

Original Article

Management of rocker-bottom deformity during Ponseti treatment of congenital idiopathic clubfoot

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Abstract: Objective: To investigate the clinical and radiographic presentations and management of rocker-bottom deformity during Ponseti treatment of congenital idiopathic clubfoot. Methods: In this retrospective study, 6 patients (9 feet) with idiopathic clubfoot compared to 185 outpatients (276 feet) admitted to Shanghai Children's Medical Center between August 2015 and July 2016 were recruited. Ponseti casting plus percutaneous Achilles tenotomy with/without Kirschner wire internal fixation of the talo-navicular joint were performed, followed by brace use. Dimeglio score, lateral radiograph of the foot in maximal dorsiflexion (the tibiocalcaneus angle, the mean calcaneus-fifth metatarsal angle and the mean talo-first metatarsal angle) and previous treatment data were recorded. Results: Among the 6 clubfoot patients (9 feet), 3 were male and 3 female. Three patients had bilateral clubfeet, 5 cases of right clubfoot and 4 cases of left clubfoot. All patients were aged 5-19 months. Lateral radiograph showed that the mean tibio-calcaneus angle during postoperative follow-up was $(71.23 \pm 13.62)^\circ$, significantly lower compared with preoperative angle of $(82.41 \pm 15.10)^\circ$. The postoperative mean calcaneus-fifth metatarsal angle was $(24.98 \pm 8.81)^\circ$, significantly lower than $(43.77 \pm 13.41)^\circ$ before operation. The postoperative mean talo-first metatarsal angle was $(22.82 \pm 11.62)^\circ$, considerably lower compared with $(34.77 \pm 6.20)^\circ$ before surgery. At the final follow-up, eight clubfeet were successfully corrected and merely one clubfoot was not effectively treated. Conclusions: Rocker-bottom deformity should be identified during Ponseti management of congenital idiopathic clubfoot. Lateral radiographs of the foot in the maximal dorsiflexion can assist the diagnosis. Achilles tenotomy probably lowers the risk of rocker-bottom deformity.

Keywords: Clubfoot, rocker-bottom deformity, Ponseti method, pediatrics

Introduction

Congenital clubfoot is one of the most common birth defects in the musculoskeletal system, whereas the underlying pathogenesis remains elusive. Conservative treatment is primarily adopted during the early stage [1, 2], of which the Ponseti method is currently recognized as the first therapeutic option [3]. Although early correction yields a high success rate and good repeatability, it still faces the issues of residual deformity and high recurrence in certain patients [4-7].

Through thorough investigation of subtalar joint biomechanics, Professor Ponseti has attempted to utilize a specialized manual and well-modeling cast to achieve the successful correction, and adopt percutaneous Achilles

tendon releasing to avoid the rocker-bottom deformity [3, 4]. The rocker-bottom deformity has not been reported by Professor Ponseti as one of the complications in his clubfoot studies. That is why it is regarded as an iatrogenic disease.

However, favorable clinical results not only depend upon the strict adherence to the Ponseti protocol, but also rely on detailed quality controls throughout all three stages of intervention including cast, tenotomy and brace management [8, 9]. Nevertheless, one study of radiographic images during Ponseti treatment of clubfoot has identified 7 patients presenting with rocker-bottom deformity among 87 cases of Ponseti treatment for clubfoot. Among the 7 cases, 6 feet were significantly improved after Achilles tendon release and only one foot was

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not effectively corrected [10]. Because the feet of infants mostly consist of cartilage, plain X-ray can only provide extremely limited information. In addition, X-ray evaluation of infantile feet should be performed with caution due to the damage caused by radiation exposure [11, 12]. Plain X-ray of the feet in the maximum dorsiflexion position has been proven to provide significant guidance for the treatment of infantile clubfoot [13-15]. The incidence of mild rocker-bottom deformity during Ponseti treatment has been identified by X-ray evaluation of the feet [10], suggesting that plain X-ray of the feet can identify potential abnormalities that are difficult to detect by clinical observation, such as mild rocker-bottom deformity.

We hypothesize that the risk of rocker-bottom deformity occurring during Ponseti treatment still exists. Nevertheless, the underlying mechanisms are still unknown, and relevant clinical manifestations and risk factors have been rarely studied. Therefore, the present study aimed to investigate the clinical and radiographic manifestations of rocker-bottom deformity during Ponseti management of idiopathic clubfoot and the preliminary treatment results were presented as follows.

Materials and methods

Study design

In this retrospective study, clinical data of children diagnosed with clubfoot admitted to the clubfoot specialty outpatient clinic at Shanghai Children's Medical Center between August 1, 2015, and July 31, 2016 were retrospectively analyzed. The inclusion criteria were: 1) diagnosis with congenital clubfoot; and 2) a brace is being fitted during Ponseti treatment. The exclusion criteria were: 1) complications with other musculoskeletal system disorders; 2) those with clear nervous system disorders; 3) those receiving other interventional therapies; and 4) those failing to complete follow-up treatment or with incomplete records from at least 1-year follow-up. The study was approved by the ethics committee of the Shanghai Children's Medical Center. Written informed consent was obtained from the parents of patients.

Treatment procedures

After notifying the family members and obtaining their informed consent, the following treat-

ments were performed to confirm the diagnosis of rocker-bottom deformity in the pediatric patients. 1) For continuous fixation, arch support clubfoot plaster casts were changed once per week for 4 consecutive weeks to restore the midfoot alignment. 2) Under general anesthesia, fluoroscopy of the lateral talo-navicular joint was performed with the feet in the maximal dorsiflexion position. If the talo-navicular joint was dorsally dislocated, it was manually reduced and fixed with a 1.6-mm diameter Kirschner wire. Then, percutaneous Achilles tenotomy was performed, as described by Dobbs *et al.* [16]. If the talo-navicular joint was properly aligned, the percutaneous Achilles tenotomy alone was sufficient. 3) The ankle joint was held in a neutral position with a long-leg cast with dorsiflexion at 15° and arch support cast for 4 consecutive weeks. The hardware was removed under local anesthesia, and another cast was placed for 2 consecutive weeks. Those without internal-fixation were fixed with the final cast for 6 consecutive weeks. 4) The original Ponseti external brace was refitted. Brace management was adopted initially for 23 h/day for 3 months, followed by wearing only while sleeping for at least 2 years.

Follow-up

After corresponding treatment, all patients were subject to follow-up once a year for 2 years. During the follow-up, the appearance and function of bilateral feet were evaluated. The compliance of wearing the brace was also observed and recorded. Lateral radiographs were taken starting at 6 months after corresponding treatment for 2 years, and then once a year for 3 years afterwards.

Evaluation methods

The Dimeglio score system has become the most universally adopted classification system of the severity of deformity [17]. The independent sagittal and total scores were recorded. Medical history of each patient was extracted from the medical charts.

During the physical examinations of the ankle dorsi-flexion, forefoot position was carefully checked in the neutral and supination positions. If the dorsiflexion angle in the forefoot supination position was smaller compared with that in the forefoot neutral position, the patient is suspected to have rocker-bottom deformity.

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Figure 1. Radiograph of rocker-bottom deformity. A. Lateral radiograph of the feet in the maximal dorsiflexion position. B. Radiograph of rocker-bottom deformity. C. The measurement of the tibio-calcaneal (Ti-C), calcaneo-fifth metatarsal (C-M5), and talo-first metatarsal (Ta-M1) angles.

Hence, radiographic examinations of this patient should be considered.

For radiographic examinations, lateral radiographs of bilateral feet were taken with the feet in the maximal dorsiflexion position (**Figure 1A**). Projection methods were conducted by using the lateral foot radiography technique as previously described [18]. Each of the two feet was supported and maintained with the maximal flexion of the ankle joint by the parents or guardians of the patient. After radiography, the talo-first metatarsal (Ta-M1), calcaneus-fifth metatarsal (C-M5) and tibio-calcaneus (Ti-C) angles were measured for subsequent evaluation (**Figure 1B, 1C**).

If the Ti-C angle was more than 90 degree, the Ta-M1 angle or C-M5 angle was either or both more than 15 degrees, the diagnosis of rocker-bottom deformity could be confirmed. If the diagnosis of rocker-bottom deformity was confirmed, the medical history of clinical management was reviewed with the family members. Several detailed questions were asked including the age of initial treatment, number of casts and whether the percutaneous Achilles tenotomy was performed. The Dimeglio classification system with images was explained to the parents to explain the infantile deformity before the treatment. If the score is below 5, the foot will be classified as normal (N). If the score is between 5 and 10, the foot will be classified as mild deformity (M). If the score is between 11 and 15, the foot is classified as severe deformity (S). If the score is more than 16, the foot is classified as very severe deformity (VS).

Statistical analysis

SPSS 16.0 (IBM, Armonk, NY, USA) was used for statistical analyses. Continuous variables were expressed as mean \pm standard deviation

(SD). Paired *t*-test was utilized to statistically compare the differences between the angles measured before and after treatment. A *P* value of less than 0.05 was considered as statistically significant.

Results

Baseline characteristics

During the study, 276 feet from 185 clubfoot children were examined at the outpatient clinic of Shanghai Children's Medical Center. Among them, 15 feet from 10 children were clinically suspected with rocker-bottom deformity evaluated by radiography. Eventually, the diagnosis of rocker-bottom deformity was confirmed in 13 feet from 6 children. All these 6 children were transferred to our hospital from local hospitals. The medical history of Ponseti treatment was carefully reviewed by discussing the details with their family members, as illustrated in **Table 1**.

Treatment outcomes

Prior to treatment, the severity of rocker-bottom deformity was preliminarily determined according to the Dimeglio scores as severe (11-15 points) or very severe (16-20 points) based on the descriptions and images provided by their family members. Plaster casts were used for external fixation for roughly 3.6 times on average. Achilles tendon release was performed on 4 feet from 3 children.

Follow-up

At least a 1-year follow-up was conducted in 6 children (9 feet). Radiologic results of the feet validated the diagnosis of rocker-bottom deformity. Radiologic measurements of the Ti-C, C-M5 and Ta-M1 angles were carried out. The

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Table 1. Baseline information of enrolled children

Patient	Sex	Age (months) upon Ponseti treatment	Age (months) upon the detection of the deformity	Laterality	Number of casts	Tenotomy	Dimegilo classification before casting	Follow-up duration (months)
1	Male	7	12	Left Right	4	Yes	Very severe	24
2	Female	1	14	Right	4	Yes	Very severe	23
3	Male	0.25	12	Left	2	No	severe	14
4	Female	0.25	12	Right	3	Yes	severe	23
5	Male	0.5	5	Left Right	4	No	Very severe	23
6	Female	0.25	19	Left Right	4	No	Very severe	12

Follow-up treatment and complete records from at least the 1-year follow-up were available for 9 feet in 6 patients. Of the 6 patients, 3 cases were male and 3 female, aged 10.4 months on average (range: 4-19 months). Of the nine affected feet, rocker-bottom deformity was observed in the 5 right and 4 left feet.

Ti-C angle was measured between 59.5° and 117.0° (mean value: 82.4±15.1°), 21.0°-48.5° for the C-M5 angle (mean value: 34.8±6.2°) and 24.8°-63.7° for the Ta-M1 angle (mean value: 43.8±13.4°), respectively (Tables 2, 3).

As illustrated in Tables 2, 3, the Ti-C, C-M5 and Ta-M1 angles were significantly altered before and after treatment. After the operation, the Ta-M1 and C-M5 angles were considerably improved (both $P < 0.001$). At 1-year follow-up, the Ti-C angle ($P < 0.001$), Ta-M1 angle ($P < 0.05$) and C-M5 angle ($P < 0.05$) were remarkably improved compared with the angles before surgery.

Arch support clubfoot plaster casts were utilized four times on the nine feet. Of these, the midfoot of one foot was determined to be stable during the procedure. Simple percutaneous Achilles tendon release was performed, and the foot was fixed in a neutral position in a plaster cast for 3 weeks. The midfoot was unstable during the procedure for the remaining eight feet. Kirschner wire internal fixation of the talonavicular joint was performed with adjuvant percutaneous Achilles tendon release, and the foot was fixed in a neutral position in a plaster cast for 4 weeks and fixed in a plaster cast for an additional 2 weeks after hardware removal. During subsequent follow-up, the Ponseti brace was used initially for 23 h/day for 3 months, followed by wearing only while sleeping.

During the final follow-up, radiograph of the nine feet with maximal dorsiflexion or lateral

radiograph with weight bearing was obtained for evaluation. Of the nine feet, eight were restored to the central axis. One female infant (No. 2) who began receiving Ponseti treatment at the age of one month had 4 casts with Achilles tenotomy. She came to our hospital because she kept slipping out from the brace and was diagnosed with rocker-bottom deformity at the age of 14 months. The lateral radiograph images were illustrated in Figure 2.

Discussion

Rocker-bottom deformity is a complication which occurs after nonsurgical treatment of clubfoot with an incidence rate of 3.2% worldwide [19]. However, how to identify and confirm the diagnosis of rocker-bottom deformity has been rarely investigated. Specifically, the diagnostic criteria of rocker-bottom deformity have not been described before. Similar or mild deformity has been defined as pseudocorrections in a radiography study of Ponseti treatment [20].

In the present study, the incidence rate of rocker-bottom deformity treated by Ponseti method in was 4.32% (8/185) by patients and 4.71% (13/276) by clubfeet. The diagnosis of rocker-bottom deformity could be validated if the tibio-calcaneal angle was more than 90 degrees in the meanwhile the talo-first-metatarsal angle or calcaneo-fifth-metatarsal angle was either or both >15 degrees on the radiography. This study was the first to report this iatrogenic problem during Ponseti treatment in the Chinese children in clinical practice. Moreover,

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Table 2. Radiologic results of all children at different time points

Patient	Laterality	Pre-operation			Post-operation (°)			Final follow-up (°)		
		Tibiocalcaneus angle (°)	Talo-1st metatarsal angle (°)	Calcaneo-5th metatarsal angle (°)	Tibiocalcaneus angle (°)	Talo-1st metatarsal angle (°)	Calcaneo-5th metatarsal angle (°)	Tibiocalcaneus angle (°)	Talo-1st metatarsal angle (°)	Calcaneo-5th metatarsal angle (°)
1	Left	84.25	20.6	42.1	71.12	8.39	8	71	39.3	26.5
	Right	85.8	39.3	36.1	76.7	1.2	3.5	75.7	28.8	32.9
2	Right	79.89	46.81	30.2	87.5	3.8	14.6	60.1	14.1	34.88
3	Left	84.44	31.3	45.1	102.27	9	33.8	80.2	22	35.1
4	Right	90.2	39.4	26.5	118.53	2.1	19.4	81.6	32.4	26.3
5	Left	52.68	63.45	35.67	82.3	11.8	12.5	43.45	31.1	20.5
	Right	66	60.1	34.5	80.9	2.6	4.2	62.3	26.1	13.1
6	Left	102.3	50.26	27.35	84.2	6.4	14.1	84.86	15.8	15.8
	Right	96.13	42.68	35.38	78.3	1.2	9.3	81.9	15.2	0.3

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Table 3. Radiological angle measurement at different time points

Angle measurement	Pre-operation (°)	Post-operation (°)	Final follow-up (°)
Tibiocalcaneus angle	82.4±15.1	86.9±14.7	71.2±13.6**
Talo-1st metatarsal angle	43.8±13.4	5.2±3.9**	25.0±8.8*
Calcaneo-5th metatarsal angle	34.8±6.2	13.3±9.2**	22.8±11.6*

* $P < 0.05$ and ** $P < 0.001$ vs. preoperative measurement results.



Figure 2. Lateral radiograph of a female patient 14-months old. A. Before treatment. B. One day after operation. C. 23 months after operation.

it was also the first to identify this deformity quantification on radiograph.

Clinical efficacy and safety of Ponseti treatment definitely depends upon the strict adherence of the protocols and detailed quality control of practice in every stage throughout the whole management [8, 9]. This might explain the existence of this iatrogenic problem. The other reasons why the incidence rate of rocker-bottom deformity reported in this study was slightly higher than previous investigations probably ascribed to the following limitations of this study including the small sample size, the uncertain definition of rocker-bottom deformity, as well as low awareness of rocker-bottom deformity. In spite of these limitations, this study also adds evidence to the high requirement of quality control during the education as well as the practice throughout Ponseti treatment.

In clinical practice, how to treat rocker-bottom deformity or pseudocorrection deformity is still controversial. In previous reports of pseudocorrections, a large angle of correction can be obtained with repeated Achilles tendon release after arch support plaster cast fixation [20]. In the present investigation, arch support plaster fixation of the clubfoot was also performed in the 9 children. Fluoroscopy confirmed the midfoot stability. Simple percutaneous Achilles tendon release was performed in only one

patient. For the remaining eight feet, the midfoot was still unstable, and percutaneous hardware fixation was adopted to stabilize the joint in these patients, which is similar to the method for early treatment of patients with vertical talus reported by Dobbs *et al.* [16].

In this study, application of the cast alone failed to increase the midfoot stability, probably due to the older age of the children, longer sustained time of residual deformity, or uncorrected clubfoot in the hindfoot at the time the clubfoot brace was used. Nevertheless, the brace use still could maintain the foot in the dorsiflexion of the forefoot. Percutaneous hardware fixation of the talo-navicular joint allows certain patients to achieve the midfoot stability. Percutaneous Achilles tendon release can also eliminate a driving factor for the progression of rocker-bottom deformity [16]. The above-mentioned factors are probably correlated with the excellent short-term results obtained in seven of eight feet in the present investigation.

One patient was not effectively treated, probably because the older age of the child and the higher severity of the midfoot deformity. Upon the date of manuscript submission, this patient was still under direct observation. The possibility of further surgical correction cannot be eliminated in the future. This case promotes that early detection and diagnosis of rocker-bottom

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deformity plays a significant role in enhancing clinical prognosis.

Taken together, the severity of rocker-bottom deformity, lack of percutaneous Achilles tendon release, and difficulty in both fitting the brace and wearing it are key processes throughout Ponseti treatment. In suspected clinical cases, lateral radiograph of the feet with the ankle joint in the maximal dorsiflexion can provide supplementary information for accurate diagnosis.

Disclosure of conflict of interest

None.

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