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RESEARCH ARTICLE

The treatment of neurotrophic foot and ankle deformity of spinal bifida: 248 cases in single center

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ABSTRACT

Objective: To evaluate the functional outcome of foot and ankle deformity secondary to spinal bifida treated by various methods.

Methods: A retrospective analysis of 248 patients with foot and ankle deformity secondary to spina bifida with an average age of 25.5 years and underwent surgical treatment in our hospital from March 2012 to April 2016. The deformity correction was achieved by various methods like soft tissue procedure, bony procedure combined with external fixator application. All the patients were followed up at regular interval. Post operative rehabilitation and protective splints were provided after fixator removal. The final outcome was evaluated with American Orthopedic Foot and Ankle Society (AOFAS) score and Qin's criteria for deformity correction, matched T-test was used to compare the data pre and post surgery.

Results: According to the short term follow up, all these patients were achieved complete correction and able to achieve full weight bearing. Out of 248 patients, 13 patients were lost follow up. 235 patients were followed up for an average of 28.5 months. We noted various minor complications like superficial pin tract infection was seen in 5, pin breakage in 4, pin tract burn injury in 1, and recurrence of deformity was noted in 20 patients especially in children, anterior dislocation of the tibiotalar joint in 3 patients with severe clubfoot deformity. At the final follow up, the mean AOFAS score increased to 88.7, with a significant improvement compared with the score before surgery ($P < 0.05$). Based on Qin's criteria for deformity correction, the outcome was graded as excellent in 180 patients, good in 55, and fair in none of the cases.

Conclusion: Through orthopaedic treatment, combination of soft tissue and bony procedures along with external fixator helps to achieve complete correction of deformity, healing of ulcer, restoration of functional activity for spinal bifida sequelae patients.

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1 Introduction

Spina bifida is the most common congenital anomaly of the central nervous system [1]. The level of the lesion determines the type of foot and ankle deformity [2]. The involvement of the foot and ankle in spina bifida is seen in most of the patients [3, 4] which may present as club foot, calcaneus foot, equinus, varus, valgus or a combination of deformities.

Foot and ankle deformities affect the cosmetic appearance, difficulty in ambulation, difficulty in bracing and shoe wear. These deformities can also lead to skin irritation due to abnormal loading and result in pressure sore [5]. It is a main factor which affects the quality of life in spina bifida sequelae patients.

Therapeutic management of spina bifida anomalies is not uniform currently [6]. Neuro-surgeons believe that neurolysis should be performed [7], spinal surgeons support skeletal fixation and fusion [8], and recent research studies were performed with cell therapy [9] in order to find out neurorestorative methods. As orthopaedic surgeons, in this study we had summarized our experience in the management of foot and ankle deformities in spina bifida sequelae patients.

2 Materials and methods

2.1 Patients

In this study 248 patients with foot and ankle deformity secondary to spina bifida admitted in our hospital orthopaedic department from March 2012 to April 2016 were retrospectively analysed. Out of 248, 121 were male and 127 were female. The patients included in this study were aged 5 to 50 with an average of 25.5 years old. The side of involvement was left sided in 49 patients (19.8%), right sided in 53 patients (21.4%) and bilateral in 146 patients (58.8%). Inclusion criteria: All patients with acquired foot and ankle deformity secondary to spina bifida of any age group. Exclusion criteria: Patient with congenital foot and ankle deformity, foot and ankle deformities as sequelae of poliomyelitis, cerebral palsy. Detailed history of acquired foot and ankle deformities, any sensory impairment, any bowel and bladder involvement were obtained. Thorough evaluation of lumbosacral region to look for any cystic lumps, skin depression, pigmentation, hairy tuft, any previous operated scar, type of deformity of foot and ankle, muscle power, degree of sensory involvement, any ulceration were performed. Radiographic evaluation of lumbosacral region was done in all patients to assess the level of spinal lesion. An informed consent was obtained from all the patients involved in this study.

2.2 Surgical method

All the patients were operated by the same team of orthopaedic surgeons under general anaesthesia. The surgical plan was designed based on initial clinical evaluation, component of involvement soft tissue or bony component, type of deformity, and suppleness of the deformity. For only soft tissue involvement, tendoachilles lengthening, tibialis posterior lengthening or transfer, tibialis anterior transfer was performed based on the type of deformity. For bony involvement, corrective extra articular osteotomies and subtalar arthrodesis were performed. After initial correction, external fixator was applied to all the patients. Hybrid fixator was used for acute correction. If the deformity cannot be corrected acutely, then Ilizarov fixator was used for gradual correction of the deformity.

2.3 Post operative management

In the initial 1st to 4th postoperative day, patients were advised to do exercise in bed. Empirical antibiotic was given to all the patients. Wound

inspection was done on the 4th postoperative day and postoperative radiograph were taken. Those patients who had residual deformity gradual correction by frame adjustment were started on the 5th postoperative day. All patients were mobilised initially with crutches, transited from non-weight bearing walking gradually to partial weight bearing, and at last full loading was allowed. All the patients were followed up at regular intervals. Radiographs were taken at each visit to assess the bone healing. After achieving complete correction of the deformity, the fixator was locked in that position and allowed full weight bearing. Those patients who had ulceration were treated by regular dressing every 1 or 2 days. Once the signs of bony union noticed in follow up radiograph, the external fixator was removed. Then custom made ankle foot orthosis was provided and allowed to walk with full weight bearing. After 4 to 8 weeks, patients were gradually transited to unprotected walking.

At final follow up, the results were assessed by AOFAS (American Foot and Ankle Society) score [10] and Qin's criteria for the deformity correction [11] (Table 1). Matched T-test was applied in SPSS 20.0 software to compare the score preoperatively and at the final follow up.

3 Results

Out of 248 patients, 13 patients were lost follow up. 235 patients were followed up for an average of 28.5 months. In terms of the type of deformities of foot and ankle, there were 134 patients with clubfoot, 30 with valgus foot, 37 with calcaneal foot, and 34 with cavus foot. 43 patients had ulcer in the weight bearing area. 9 patients had hip dislocation and 12 had scoliosis as associated deformity which was treated accordingly. According to the type of external fixator, Ilizarov external fixator in 115 cases (48.9%) and 107 cases (45.6%) were treated with Hybrid external fixator. 11 cases were treated with both kinds of fixator. Plaster fixation was used for the other 2 patients.

All the patients achieved complete correction of deformity with plantigrade foot and able to restore full weight bearing without serious complications such as neurovascular injury, deep infection or amputation.

However minor complications occurred in a few patients. 20 patients developed recurrence of foot and ankle deformity notably, and all these patients were paediatric age group who had remnant growth period. These patients were all treated accordingly and achieved full correction of deformity, and AFO (Ankle and Foot Orthosis) was given for long duration for the maintenance of correction and prevention of recurrence. Superficial pin tract infection was noted in 5 adult patients who were treated with regular pin tract dressing and appropriate antibiotics. Wire breakage was noted in 4 patients, all of which occurred in forefoot and was treated with replacement of broken wire by new wire insertion into the frame. Pin track burn injury was observed in 1 patient at the calcaneal region which healed up after regular dressing. 1 patient had allergic eczema related to external fixator. 3 patients with severe clubfoot deformity developed anterior dislocation of tibiotalar joint which was corrected successfully by changing the configuration of external fixator. The ulceration in weight bearing area of foot recurred in 2 patients and was healed after second surgery.

At the final follow up, the mean AOFAS score increased to 88.7 post operatively. Based on Qin's criteria for deformity correction, the outcome was graded as excellent in 180 patients, good in 55 and none were fair or poor. By comparing the pre and post operative AOFAS score using matched T-test, significant improvement ($P < 0.05$) was observed (Tables 1 and 2).

Table 1 Qin's postoperative evaluation criteria for lower limb reconstruction.

Name	Gender	country	Age	Age of follow-up	
Address				Phone number	
Cause of deformity Deformity : Osseous	Soft tissue contracture	Combined deformity	Limb shortening		
Gait (preoperative) Crawling Squatting	Crutches	Single crutch	Severe limp	Moderate limp	Mild limp Normal
Operation procedures and times:					
Remarks	Physical therapy: Reoperation: YES No				
_____ years _____ months _____ days after the last surgery					
Evaluation of muscle transfer surgery	Unbalanced muscle (preoperative)				
	Strength balance	Excellent	Well	Good	Poor
	Scores	3	2	1	0
Evaluation of deformity correction(X-ray)	Deformity part and degree (preoperative)				
	Deformity correction	Excellent	Well	Good	Poor
	Scores	3	2	1	0
Evaluation of walking ability	Disabled part				
	Improvement	Excellent	Well	Good	Poor
	Scores	3	2	1	0
Patient's self-evaluation	(Parents represent for children)				
	Self-evaluation	Excellent	Well	Good	Poor
	Scores	3	2	1	0
Complications	(E.g. nerve injury, anchyloses, severe infection, osteotomy nonunion etc.)				
	Complication degrees	Excellent	Well	Good	Poor
	Scores	3	2	1	0
Average scores	2.5		2		1
	Excellent	Well		Good	Poor
Cause of complication:					
Surgery performed by: Follow-up date:		Followed by:			

The patients were evaluated in terms of muscle strength, deformity correction (including imaging), assessment of motor and walking function, self-perception of surgical effect, and complications & problems, and so on. Each item was divided into 4 levels.
For patients with bony surgery but without tendon transfer, 4 items were applied when follow-up, the total score divided by 4. For patients with compound surgery, 5 items were applied when follow-up, total score divided by 5.
Average scores: > 2.5, excellent; > 2, well; > 1, good; < 1, poor.

Table 2 The pre and post operative AOFAS scores.

	Preoperative score (n)	Postoperative score (n)	P value
Clubfoot	67.05 ± 5.5 (134)	86.16 ± 3.62 (134)	0.046
Valgus foot	70.45 ± 10.3 (30)	85.45 ± 6.55 (30)	0.039
Calcaneal foot	68.46 ± 7.3 (37)	83.41 ± 5.42 (37)	0.035
Cavus foot	72.26 ± 4.7 (34)	84.92 ± 5.18 (34)	0.045

$P < 0.05$, significantly different.

Table 3 The outcome based on Qin's criteria for deformity correction.

Category	Excellent	Good	Fair	Poor
Clubfoot	97	37	0	0
Valgus foot	26	4	0	0
Calcaneal foot	32	5	0	0
Cavus foot	25	9	0	0
Total	180	55	0	0

4 Discussion

Spina bifida is a developmental defect caused by an incomplete closure of the embryonic neural tube during the first 3~4 weeks of gestation. This developmental vertebral defect may lead to motor and/or sensory loss below the lesion causing variable spasticity, paralysis, and muscle imbalance of the lower extremities [12]. Different spinal segment defects cause different types and degrees of foot and ankle deformities, and bowel and bladder disturbances [2]. The treatment of this clinical syndrome involves pediatrics, neurosurgery, urology, orthopaedics, rehabilitation and other disciplines, each one with their indication achieved good result based on their principles [13].

Orthopaedic method [14–16] is useful in the treatment of various deformities occur in these

Fig. 1 A typical case: female, 18 years old, bilateral equinovarus foot deformities of spinal bifida sequela. (A) Equinovarus foot deformity bilateral sides. (B) X-ray showed vertebral defect in lumbosacral region. (C) The operation included posterior tibia muscle lengthening, the triple joint osteotomy and fixation with an Ilizarov fixator gradually correcting the deformity to neutral position. AFO was applied when fixator removal. (D) 2 years follow-up, her foot and ankle deformities were corrected completely.

spinal bifida patients and help to improve the functional quality of life. In addition to tendon release and bony osteotomy, external fixation has obvious advantages over other fixation methods like plaster, brace, and internal fixation. The external fixation is not only simple, adjustable and minimally invasive, but also suitable for various types and degrees of deformities. It is the result of method optimization. The choice of external fixator configuration is related to the degree of deformity of foot and ankle, and the extent of deformity correction. After limited bony or soft tissue surgery, hybrid fixator was usually used for acute correction. If the deformity could not be corrected fully, the correction of residual part relay on Ilizarov ring fixator, which can be done without adjustment or fine adjustment until fixator removal.

Ilizarov technique [17] is a representative of external fixation for the treatment of complex fracture, severe limb deformity and function reconstruction. It also creates a vast space in the treatment of complex foot and ankle deformity of spina bifida sequela. Patients could walk partial to full weight bearing under the control of external fixation. Some complications of skin and soft tissue necrosis, and neurovascular injury could be prevented as much as possible.

Lee et al. [18] evaluated the role of Ilizarov fixator in correction of rigid neurologic equinocavovarus foot deformities. They found that Ilizarov allows a greater degree of correction even in rigid foot deformities, and can minimize the further surgical intervention and scarring.

External fixation has some disadvantages in the treatment of lower limb deformities of spina bifida. Burn injury of pin tract is also a problem that needs to be paid attention in clinic practice. The wire can be broken during the walking weight bearing, which may result in below-expected bone healing effect and delayed bone formation in neurogenic diseases. Fatigue breakage under long-term abnormal stress occurred on the wires and pins of external frame. Complications above can be avoided or minimized by standard installation of surgical procedures.

Regarding the limitation of this observed paper from a single center, in which patients suffered from spinal bifida and underwent tethered surgery, we hope the experience of deformity correction treatment could be confirmed by more centers.

When designing the plan for treatment of foot and ankle in spinal bifida sequela, we should pay attention to macro-thinking and the overall concept, and consider the application of individualized strategy of orthopedic surgery based on the theory of "Natural Reconstruction". Some children have developed to ankle deformity again in short-term, which are typical examples of the recurrence cases in treated group. If comprehensive methods above are applied in time, the natural course of deformity development may be blocked. Moreover, if patients are treated combine with other neurorestoring methods, such as cell therapy [9], neuromodulation, suitable rehabilitation etc. [19, 20], they may have less recurrence and achieve better therapeutic effect. This could also be the direction of future research.

5 Conclusion

A combination of soft tissue and bony procedures along with external fixator helps to achieve complete correction of deformity and plantigrade foot, healing of ulcer, and restoration of functional activity. Through orthopaedic treatment, we can reconstruct the deformity, paralysis, sensory disorders and ulceration of foot and ankle suffered from spinal bifida sequela.

Regarding the recurrence cases of this deformity in developing age, orthopedic surgery should combine with the neurorestoring therapies, which may make suffering children to get more benefits.

References

[1] Chong C, Molet J, Oliver B, et al. The tethered cord syndrome: a review of causes. *Neurologia*. 1994, **9**(1): 12–18.

[2] Gunay H, Sozbilen MC, Gurbuz Y, et al. Incidence and type of foot deformities in patients with spina bifida according to level of lesion. *Childs Nerv Syst*. 2016, **32**(2): 315–319.

[3] Westcott MA, Dynes MC, Remer EM, et al. Congenital and acquired orthopedic abnormalities in patients with myelomeningocele. *Radiographics*. 1992, **12**(6): 1155–1173.

[4] Noonan KJ, Didelot WP, Lindseth RE. Care of the pediatric foot in myelodysplasia. *Foot Ankle Clin*. 2000, **5**(2): 281– 304, vi.

[5] Swaroop VT, Dias L. Orthopaedic management of spina bifida-part II: foot and ankle deformities. *J Child Orthop*. 2011, **5**(6): 403–414.

[6] Kameshita K. Therapy of foot deformities in spina bifida. *Nippon Seikeigeka Gakkai Zasshi*. 1987, **61**(10): 1159–1173.

[7] Tita AC, Frampton JR, Roehmer C, et al. Correlation between neurologic impairment grade and ambulation status in the adult spina bifida population. *Am J Phys Med Rehabil*. 2019, in press, DOI 10.1097/PHM.0000000000001188.

[8] Bradko V, Castillo H, Janardhan S, et al. Towards guideline-based management of tethered cord syndrome in spina bifida: A global health paradigm shift in the era of prenatal surgery. *Neurospine*. 2019, in press, DOI 10.14245/ns.1836342.171.

[9] Chen L, Nie H, Huang H, et al. Three cases of tethered release combined with olfactory ensheathing cell transplantation (in Chinese). *Chin J Clinicians (Electronic Edition)*. 2010, **4**(11):2311-2312.

[10] Wang ZH, Gao JQ, Zhan XH, et al. One-stage debridement and two-stage Ilizarov bone transport technology for post-traumatic lateral malleolus defect (in Chinese). *Chin J Reparative Reconstr Surg*. 2019, **33**(7): 865–870.

[11] Zang JC, Qin SH, Qin XL, et al. Treatment strategy for flail foot with sensory disorder of spina bifida sequela in adult (in Chinese). *Chin J Repar Reconstr Surg*. 2018, **32**(10): 1255–1260.

[12] Cotter AM, Daly SF. Neural tube defects: is a decreasing prevalence associated with a decrease in severity? *Eur J Obstet Gynecol Reprod Biol*. 2005, **119**(2): 161–163.

[13] Dicianno BE, Beierwaltes P, Dosa N, et al. Scientific methodology of the development of the guidelines for the care of people with spina bifida: an initiative of the spina bifida association. *Disabil Health J*. 2019: S1936–S6574(19)30106–2.

[14] Carroll NC. Assessment and management of the lower extremity in myelodysplasia. *Orthop Clin North Am*. 1987, **18**(4): 709–724.

[15] Shanahan MD, Douglas DL, Sharrard WJ, et al. The long-term results of the surgical management of paralytic pes cavus by soft tissue release and tendon transfer. *Surg Infancy Child*. 1985, **40**(Suppl 1): 37–41.

[16] Nicol RO, Menelaus MB. Correction of combined tibial torsion and *Valgus* deformity of the foot. *J Bone Joint Surg Br*. 1983, **65**(5): 641–645.

[17] Zhang Z, Zang JC, Qin S. Effectiveness of Ilizarov technique in treatment of clubfoot after burns (in Chinese). *Chin J Repar Reconstr Surg*. 2018, **32**(2): 178–181.

[18] Lee DY, Choi IH, Yoo WJ, et al. Application of the Ilizarov technique to the correction of neurologic equinocavovarus foot deformity. *Clin Orthop Relat Res*. 2011, **469**(3): 860–867.

[19] Panda PK, Mallik KC, Patel R, et al. Molecular basis of spina bifida: recent advances and future prospectives. *J Pediatr Neurosci*. 2019, **14**(1): 16–19.

[20] Huang HY, Mao GS, Feng SQ, et al. 2016 yearbook of Neurorestoratology. *J Neurorestoratology*. 2017, **5**: 111–115.

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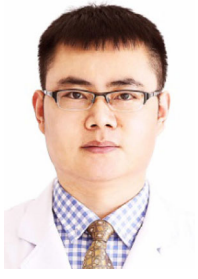

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