Results of Primary Talectomy for Clubfoot in Infants and Toddlers with Arthrogryposis Multiplex Congenita

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Background: *Equinovarus deformity in arthrogryposis multiplex congenita patients is rigid and difficult to treat. Radical soft tissue operations yielded good results though recurrence of deformity was high. Talectomy is a bony procedure recommended as either a salvage procedure or a primary operation.*

Objective: *To evaluate the results of primary talectomy in infant and toddler patients retrospectively.*

Material and Method: *Arthrogryposis multiplex congenita patients with rigid equinovarus deformity treated with talectomy initially were retrospectively reviewed. Pain score, residual foot deformity, shoe modification, and ambulatory status were assessed.*

Results: *Talectomy were performed in 19 arthrogrypotic feet in 10 infants and toddlers. There were 6 males and 4 females. The mean age at surgery was 1.3 years old and the mean age of the follow-up time was 4.9 years. All patients had plantigrade foot without pain. One arthrogrypotic foot required posteromedial release 2 years after index surgery due to recurrent deformity.*

Conclusion: *Talectomy as the primary procedure in arthrogrypotic infants and toddlers demonstrated good results with few residual deformities and recurrence. Patients could achieve plantigrade position.*

Keywords: *Infant, Toddler, Arthrogryposis multiplex congenita, Clubfoot and talectomy*

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Arthrogryposis multiplex congenita is one of the most common disorders of the multiple congenital joint contracture syndromes. This disorder involves multiple joints including hip, knee, ankle, foot, shoulder, elbow, wrist and hand[1]. The ankle and foot are the most commonly affected areas and the equinovarus deformity is the most common deformity[2]. Equinovarus or clubfoot deformity in arthrogryposis multiplex congenita is usually rigid and resistant to manipulation. Radical soft tissue operations provided good results especially in children under 1 year old[3]. However, this operation has high recurrent rate[4-7]. Talectomy is a salvage procedure for severe rigid clubfoot and is recommended as a primary procedure by many authors[4-10]. Talectomy would improve equinovarus deformity, whereas correction of deformity without tension could be easily achieved. Primary talectomy also provided better results compared to salvage talectomies especially in younger children[5,6]. Hsu et al[8] stated good results and functional feet in all patients undergoing talectomy after the follow-up mean of 8 year olds. Cassis and Capdevila[6] reviewed 101 talectomies in 56 patients. Good results could be obtained in 63% of cases despite that most patients in the study had previous procedures. Other procedures such as tibiocalcaneal fusion, calcaneocuboid fusion or naviculectomy may also be added to reduce postoperative recurrence deformity and pain[4,11,12].

The purpose of this study was to evaluate the functional results of talectomy in arthrogrypotic clubfeet.

Material and Method

The study population included 0.5-3-year old, arthrogrypotic patients undergoing talectomy as single or combined procedure at Faculty of Medicine, Siriraj Hospital, Mahidol University between 2000-2013. Data collection included demographic data consisting of gender, age and affected sides. Pre-operative and postoperative ambulatory statuses were defined as

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Table 1. Individual data

ID	Age at surgery (years)	Gender	Involved limb	AMC type	Surgery	F/U (years)	Additional surgery	Shoe modification
1	2.33	Female	Both	Distal	Talectomy	2.75	None	None
2	1.33	Male	Both	Distal	Talectomy with naviculectomy	9.75	None	None
3	3.00	Male	Both	Classic	Talectomy	4.50	None	None
4	1.33	Male	Both	Classic	Talectomy	4.83	None	None
5	0.58	Female	Both	Classic	Talectomy	6.50	None	None
6	0.50	Female	Both	Classic	Talectomy	5.83	PMR	Needed
7	0.58	Female	Left	Classic	Talectomy	5.25	None	None
8	1.83	Male	Both	Classic	Talectomy	3.67	None	Needed
9	1.17	Male	Both	Classic	Talectomy	2.08	None	None
10	0.50	Male	Both	Classic	Talectomy	3.50	None	None

AMC = Arthrogypoisis multiplex congenita; PMR = Posteromedial release

Table 2. Summary data

Factors	Population
Male: female	6:4
Preoperative ambulatory status	
Community	0
Household	1
Nonfunctional	1
Nonambulator	-
Nonspecified	8
Age at surgery (years)	1.32 (0.5-3)
Simultaneous procedure	1 Naviculectomy
Complications	
Infection	0
Reoperation	1 posteromedial release
Follow-up period (years)	4.9 (2-9.8)
Postoperative ambulatory status	
Community	6
Household	2
Nonfunctional	2
Nonambulator	-
VRS at last F/U	0
Shoe modification needed	2/10
Residual foot deformity	10/10 plantigrade foot

Values are number (percentage) and the mean (min-max)

community ambulators, household ambulators, non-functional ambulators and non-ambulators. Simultaneous procedures and complications were also evaluated. At the last follow-up, verbal rating scale (VRS) pain score, shoe modification and residual foot deformity were graded.

Results

Nineteen arthrogrypotic feet in 10 patients were evaluated. Nine patients had bilateral involvement while one patient had left foot-involvement. Most patients were classic AMC. There were 6 males and 4 females. Eight patients could not be classified for any group (ergo, non-specific group) because they were less than 2 years old.

Primary talectomy alone were performed in 18 arthrogrypotic feet. Talectomy with naviculectomy in one arthrogrypotic foot was justified. One patient required posteromedial release in 2 years later. The follow-up mean was 4.9 years. Six patients (60%) became community ambulators while two patients were capable of household ambulators and non-functional ambulators equally. Two out of ten patients required shoe modification to improve walking ability while most patients could wear normal shoes.

Discussion

Treatment of arthrogrypotic clubfoot deformity is controversial. Goal of the treatment is to provide stable, plantigrade, pain-free feet, though radical soft tissue release[3] and Ponseti technique[13-15] have been used successfully to treat these patients. Talectomy has been recommended as primary or salvage operation and especially after clubfoot relapse[4-6,8,9,16]. Legaspi et al[9] reviewed twenty-year follow-up of 24 recurrent clubfeet with

75% fair to good results after talectomy. In cases of recurrent equinovarus deformity, tibiocalcaneal fusion was a salvage operation after talectomy. Tibiocalcaneal fusion could improve patients' overall function and pain. Calcaneocuboid fusion combined with talectomy decreased rates of recurrent deformity and less pain in severe rigid clubfoot[11]. Naviculectomy could be combined with talectomy in the cases of inadequate correction after talectomy alone[4]. Most studies[2,3,6,8,9,13] reported good outcome in the short and medium follow-up time. The longest follow-up study[9] demonstrated fair to good results in 75% of patients at the follow-up mean of 20 years old. Recurrent rate and tibiocalcaneal arthritis were still high (67% at 10-year follow-up and 33% at 8 to 10-year follow-up).

Primary talectomy may be appropriate in some situations such as low compliance for weekly follow-up or in rural area. The presented data demonstrated good results after primary talectomy with few residual deformities and low recurrence. The improvement of ambulatory status of the patients was evident. The authors hypothesized that ambulatory status may not associate with existence or severity of deformity but this improvement may be due to increasing age. The present study could not demonstrate the benefit of concomitant naviculectomy with primary talectomy because it was performed in only one of nineteen feet. One of 19 (5.3%) arthrogrypotic clubfeet in this study needed revision surgery 2 years after index surgery. This low recurrence may be due to younger age of the study population and the fact that most of the patients underwent talectomy as primary procedure. Longer follow-up is needed because the follow-up mean of this study was 4.6 years old. Despite this fact, primary talectomy is a reliable procedure for treating rigid equinovarus deformity.

Conclusion

Primary talectomy could be a suitable alternative option for rigid clubfeet in arthrogrypotic infants and toddlers and low recurrence could be expected.

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Potential conflicts of interest

None.

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ผลการรักษาโรคเท้าปุกโดยวิธีการผ่าตัดเอากระดูกทาลัสออกในผู้ป่วยอาไทรไกรโพซีส มัลติเพล็กซ์ คอนเจนิตา ในเด็กทารกจนถึงวัยหัดเดิน

จตุพร โชติกวณิชย์, ธเนศ อริยะวัตรกุล, พีระจิตร เอี่ยมโสภณา, กมลพร แก้วพรสวรรค์

ภูมิหลัง: *ภาวะเท้าปุกในผู้ป่วยอาโทรไกรโพซีส มัลติเพล็กซ์ คอนเจนิตาในเด็กเป็นภาวะที่ยากลำบากต่อการรักษา การรักษาโดยการผ่าตัดแก้ไขโดยผ่าตัดเลาะพังผืด มีอัตราการกลับเป็นใหม่ค่อนข้างสูง การผ่าตัดเอากระดูกทาลัสออก มักทำได้ในเท้าที่ผ่านการรักษาด้วยวิธีการผ่าตัดเลาะพังผืดมาแล้วหรืออาจทำได้ในการผ่าตัดแก้ไขครั้งแรกในเด็กโต*

วัตถุประสงค์: *รายงานผลการรักษาโดยวิธีการผ่าตัดโรคเท้าปุกโดยวิธีการผ่าตัดเอากระดูกทาลัสออกในผู้ป่วยอาโทรไกรโพซีส มัลติเพล็กซ์ คอนเจนิตา ในเด็กทารกจนถึงในวัยหัดเดิน คือที่อายุน้อยกว่าประมาณ 3 ปี*

วัสดุและวิธีการ: *การศึกษาย้อนหลังผู้ป่วยที่มีภาวะเท้าปุกในผู้ป่วยอาไทรไกรโพซีส มัลติเพล็กซ์ คอนเจนิตา ที่ได้รับการผ่าตัดโดยวิธีการผ่าตัดเอากระดูกทาลัสออกโดยเก็บข้อมูลของอายุ เพศ คะแนนความเจ็บปวด ภาวะเท้าที่ผิดรูปที่เหลืออยู่ ความสามารถในการใส่รองเท้าแบบปกติและการเดิน*

ผลการรักษา: *ผู้ป่วย 10 ราย 19 เท้า ที่ได้รับการรักษาโรคเท้าปุกโดยวิธีการผ่าตัดเอากระดูกทาลัสออกในผู้ป่วย อาโทรไกรโพซีส มัลติเพล็กซ์ คอนเจนิตา เป็นเด็กชาย 6 ราย เด็กหญิง 4 ราย อายุเฉลี่ยที่ได้รับการผ่าตัด 1.3 ปี (0.5-3 ปี) ระยะเวลาติดตามเฉลี่ย 4.9 ปี ผู้ป่วยทุกรายสามารถลงน้ำหนักเท้าในสภาวะแพลติเกตได้โดยไม่เจ็บ มีหนึ่งเท้าต้องได้รับการผ่าตัดแก้ไขเนื่องจากการกลับเป็นซ้ำ*

สรุป: *ผลการรักษาโรคเท้าปุกโดยวิธีการผ่าตัดเอากระดูกทาลัสออกในผู้ป่วยอาโทรไกรโพซีส มัลติเพล็กซ์ คอนเจนิตาในเด็กที่อายุน้อยกว่า 3 ปี ได้ผลดี ถึงแม้ว่าอาจมีรูปเท้าผิดปกติเล็กน้อยหลงเหลืออยู่บ้าง*
