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Original Article

Hip Development After Selective Dorsal Rhizotomy in Patients with Cerebral Palsy

腦癱患者接受選擇性脊神經背根切斷術後對髖關節發育的影響

Chan Wai-Ming [a,*], Choi Kai-Yiu Alexander [a], Sun Kin-Wai [a], Fong Dawson [b], Yam Kwong-Yui [b]

[a] Department of Orthopaedics & Traumatology, Tuen Mun Hospital, Hong Kong Special Administrative Region, China
[b] Department of Neurosurgery, Tuen Mun Hospital, Hong Kong Special Administrative Region, China

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ABSTRACT

Objective: The effects of selective dorsal rhizotomy (SDR) on the hip development in children with spastic cerebral palsy (CP) are not well defined. The present study was performed to determine the effects of SDR and other associated clinical and radiological factors on the outcome of hip development after SDR.
Methods: The study included 53 patients who were skeletally immature at the time of SDR. Between 2003 and 2010, they underwent SDR at our institute. The age ranged from 4 to 15 years old. Their preoperative hip status was divided into two groups: normal and abnormal. The final outcome of the hip was considered good if the centre-edge angle of Wiberg (CEA) at last follow-up was more than 20 degrees without the need for orthopaedic intervention. Thirty-seven patients satisfied the inclusion criteria for statistical analysis.
Results: Seventeen patients were in the“Normal pre-op” group. In all patients (except for two patients), the hip status remained normal after the SDR. Twenty patients were in the “Abnormal pre-op” group. In this group, only two patients returned to normal hips, whereas 11 patients required orthopaedic hip surgery within 5 years after the SDR. The remaining seven patients had hip subluxation, but not to the extent of hip dislocation. The preoperative hip radiological measurements and functional status were positively correlated with the postoperative hip status. The preoperative radiological measurements showed superior predictive value when other covariance were considered. No difference of outcome existed in regard to the different surgical approaches of SDR.
Conclusion: Selective dorsal rhizotomy has a neutral effect on hip development. The preoperative hip radiological measurement is the most important predictive factor to determine hip status after SDR. Good collaboration between neurosurgeons and paediatric orthopaedists is essential for the best management of these patients.

中文摘要

目標: 選擇性脊神經背根切斷術（SDR）對痙攣性腦癱（CP）兒童的髖關節發育的影響並沒有明確的結論。這項研究是為了確定該手術，其他臨床和放射學上的相關因素，對髖關節發育的影響。
方法:研究對象包括53名骨骼未發育成熟的患者，他們在2003年至2010年間在本院接受SDR，年齡介乎4歲至15歲。他們在術前以髖關節的評估分為兩組：正常組和異常組。如他們在最後一次檢查時，髖關節X光的中心邊緣角(CEA)為20度以上並且沒有接受任何骨科手術，定義為有良好結果。有37名患者符合納入條件以進行統計分析。
結果:在正常組中有17名患者。除2名患者外，其餘在SDR手術後髖關節仍然發育正常。 20名患者屬於異常組，當中除2名患者手術後髖關節回復正常外，有11名患者需要在術後五年內進行骨科髖關節手術。術前髖關節X光測量數據和功能狀態與術後髖關節的狀態呈正相關。在排除其他協變量的影響下，術前髖關節X光測量數據更能預測SDR後髖關節的狀況。不同的SDR手術對結果沒有影響。
結論: SDR對髖關節發育的影響是中性的。術前髖關節X光測量數據是最重要的術後髖關節發育的預測因素。神經外科醫生及小兒骨科醫生之間的良好合作，是對這些患者的治療所不可或缺的。

* Corresponding author. *E-mail:* rockycwm@yahoo.com.hk.

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Introduction

Hip instability is common in children with spastic cerebral palsy (CP). It is the second commonest orthopaedic problem resulting in significant morbidity, especially among walkers.[1–5] It is believed to be caused by the combination of muscle spasticity and imbalance with secondary acetabular dysplasia, coxa valga and excessive femoral anteversion, and it is associated with scoliosis and pelvic obliquity. Soft tissue release of the hip adductors and iliopsoas effectively reverts early hip subluxation, whereas osteotomy may be required in late and severe cases of hip subluxation.[6–10] Selective dorsal rhizotomy (SDR) is evolving as an important treatment modality to reduce lower limb spasticity. Hip adductor and flexor tone is theoretically reduced, and thus SDR may act as a soft tissue release. However, early results of SDR show the contrary.[11] Subsequent studies of SDR on hip subluxation has had variable results (Figures 1–3) and inclusive results.[12–14] There is lack of reference in the Asian population. The object of the present study is to analyze the effects of SDR and other factors on hip development in patients with CP. Hence, the timing and role of neurosurgery and orthopaedic surgery with respect to hip development in CP patients may be delineated.

Methods

We retrospectively reviewed 53 consecutive children with cerebral palsy who had undergone bilateral selective dorsal rhizotomy (SDR) from June 2003 to August 2010 in our hospital. The selection criteria for SDR were spasticity of the lower limbs that interfered with normal function and gait; fair to good lower limb control and muscle strength; and fair to good trunk control with no fixed orthopaedic deformity.

The SDR was performed by the same group of neurosurgeons. The amount of bilateral afferent rootlets to be resected was determined by intraoperative trigger electromyography and by the on-table clinical response during dorsal rootlet stimulation. Two surgical approaches were employed. Selective dorsal rhizotomies performed from 2003 to 2006 were fashioned with L2-S1 laminoplasty. After 2006, the operative technique was modified to a single level laminectomy at the level of the clonus medullaris to reduce the surgical time and minimize the perioperative complications such as blood loss and spinal deformities.[15]

The children were followed up by their neurosurgeons and paediatric orthopaedic surgeons. Clinical data were extracted from the clinical management system (CMS) of the Hospital Authority (Hong Kong, China). The data included gender and the age at which the operations were performed, preoperative and postoperative ambulatory status. The Gross Motor Function Classification System (GMFCS) was used for staging the functional status. The centre-edge angle of Wiberg (CEA) and the Reimer's migration index (RMI) were used in the radiological assessment of hip coverage and subluxation (Figure 4).

Outcome Analysis

The patients had orthopaedic interventions before or within 1 year after SDR were excluded. Only those with a full set of radiographs with a minimum 1 year of follow-up were included in the data analysis. They were then categorized into two groups. Because radiological measurements vary with age, we defined the "Normal pre-op" group as patients having radiologically normal hips with a RMI less than 33% (for patients younger than 8 years old) or a CEA greater than 20 degrees (for patients 8 years old or older).[16–18] The patients were otherwise considered to have subluxed hips or dysplastic hips and were categorized in the "Abnormal pre-op" group.

For the outcome analysis of the hip, a "good" outcome was defined as patients who had a normal radiological hip measurement with a CEA of 20 degrees or greater without orthopaedic intervention at the last follow-up assessment of the study period. Patients who had a CEA less than 20 degrees or an osteotomy around the hip for residual dysplasia were considered a "poor" outcome.

Statistical analysis was performed by using SPSS 19.0 for Windows (IBM Statistical Package for the Social Sciences version 19.0, New York, USA). Analysis was performed on the following possible covariates: (1) preoperative hip status; (2) age at which the SDR was performed; (3) gender; (4) pre-op GMFCS; and (5) surgical exposure of SDR for the association with the outcome variable.

Results

Demographic Data

From 2003 to 2010, 53 skeletally immature patients underwent a SDR. There were 23 girls and 30 boys. Forty-five (85%) patients were classified as having spastic diplegia and three patients were diagnosed as having triplegia (diplegia + single upper limb involvement). At the time of the SDR, the mean age of the children was 7.9 ± 2.2 years. The average follow-up time was 5.3 years with minimal follow up of 12 months. Follow up continued for 46% of the children until closure of the pelvic tri-radiate cartilage. Before the operation, 15 patients were at GMFCS Level I; 9 patients, at GMFCS Level II; 33 patients, at GMFCS III; 5 patients, at GMFCS Level IV; and 2 patients, at GMFCS Level V (Figure 5). None of the patients had significant scoliosis since they had a Cobb's angle greater than 20 degrees preoperatively.

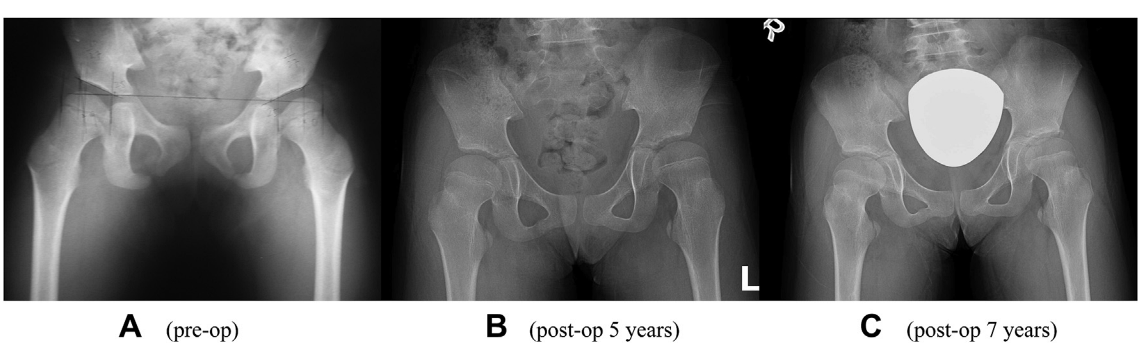

Figure 1. A spastic diplegic girl underwent selective dorsal rhizotomy (SDC) at 5 years. She had improved left hip subluxation after the SDR. (A) Pre-op image. (B) Post-op image at 5 years. (C) Post-op image at 7 years. Post-op = postoperative; Pre-op = preoperative.

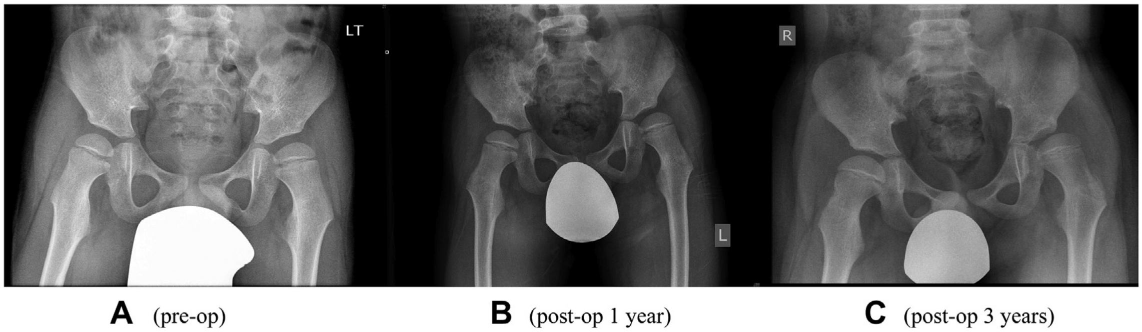

Figure 2. Progression in left hip subluxation in a spastic diplegic boy who underwent SDR at the age of 5 years. (A) Pre-op image. (B) Post-op image at 1 year. (C) Post-op image at 3 years. Post-op = postoperative; Pre-op = preoperative.

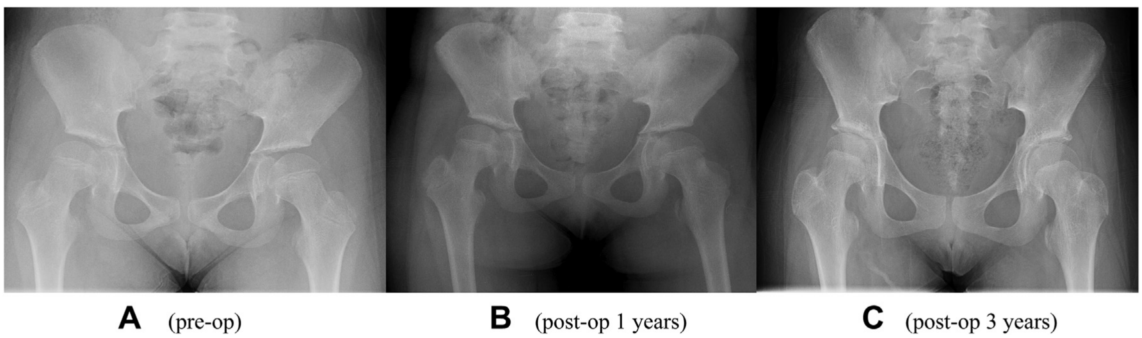

Figure 3. The hips remained normal during a 3-year follow-up period for a spastic diplegic boy who underwent SDR at the age of 7 years. (B) Post-op image at 1 year. (C) Post-op image at 3 years. Post-op = postoperative; Pre-op = preoperative.

Outcome

In regard to the SDR operation, there were no significant postoperative complications such as deep wound infection, cerebrospinal fluid leak, or pseudomeningocele. There was also no significant neurological deterioration. None of the patients had progressive spinal deformities on follow up. All patients had reduced hip adductor tone after the SDR.

Before the SDR, 10 patients had unilateral hip subluxation and 13 patients had subluxation in both hips. Twenty of these 23 patients satisfied the selection criteria for analysis and 18 (90%) of the patients continued to have poor hip development. Two patients with subluxed hips returned to normal after the SDR (Figure 1A–C). Eleven patients (representing 19 hips) subsequently required osteotomy for residual or progressive hip dysplasia (Table 1).

Seventeen patients had normal hips at the outset and satisfied the inclusion criteria. Fifteen (88%) patients remained normal at the final assessment. Two patients deteriorated. One patient had poor coverage in one hip, and both hips became subluxed in another patient .

Statistical Analysis

Statistical analysis was performed to determine whether there were any other associated factors on hip development (Table 2).

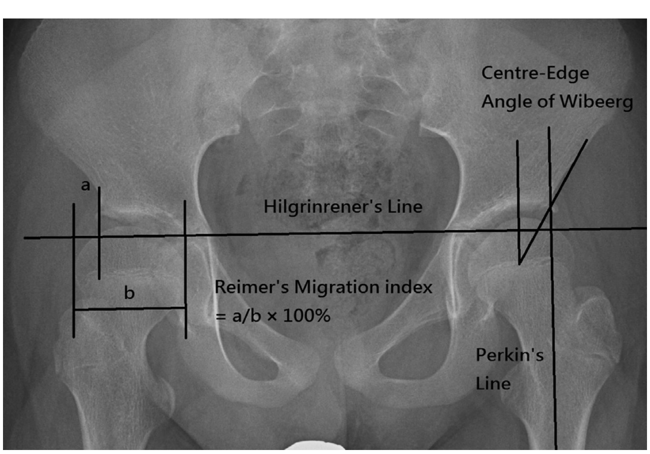

Figure 4. Centre-edge angle of Wiberg and Reimer's migration index (a/b) measurement on a pelvis radiograph.

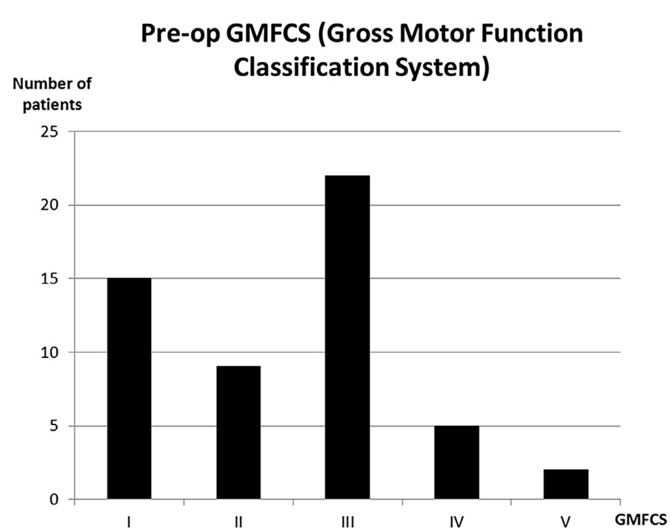

Figure 5. The preoperative GMFCS.

Table 1
Orthopaedic procedures performed after SDR

Orthopaedic procedures	Number
Hip osteotomy (pelvic/femoral)	19
Iliopsoas release	21
Adductor release	13
Others	46
Total	99

SDR = selective dorsal rhizotomy.

The factors that where assessed were (1) preoperative hip status; (2) age at which SDR was performed; (3) type of surgical exposure in SDR ; (4) gender; and (5) pre-op GMFCS.

Bivariate correlation analysis (Table 3) showed that the variables that were correlated with the postoperative hip status were (1) the preoperative hip status, based on radiological hip measurements ($p < 0.01$) and (2) the preoperative functional status in GMFCS ($p < 0.01$). Binary logistic regression analysis was then performed to create a model that would predict which patients may have normal hip status or abnormal hip status after SDR. The only variable showing a positive correlation with the postoperative hip status was the preopeative hip status ($p = 0.002$). The correlation was satistically significant, even in the presence of other covariates.

Discussion

The overall prevalence of cerebral palsy in Hong Kong is 1.3 per 1000 children.[19] The reported incidence of hip dislocation of children with CP ranges from 2.6% to 75%.[20–22] The incidence of hip subluxation or dislocation in our study was 45%, which was slightly higher than expected, given the relatively good functional status of this group of CP patients. Twenty-one percent of the patients subsequently required orthopaedic intervention for their hips. This percentage was similar to the 25% described in 1998 by Carroll and Moore.[3]

The natural history of hip subluxation and dislocation in patients with cerebral palsy is progressive,[5] especially in patients with hip flexor and adductor spasticity, and in nonambulatory patients.[5] For ambulatory patients, hip instability results in lever arm dysfunction and it predisposes patients to early labral tear and subsequent hip degeneration. Even in nonambulatory patients, hip instability may cause significant pain, deformity, and disability in up to 50% of these children, if left untreated.

The pathophysiology of hip subluxation in patients with cerebral palsy is multifactorial. The risk factors include ambulatory and functional status; severity of acetabular dysplasia, and the pattern of neurological involvement. As early as 18 months old, migration percentages were significantly greater in children with CP than in the normally developing population.[20] The progress is faster before 5 years old and becomes more static after 18 years old.

Numerous treatments are available for managing hip subluxation such as early soft tissue release or later femoral and/or acetabular osteotomy. Soft tissue release, particularly adductor release provides a favorable outcome in approximately two-thirds of patients in the younger age group.[7–10,23] Botulinum toxin combined with an abduction brace has a small benefit but does not prevent progressive hip subluxation.[24] Selective dorsal rhizotomy is a well demonstrated procedure that is effective in reducing spasticity and enhancing muscle strength.[25–27] The reduction of hip adductor and flexor tone with this procedure theoretically helps to prevent or revert hip subluxation and is similar the reduction provided by a tenotomy. Our study could only demonstrate that SDR had a neutral effect on hip development. Selective dorsal rhizotomy has a limited role in normalizing the subluxed or dislocated hip. No significant detrimental effect of SDR on hip development was otherwise shown. Selective dorsal rhizotomy may provide a role in maintaining the hips in normal status. Nearly 90% of normal hips continued to develop normally after the SDR. However, it may also be futile for the development of hips in CP.

This result provides us with a scientific basis for deciding the timing of surgery for CP patients with hip subluxation who are also candidates for SDR. Adductor or psoas release is still preferable for children who have early hip subluxation before SDR. For patients who are older or with a more severe degree of hip subluxation, the priority of SDR versus hip osteotomy should be individually considered. A close liaison between the paediatric orthopaedic surgeon and the neurosurgeon is invaluable for the best surgical planning for these patients who can anticipate at least two different major operations.

Two of the important predictive factors for hip development after SDR were the preoperative hip status and the functional status of CP, as indicated by the GMFCS.[12] The age at which the SDR was performed, the sex of the patients, and the surgical techniques had no significant correlation. The preoperative hip status was the superior predictive factor, if other covariant were also taken into consideration. Therefore, a good hip surveillance practice such as the Australian Consensus Statement is essential. Even for patients with “normal” hips, we recommend continual hip surveillance after the SDR.

There are no longterm follow-up studies in the medical literature concerning the stabilizing effect of SDR on hip development.[12,27,28] In previous study, the RMI and the CEA were both used for the radiological measurement of hip status. However, the effect of age on these measurements was not well addressed.

Some limitations of our study include the lack of a control group and a standard protocol for the follow up these children. Therefore, a significant portion (28%) of the patients was excluded from our study because of the lack of complete sets of pelvic X-rays or because early orthopaedic intervention may have masked the effect of the SDR.

Table 2
Summary of study group variables

Pre-op hip status	Age at SDR (Mean)	Type of exposure in SDR surgery	Gender (male:female)	Pre-op GMFCS (I to V)	Final outcomes of hip
“Normal” = 17	7.9	Laminoplasty (9 patients) Laminectomy (8 patients)	10:7	I = 9 II = 3 III = 4 IV = 1 V = 0	Good = 15 (88%) Poor = 2 (12%)
“Abnormal” = 20	8.0	Laminoplasty (6 patients) Laminectomy (14 patients)	12:8	I = 1 II = 3 III = 12 IV = 4 V = 0	Good = 2 (10%) Poor = 18 (90%)

Table 3
Bivariate correlation between post-op hip status and various covariants

		Pre-op hip status	Age at SDR	Type of exposure in SDR surgery	Gender	Pre-op GMFCS
Post-op hip status	Pearson correlation	0.782*	0.029	0.180	−0.122	0.521*
	Significance (2-tailed)	0.000	0.864	0.285	0.470	0.001

* The correlation is significant at $r \leq 0.01$ (2-tailed test).

Conclusion

In conclusion, SDR has a neutral effect on hip development. Preoperative hip status is the most important predictive factor to determine the hip status after SDR. It may also help to predict the need for subsequent orthopaedic hip surgeries. Constant hip surveillance is still indicated when SDR is contemplated. Good collaboration between neurosurgeons and paediatric orthopaedic surgeons is essential for the best management of these patients.

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