who, after serial casting (b), followed by Denis-Browne splint (c) maintained plantigrade foot (d).

Table 1: Socio-demographic characteristics of the participant with etiology, foot involvement, and type of care.

	n (%)
Gender	
Male	21 (55.3)
Female	17 (44.7)
Ethnicity	
Brahmin/Chhetri	23 (60.5)
Janajati	8 (21.1)
Dalit	7 (18.4)
Age distribution in months	
3–12	9 (23.7)
13–24	8 (21.0)
25–36	8 (21.0)
37–48	6 (15.8)
49–60	7 (18.4)
Foot involvement	
Bilateral	21 (55.3)
Unilateral (Right)	12 (31.6)
Unilateral (Left)	5 (13.2)
Etiology	
Streeter's dysplasia	14 (36.9)
AMC	9 (23.7)
CP	4 (10.5)
Spina Bifida	4 (10.5)
DDH	2 (5.3)
SPS	2 (5.3)
CMTD	1 (2.6)
Larsen's syndrome	1 (2.6)
Pterygia syndrome	1 (2.6)
Type of care	
Inpatient	21 (55.3)
Outpatient	17 (44.7)

AMC: Arthrogryposis multiplex congenita, CMTD: Charcot-Marie-Tooth disease, CP: Cerebral palsy, DDH: Developmental dysplasia of the hip, SPS: Stiff Person syndrome

the two groups. Analysis of Variance was used to compare the means between more than two groups. A $P < 0.05$ was assigned statistically significant.

Results

Among the 38 patients (59 feet), 17 were female (44.7%), and 21 were male (55.3%), ranging from 3 months to 60 months. Foot involvement was observed on the right (12, 31.6%), left (5, 13.2%), and bilateral (21, 55.3%) sides. Among the various etiologies observed, the majority of patients had streeter's dysplasia (14, 36.9%), while Charcot-Marie-Tooth Disease (CMTD) (1, 2.6%), Larsen's syndrome (1, 2.6%), and Pterygium[TPS1.1] syndrome (1, 2.6%) were the least prevalent causes.

Patients were treated as either inpatients (21, 55.3%) or outpatients (17, 44.7%) (Table 1).

All of the foot of NICF cases were inflexible (Diméglio score >5). Diméglio grading system demonstrated grade II (5, 8.5%), grade III (4, 6.8%), and grade IV in (50, 84.7%) feet.

A significant improvement was observed between pre-cast and post-cast Pirani scores across various age groups and etiologies ($P < 0.001$) (Table 2).

The mean number of casts required was 6.84 (4-10). Patients with lower age groups (3–12 months) and patients with CMTD required the highest mean number of casts (7.6 and 13, respectively) (Table 3). Cast sores were encountered in three patients and were managed accordingly.

The ankle range of motion (ROM) was assessed at initial and final follow-up. A decreasing trend of ROM score was seen in all age groups during the follow-up, with a maximum in the 49–60 months age group. In terms of etiology, dorsiflexion demonstrated a maximum decreasing trend seen in AMC, CMTD, and Larsen's syndrome. No change was seen in Pterygia syndrome. The immediate and final follow-up mean plantar flexion were 28 ± 11.4 and 23.38 ± 13.4, respectively ($P = 0.51$). The immediate mean of abduction of the foot was 28 ± 15.9, and the final follow-up mean was 28.5 ± 16.7 ($P < 0.05$). An almost similar rate of reduction in abduction was observed during follow-up in all age groups except in 49–60 months of age (Table 4).

One patient was lost to follow-up, and 37 patients (58 feet) were followed up for a mean duration of 10.58 months (3–24 months). Recurrence was noticed in eight patients (11 feet). They were managed with posterior medial release (5 feet), talectomy (3 feet) (Fig. 1), talar decancellation (2 feet), and capsular release (1 foot).

Parents were enquired regarding compliance with the DB splint, with compliance in 68.4% and non-compliance in 31.6%. Thirty patients' families (78.9%) were found to be satisfied with the functional and cosmetic plantigrade appearance of the foot after treatment. Evaluation revealed possible squatting in (22, 58%), while (8, 21%) were immature to undergo squatting. Percutaneous HCR was performed in (34, 57.9%) feet during the course of casting. Surgery was tailored to each foot at the end of the casting who failed the treatment. Our evaluation revealed that 36 feet (61.7%) were treated successfully with casting and HCR (Fig. 2). In the remaining (22, 37.3%) feet, surgery was performed (Table 5).

Analysis revealed that there was a significant difference in ROM scores between those who complied versus not complied to the DB splint use (Table 6).

Table 2: Pirani scores before and after casting based on age and etiology.

	Pre-cast (Mean±SD)	Post-cast (Mean±SD)	*P*-value
Overall Pirani score	5.2±1	2.9±1.4	<0.001
Age in months			
3–12	5.9±0.2	2.3±0.8	<0.001
13–24	5.6±0.5	2.7±0.8	<0.001
25–36	5.2±0.9	3±1.2	<0.001
37–48	4.1±1.3	2.4±1.3	<0.05
49–60	4.5 ±0.9	3.7±1.1	<0.05
Etiologies			
AMC	5.47 ±0.56	3.8 ±0.84	
Streeter's dysplasia	5 ±1.26	2.45 ±0.82	<0.001
CMTD	6 ±0	3.5 ±0	
CP	5.33±0.82	2.42±0.58	<0.001
DDH	6±0	2.5±0.58	<0.001
Spina Bifida	3.8±0.76	1.5±1.17	<0.001
SPS	5.5±0	1.5±0	<0.001
Larsen's syndrome	6±0	4.0±0	<0.001
Pterygia Syndrome	4.5±0	4.5±0	

AMC: Arthrogryposis multiplex congenita, CMTD: Charcot-Marie-Tooth disease, CP: Cerebral palsy, DDH: Developmental dysplasia of the hip, SPS: Stiff Person syndrome, SD: Standard deviation

Surgery was performed in the form of Talar Decancellation, Posterior Medial release, Talectomy, Dillwyn Evans procedure, and Capsular release (Table 7).

AMC: Arthrogryposis multiplex congenita, CMTD: Charcot–Marie-Tooth Disease; CP: Cerebral palsy, DDH: Developmental dysplasia of hip, HCR: Heel cord release, TAL: Tendo Achilles lengthening

Discussion

Clubfoot is one of the most prevalent congenital birth defects, occurring in about one out of every 1000 live births. The Ponseti approach, known for its outstanding results in the treatment of idiopathic clubfoot, has only a few documented instances in the literature concerning its application for NICF [9]. Prior research has shown its effectiveness against a narrower range of etiologies, while our study encompasses a broad range of etiologies, thereby enhancing the generalizability of our findings to a wider array of causes of NICF. This study demonstrates comprehensive causes of NICF, including AMC, Streeter's Dysplasia, CMTD, CP, DDH, Spina Bifida, SPS, Larsen's Syndrome, and Pterygia syndrome. In line with this finding, Moroney et al. reported AMC (n = 5), trisomy 21 (n = 4), and spina bifida (n = 4) as the etiologies of NCIF [21]. Similarly, a few previous studies reported AMC [6, 7, 9], spina bifida [10, 11, 12], or tethered cord syndrome as a common cause of NICF [13].

Boehm et al. in their study reported (11; 22 feet) as Diméglio grade IV and (1; 2 feet) as Diméglio grade II (9). Gerlach et al. reported (11, 28 feet, 39%) in the myelomeningocele as Diméglio grade IV [12]. Our study indicated Diméglio grade II (5, 8.5%), grade III (4, 6.8%), and grade IV in (50, 84.7%) feet. Our results vary with the previous literature, demonstrating more rigid feet during presentation. This variability may be attributed to a wide range of etiologies, along with the fact that patients often present late when seeking care. The latter could stem from a limited understanding of the pathology and the time constraints involved in reaching our tertiary care facility from rural areas.

In our research, we assessed Pirani scores across various age groups: 3–12 months, 13–24 months, 25–36 months, 37–48 months, and 49–60 months with average pre-casting scores of 5.9, 5.6, 5.2, 4.1, and 4.5, respectively, and post-casting scores of 2.3, 2.7, 3.0, 2.4, and 3.7, respectively, with mean correction post-casting at final follow-up being 2.82; the optimization was statistically significant ($P < 0.001$). This aligns with the research carried out by Sharma et al., where they analyzed 23 NICF patients. In this study, the mean Pirani score was 4.58 ± 1.10 at the first visit and

Table 3: Number of casts required in accordance to age and etiology

	Mean number of cast	Number of patients
Age in months		
3–12	7.6	15
13–24	6.3	12
25–36	7.7	13
37–48	7.1	9
49–60	5	10
Total	6.84	59
According to etiology		
AMC	6.9	16
Streeter's dysplasia	6.6	22
CMTD	13	2
CP	6.7	6
DDH	5.5	4
Spina Bifida	6.4	5
SPS	6.5	2
Larsen's syndrome	10	1
Pterygia syndrome	4	1
Total	6.9	59

AMC: Arthrogryposis multiplex congenita, CMTD: Charcot-Marie-Tooth Disease, CP: Cerebral palsy, DDH: Developmental dysplasia of the hip, SPS: Stiff Person syndrome

Table 4: Range of motion score (dorsiflexion, plantarflexion, abduction) at initial and final follow-up according to age and etiology

	Dorsiflexion score at initial presentation		Dorsiflexion score at final follow-up		*P*-value
	Mean	SD	Mean	SD	
Age in months					
3–12	10.4	6.1	4	14.6	0.078
13–24	27.1	8.4	20.8	10.6	<0.01
25–36	6.2	6.5	5.4	5.6	0.7
37–48	7.8	10	7.2	11	0.923
49–60	14.5	13.2	3.5	15.8	0.072
According to etiology					
AMC	5.94	10.04	0.31	11.47	
Streeter's Dysplasia	9.55	8.85	2.5	12.7	
CMTD	15	0	0	0	-
CP	9.17	7.36	7.5	6.12	<0.001
DDH	8.75	2.5	10	0	-
Spina Bifida	12	2.74	15	5	<0.001
SPS	12.5	3.54	5	21.21	<0.001
Larsen's Syndrome	30	-	10	-	-
Pterygia Syndrome	0	-	0	-	-
	Plantarflexion score at initial presentation		**Plantarflexion score at final follow-up**		
Age in months					
3–12	37.7	9.4	37.7	11.5	1
13–24	32.5	11.2	30	11.7	0.324
25–36	28.5	15.5	31.2	16.1	<0.01
37–48	27.8	11.5	30	13.9	0.225
49–60	7.5	15.9	8	16.9	0.343
According to etiology					
AMC	18.12	17.88	13.75	15.22	
Streeter's dysplasia	30.68	15.3	32.27	15.79	<0.001
CMTD	40	0	40	0	-
CP	38.33	9.31	39.17	9.7	<0.001
DDH	32.5	8.66	37.5	2.89	<0.001
Spina Bifida	31	9.62	36	8.94	<0.001
SPS	32.5	17.68	42.5	3.54	<0.001
Larsen's syndrome	0	-	0	-	-
Pterygia syndrome	25	-	20	-	-
	Abduction score at initial presentation		**Abduction score at final follow-up**		
Age in months					
3–12	32.7	8	30	10.4	<0.05
13–24	27.1	8.4	20.8	10.6	<0.05
25–36	25.8	11	25	11.7	0.165
37–48	30.6	18.1	25	20.9	<0.05
49–60	22.5	10.9	13	10.3	<0.001
According to etiology					
AMC	21	4	12.81	3.64	
Streeter's dysplasia	28	11	23.64	11.97	<0.001
CMTD	20	0	20	0	-
CP	33	11	31.67	13.66	<0.001
DDH	40	0	40	0	-
Spina Bifida	44	11	41	15.17	<0.001
SPS	30	0	25	7.07	-
Larsen's syndrome	0	-	0	-	<0.001
Pterygia syndrome	15	-	10	-	-

AMC: Arthrogryposis multiplex congenita, CMTD: Charcot-Marie-Tooth Disease, CP: Cerebral palsy, DDH: Developmental dysplasia of the hip, SPS: Stiff Person syndrome, SD: Standard deviation

1.41 ± 1.02 at the final follow-up [22].

The mean number of casts required based on our data was 6.84. Similarly, Janicki et al. reported the mean number of casts applied being 6.4 [23]. In a study by Boehm et al., initial correction was achieved in all clubfeet with a mean of 6.9 ± 2.1 casts [9]. Dunkley et al. reported the NICF group requiring a median of 7 casts to correct the clubfoot deformity [24]. Matar et al. reported initial correction being achieved in all NICF cases with an average of 8 (4–10) casts, and 7 (4–9) casts in two different studies from the same author [7, 10]. This aligns with the mean number of casts used in our study.

Correction without need for surgery other than HCR in our study was 61%, while Gerlach et al. in their study found 24 of 28 clubfeet in the myelomeningocele group with satisfactory result [12]. Boehm et al. in their study found correction in 18 of 22 feet in AMC [9]. Janicki et al. demonstrated correction in 29 of 40 feet in NICF due to neuromuscular or syndrome-associated cause [23]. Moroney et al. reported 73% of NICF with better results [21]. Kowalczyk et al. reported 8 of 10 feet of AMC gaining correction [6]. Dunkley et al. reported 64.6% of NICF correction [24]. El-Fadl et al. demonstrated 43 of 48 feet showing some result with serial casting, while 5 of 48 failed to show any result for myelomeningocele [11]. Matar et al. demonstrated two-thirds of patients having a satisfactory outcome at final follow-up, with functional plantigrade, pain-free feet in AMC with Ponseti casts and a HCR [7]. Similarly, nine children with 15 of 18 (83.3%) myelomeningocele-associated clubfeet achieved a satisfactory outcome at the final follow-up, with functional, pain-free feet from the same author [10]. These reports indicate that results vary across different studies, with isolated cases, such as myelomeningocele, demonstrating higher correction rates. In contrast, studies that included a range of causes yielded results similar to those of our study. This indicates that the correction rate can be estimated to be approximately two-thirds among patients with NICF, and that casting efforts are warranted before proceeding to surgical interventions.

In our study, 31.6% of the patients demonstrated non-compliance with the splint. Similarly, Boehm et al. reported 6 feet among 24 feet (25%) having a relapse after initial successful treatment, and all relapses were related to non-compliance with prescribed brace wear [9]. Our study revealed eight patients (11 feet; 18.6%) with relapse. These patients were managed with posterior medial release in 5 feet, talectomy in 3 feet, talar decancellation in 2 feet, and capsular release in 1 foot. Jackson et al. demonstrated the recurrence within the first 2 years after casting initiation, being 42% in the tethered

Table 5: Rate of success, failure, relapse, compliance, and satisfaction in patient undergoing Ponseti for non-idiopathic clubfoot

	% of cases	Number of feet
Success rate	61.7	36
Failure rate	38.3	22
Relapse rate	18.97	11
Compliance rate to DB splint	68.4	40
Parent reported satisfaction	78.9	30 parents

DB: Denis-Browne,

cord syndrome [13]. Recurrence requiring additional treatment occurred in (16, 44%) of 36 feet in a study by Janicki et al. [23] and recurrence of deformity being 44% of NICF in a study by Moroney et al. [21]. Dunley et al. reported surgery requirement in 26 % of relapsed NICF [24]. Gelfer et al. reported recurrence being noted in (14, 48.3%) [25]. Parent-reported satisfaction has been observed in 78.9% of the cases in our study. We couldn't found any reports on parents' reported satisfaction during treatment of NICF.

This study's strength lies in its prospective follow-up design and its thorough demonstration and discussion of a rare entity in pediatric orthopedic surgery. This work will contribute to and enhance the understanding of the efficacy of the Ponseti method of casting in NICF, addressing gaps in the existing literature. Alongside evaluating the success and failure rates of the Ponseti method, our study has examined splint compliance and investigated parent-reported satisfaction. Our limitation is the inability to perform long-term follow-up after the tenure of research work, being a small sample-sized study and having no control groups for comparison. This study would serve as a foundation for future trials or studies with a higher level of evidence.

Conclusion

The success rate of Ponseti casting in NICF treatment was reasonable (61%). For the majority of the foot (57.9%), only percutaneous HCR was necessary. The number of casts required varied based on the etiology, with CMTD requiring the most casts for correction. The younger age group of 3–12 months required a greater number of casts, showing the most significant improvement in Pirani scores at the final follow-up. 80% of families indicated they were satisfied with the treatment. Only a few complications arose from the treatment method, with cast sores occurring in three patients.

Table 6: Mean score comparison of DB Splint and ROM

ROM	DB splint				
	Complied (68.4%)		Not complied (31.6%)		*P*- value
	Mean	SD	Mean	SD	
Plantar flexion	36.15	9.7	13.5	17.48	<0.001
Dorsiflexion	5.64	10.4	0	12.57	0.071
Abduction	28.33	13.1	13.75	8.56	<0.001

DB: Denis-Browne, ROM: Range of motion

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