



ARAŞTIRMA / RESEARCH

Effects of open adductor tenotomy and adductor muscle botulinum toxin A injection on the hip instability in spastic type cerebral palsy

Spastik tip serebral palside açık addüktör tenotomi ve addüktör kas botulinum toksin-A enjeksiyonunun kalça eklemi instabilitesine etkileri

Zeynel Mert Asfuroğlu¹, Kemal Gökhan Günel², Hakan Ömeroğlu³

¹Tekirdağ Dr. İ. Fehmi Cumalıoğlu City Hospital, Orthopedics and Traumatology Clinic, Tekirdağ, Turkey

²Yenihayat Hospital, Orthopedics and Traumatology Clinic, Osmaniye, Turkey

³Ufuk University, Faculty of Medicine, Department of Orthopedics and Traumatology, Ankara, Turkey

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Abstract

Purpose: We aimed to evaluate the clinical and radiographic outcomes of hip instability in children with spastic-type Cerebral Palsy (CP) who had undergone open Adductor Tenotomy (AT) or received Botulinum Toxin-A (BTX-A) injections to the hip adductor muscles.

Materials and Methods: Maximum Hip Abduction Angles (MHAA) with the knees and hips in extension were measured preoperatively and the latest clinical findings of all patients in the AT and BTX-A groups were recorded. Reimers' Migration Percentage (RMP) was measured as the radiographic assessment in the AT group.

Results: There were 30 patients (mean age: 8.3 years) in the AT Group and 25 (mean age: 5.9 years) in the BTX-A Group whose clinical and radiographic data were available. The mean follow-up period in the AT Group was 29.6 months. In the AT Group, the mean MHAA before surgery and at the last assessment were 21.20 and 37.10 respectively. In the BTX-A Group, the mean MHAA before the injection and at the latest assessment were 25.30 and 34.20 respectively. The change in the mean MHAA in the AT Group was 16.20 and 8.80 in the BTX-A Group. In the AT Group, the preoperative RMP was 28.2% and the latest RMP was 22.2%.

Conclusion: AT and BTX-A injections significantly improved hip abduction angles. However, AT provided better clinical improvement and considerable radiographic correction in pediatric patients with spastic-type CP and hip instability.

Keywords: Cerebral palsy, botulinum toxin-A, adductor tenotomy

Öz

Amaç: Bu çalışmada Spastik tip serebral palsi (SP) tanılı hastalarda kalça instabilitesi nedeniyle açık addüktör tenotomi (AT) yapılanlar ile addüktör kaslara Botulinum toksin-A (BTX-A) enjeksiyonu yapılanların klinik ve radyolojik sonuçlarının değerlendirilmesi ve karşılaştırılması amaçlanmıştır.

Gereç ve Yöntem: Hem AT grubunda hem de BTX-A grubunda kalça ve diz eklemleri tam ekstansiyonda iken maksimum kalça abduksiyon açısı (MKAA) ölçüldü. MKAA'nın işlem öncesi değerleri ile son klinik muayenede elde edilen değerleri karşılaştırıldı. Bu ölçüme ek olarak AT grubunda kalça radyografileri üzerinde Reimers migrasyon yüzdesi (RMY) ölçüldü.

Bulgular: AT grubunda 30 (ortalama yaş: 8.3), BTX grubunda ise 25 (ortalama yaş: 5.9) hasta mevcuttu. Ortalama takip süresi AT grubunda 29.6 ay idi. AT grubunda ameliyat öncesi muayenede ve son muayenede ölçülen MKAA değerleri sırasıyla 21.20 ve 37.10 idi. BTX-A grubunda enjeksiyon öncesi muayenede ve son muayenede ölçülen MKAA değerleri sırasıyla 25.30 ve 34.20 idi. MKAA değerinin değişim miktarı ortalaması AT grubunda 16.20, BTX-A grubunda ise 8.80 idi. AT grubunda ameliyat öncesi RMY ortalaması %28.2, son muayene ortalaması ise %22.2 idi.

Sonuç: Hem AT grubunda hem de BTX-A grubunda kalça abduksiyon açıları anlamlı düzeyde artmasına rağmen AT grubunda bu artış BTX-A grubuna kıyasla daha fazla olmaktadır. Ek olarak AT grubunda radyolojik olarak belirgin düzelme gözlenmektedir.

Anahtar kelimeler: Serebral palsi, botulinum toksin-A, addüktör tenotomi

Yazışma Adresi/Address for Correspondence: Dr. Zeynel Mert Asfuroğlu, Tekirdağ Dr. İ. Fehmi Cumalıoğlu City Hospital, Orthopedics and Traumatology Clinic, Tekirdağ, Turkey E- mail: z.mert.asfuroglu@gmail.com

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INTRODUCTION

Cerebral Palsy (CP) is a group of permanent disorders of movement and posture that can limit the performance of daily activities; it is often accompanied by disturbances in sensation, perception, cognition, communication, and behavior. In developed countries, CP is the most common physical disability of childhood and has high morbidity and mortality rates¹.

Children with CP typically have normal hips at birth, but gradual deterioration occurs in single or both hips due to growth tethering by contractures. Primary causes of hip instability are lack of weight-bearing and asymmetry. Normally, children are born with a considerable amount of femoral anteversion, which normally decreases with age. However, if typical loading conditions are absent, femoral anteversion increases, and subsequent changes occur in the acetabulum and femur. The imbalance between strong hip flexors and adductors and weaker hip extensors and abductors can lead to adductor contractures, affect growth and potentially exacerbate changes in the acetabulum and femur^{2,3}.

Hip problems occur in more than 30% of children with CP and are the second most common musculoskeletal deformity after ankle equinus⁴. Hip instability in CP is classified as “hip at risk”, “hip subluxation” and “hip dislocation”. If there is a limitation of abduction less than 45°, it is named as “hip at risk”. If the femoral head is partially migrated out of the acetabulum, the hip is called to be “subluxated”. If the all contact is lost between the acetabulum and the femoral head, then the hip is called “dislocated”⁵. Long-term effects of hip dislocation include pain and loss of ability to sit comfortably in up to 50% of the patients. Other problems include difficulty with perineal care and personal hygiene, pelvic obliquity and scoliosis, poor sitting balance, and loss of ability to stand or walk^{6,7}.

Surveillance is the first step in managing hip problems in patients with CP. Early recognition of unstable hips is a treatment hallmark and prevents late complications and necessity for salvage procedures⁶.

In patients with CP, surgical treatment of hip problems include soft tissue release, hip reconstruction surgery (including femoral and pelvic osteotomies), and salvage surgery for long-standing painful hip dislocations⁸. Adductor Tenotomy (AT)

is a commonly used procedure for treating problems interfering with patients’ daily activities and personal hygiene. The procedure reduces the tendency of lateral displacement of the femoral head and prevents subluxation and painful dislocations^{9,10}.

Nonoperative treatment options include physical therapy, abduction orthosis, casting, gait training with neurodevelopmental therapy, rhythmic auditory stimulation or neurodevelopmental therapy, whole body vibration, and Botulinum Toxin-A (BTX-A) and intrathecal baclofen pump injections². BTX-A injections, either alone or in combination with other treatments, have extensively been studied in the literature¹¹. In a recent randomized controlled trial, it was reported that BTX-A injections improved the passive range of motion, pain, and spasticity, but were ineffective for preventing hip displacement¹².

The main hypothesis of the present study is that open adductor tenotomy is protective against hip instability in spastic type CP. Our primary aim was to evaluate the clinical and radiological outcomes of isolated open AT which had been performed to prevent and treat hip instability (hip at risk or subluxation) in children who had a diagnosis of spastic type CP and were able to walk with or without support. We also aimed to compare the clinical outcomes of patients with isolated open AT to those with BTX-A injection to the hip adductor muscles only, as well as to evaluate the effect of functional capacity of the patients on these procedures. The comparison of these two minor procedures to prevent hip instability would contribute information to help method preference to the literature.

MATERIALS AND METHODS

The local institutional ethics committee (Eskişehir Osmangazi University Clinical Research Ethics Committee) approved the content and methodology of this study (04-21-2014/12). The records of patients who were diagnosed spastic type CP and were treated by the same surgeon between 2002 and 2012 were retrospectively reviewed. 126 patients were found. The patients who had undergone open hip Adductor Tenotomy (AT group) or hip adductor tendon BTX-A injection (BTX-A Group) due to hip instability (hip at risk or subluxation) were included in the study. Informed consent was obtained from the parents of all patients. Patients who had undergone additional iliopsoas tenotomies or iliopsoas BTX-A applications and those who had undergone femoral or pelvic

osteotomies were not included in the study. Adductor Tenotomy patients with a clinical and radiological follow-up period less than 12 months and BTX-A patients with a clinical follow-up of less than 3 months were excluded from the study. Most of the BTX-A patients lacked pelvic radiographs prior to the BTX-A applications and/or during the follow-up, thus the radiological outcome of adductor tendon BTX-A applications could not be assessed. All the patients included in the study were able to walk with or without crutches, tripods or walkers or to stand upright with the aid of support. A total of 55 patients

who met the inclusion and exclusion criteria were included in the study.

Clinical and radiological assessments

Functional levels of the patients were determined using the "Gross Motor Function Classification System (GMFCS)"¹³ (Table 1). The number of GMFCS II patients was limited. To obtain a stronger statistical result in evaluating the clinical and radiological outcomes of the hips GMFCS II and III groups were combined.

Table 1. Description of the levels of GMFCS

The Levels of Gross Motor Function Classification System (GMFCS)	
Level I	Walks without limitations; more advanced gross motor skills such as running and jumping are limited
Level II	Walks with limitations but without assistive devices; limitations in walking outdoors and in the community
Level III	Walks with handheld assistive mobility devices; limitations in walking outdoors and in the community
Level IV	Self-mobility with limitations; children are transported or use power mobility outdoors and in the community

Radiological CP studies have indicated that Reimers' Migration Percentage (RMP) and the acetabular index are useful measures for hip monitoring^{14,15}. The RMP quantifies the lateral displacement of the proximal femoral epiphysis with respect to the acetabulum, and is the most commonly used radiographic parameter for determining the proximal femoral displacement risk and intervention effectiveness^{16,17}. In the present study, RMP was used to quantify hip instability on standardized Anteroposterior (AP) pelvic radiographs.

Maximum Hip Abduction Angles (MHAA) of all patients were recorded in both groups. The angle was defined as the maximum hip abduction that was obtained while both knees were in maximum extension with the patient lying in supine. None of patients had any other joint contractures that would affect this measurement. For distinction between stiffness and contracture, measurements were performed quite slowly. The preoperative and latest follow-up data of MHAA of AT Group were assessed. The MHAA before BTX-A application and the largest angles obtained within the first 3 months following BTX-A injection were also assessed.

Details of the treatment procedures

All interventions were performed by or under the strict supervision of the senior author. The treatment algorithm for the management of hip instability in cerebral palsy was determined according to RMP, acetabular index, hip contracture and age (Table 2).

BOTOX® (Allergan Inc.) preparation of BTX-A was used in this study. Every 100 U was diluted with 2-4 cc of 0.9% sodium chloride. It was injected into the adductor longus muscle by the palpation technique. At least two injections, 4-5 cm apart from each other were made under inhalation anesthesia. The BTX-A dose ranged from 1 to 2.5 U/kg. In the surgical treatment, the adductor longus tendon was sectioned as proximally as possible (Figure 1). The adductor brevis tendon was partially sectioned in cases with severe hip adduction contracture. After both AT and BTX-A procedure a bilateral long-leg circular cast was applied while the hips were kept in 45° of abduction with the help of an abduction bar and the knees in full extension. The cast was kept for 3 weeks for AT and 10-14 days for BTX-A injection. The patients were then referred to physiotherapy clinics on outpatient basis.

Table 2. Our treatment algorithm of hip instability in cerebral palsy.

Reimer's Migration Percentage	Acetabular Index	Surgical Procedure
<25%	≤25%	Follow-up
<25%*	≤25%	Botulinum toxin-A injection
<25% with hip adduction contracture*	≤25%	Adductor tenotomy
25%-50%*	≤25%	Adductor tenotomy
50%-75% (≤4 years)*	≤25%	Adductor tenotomy
50%-75% (>4 years)	≤25%	Proximal femoral osteotomy
>75% (any age)	≤25%	Proximal femoral osteotomy
	>25%	Acetabuloplasty

*if hip flexion contracture is present add iliopsoas tenotomy

The patients in both AT and BTX-A groups used nighttime orthosis with an abduction bar for an additional 3 months following the cast removal. The position of the orthosis was the same as the postoperative cast.

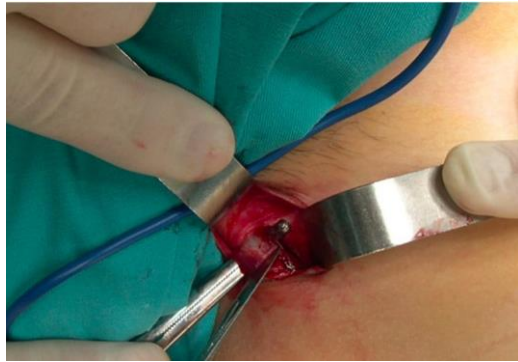


Figure 1. Open Adductor Tenotomy with adductor longus tendon exposure.

Statistical analysis

SPSS 22.0 (IBM Corporation, Armonk, New York, United States) was used for data analysis. The independent-samples bootstrapped T-test and Mann-Whitney U-test with Monte Carlo simulation were used to compare two independent groups. The paired-samples bootstrapped T-test and Wilcoxon signed rank test with Monte Carlo simulation were used to compare the repeated measures of the dependent variables. Categorical variables were compared using Pearson's chi-square and Fisher's exact test with Monte Carlo simulation. Quantitative data in the tables were expressed as mean \pm standard deviation and range (maximum–minimum). Categorical data were expressed as number (n) and percentage (%). Data were examined within 95% confidence interval, and a p-value of less than 0.05

was considered statistically significant. The power analysis was performed with reference to the MHAA variable. For 55 patients included in the study, the power value was found to be 100% and the number of the samples was sufficient (effect size=1.07, $\alpha=0.05$).

RESULTS

AT Group included 30 patients (12 females and 18 males) with a mean intervention age of 8.3 (3-18) years. BTX-A Group included 25 patients (14 females and 11 males) with a mean intervention age of 5.9 (3-10) years. Adductor muscle BTX-A application was performed twice in 4 patients. The mean follow-up period in the AT Group was 29.6 months (12-95).

The number of GMFCS Level II, III and IV patients was 3 (5 hips), 8 (15 hips) and 19 (38 hips), respectively in the AT Group. The number of GMFCS Level II, III and IV patients was 4 (7 hips), 13 (29 hips), and 8 (17 hips), respectively in the BTX-A Group.

In the AT Group, the mean MHAA significantly increased at the last assessment. In the AT Group, the mean MHAA significantly increased at the latest assessment in both the GMFCS Level II-III and GMFCS level IV hips. In the BTX-A Group, the mean MHAA significantly increased at the latest assessment. In the BTX-A Group, the mean MHAA significantly increased at the latest assessment in both GMFCS Level II-III and GMFCS Level IV hips. The change in the mean MHAA in the AT Group was significantly higher than the BTX-A group. The mean RMP in the AT Group significantly decreased at the last assessment ($P<0.001$) (Figure 2). The mean RMP in the AT Group significantly decreased in both the GMFCS Level II-III and GMFCS Level IV hips. There was no significant difference between GMFCS

Level II-III and GMFCS Level IV hips concerning the mean MHAA in the BTX-A group. Similarly, there was no significant difference concerning the RMP in the AT group. There was no significant difference between GMFCS Level II-III and GMFCS Level IV hips concerning the change in the mean MHAA in both AT and BTX-A groups.

Similarly, there was no significant difference concerning the RMP in the AT group (Table 3). In one hip with a preoperative RMP of 75%, an isolated open adductor tenotomy did not prevent the further hip dislocation. The GMFCS level was IV in this patient. None of the previously defined serious side effects of BTX-A were observed in any of the patients.

Table 3. Summary of the statistical data

	GMFCS		p value (between GMFCS groups)	All Hips Mean \pm SD (Min-Max)
	2-3 Mean \pm SD (Min.- Max.)	4 Mean \pm SD (Min.- Max.)		
BTX-A MHAA				
Preoperative	24.58 \pm 5.78 (10-30)	27.06 \pm 5.32 (20-35)	0.142	25.3 \pm 5.71 (10-53)
Postoperative	33.47 \pm 7.82 (20-50)	35.88 \pm 5.93 (30-45)	0.266	34.2 \pm 7.29 (20-50)
Changes	8.89 \pm 5.23 (0-20)	8.82 \pm 6.26 (0-20)	0.968	8.8 \pm 5.51 (0-20)
p value (Intra-groups)	<0.001	<0.001		<0.001
AT MHAA				
Preoperative	24.50 \pm 4.56 (20-30)	19.47 \pm 7.78 (5-30)	0.001	21.2 \pm 7.21 (5-30)
Postoperative	43.0 \pm 10.05 (25-60)	34.08 \pm 9.07 (15-50)	0.002	37.1 \pm 10.26(15-60)
Changes	18.50 \pm 10.65 (0-40)	15.13 \pm 11.48 (-5-45)	0.232	16.2 \pm 11.21 (-5-45)
p value (Intra-groups)	<0.001	<0.001		<0.001
AT RMP				
Preoperative	26.00 \pm 5.81 (20-38)	28.72 \pm 10.82 (10-75)	0.497	28.2 \pm 10.10 (10-75)
Postoperative	19.38 \pm 7.27 (10-30)	22.83 \pm 15.78 (0-100)	0.551	22.20 \pm 14.59 (0-100)
Changes	-6.63 \pm 9.05 (-26-5)	-5.89 \pm 9.71 (-25-25)	0.845	6.02 \pm 9.49 (-25-26)
p value (Intra-groups)	0.077	0.001		<0.001

General Linear Model Repeated Anova (Wilks' Lambda) / Paired T Test (Bootstrap), SD: Standart Deviation, Min: Minimum, Max: Maximum, GMFCS: Gross Motor Function Classification System, BTX-A: Botulinum Toxin-A, AT: Adductor Tenotomy, MHAA: Maximum Hip Abduction Angles, RMP: Reimers' Migration Percentage



Figure 2. A 4-year-old girl with an RI of 25%, which decreased to 8% in 12 months after performing Adductor Tenotomy.

DISCUSSION

The aim of this case-control study was to assess the effectiveness of isolated Adductor Tenotomy or

BTX-A injection to adductor muscles on preventing and treating hip instability (hip at risk or subluxation not dislocation) in 55 spastic-type CP patients with GMFCS Levels between II and IV.

Surgical options for hip problems include preventive surgery, which involves lengthening the soft tissue structures around the hip, starting with the tendon of the adductor longus and including the gracilis tendon, iliopsoas tendon, and adductor brevis muscle as necessary. Reconstructive and salvage surgeries can also be performed¹². Accepted indications for preventive surgery include an RI of >33% or an increase in the RI by >7% over the last year and an acetabular index of >30°¹⁴.

Reimers found that adductor muscle contracture is the primary cause of spastic hip subluxation and demonstrated that after subluxation, there is a risk of a 10% increase in the migration percentage every year. Reimers also stated that bilateral adductor

myotomy should be performed as soon as hip subluxation was noted¹⁶. Miller et al. stated that spastic hip subluxation did not spontaneously improve in patients with CP and reported that the progression risk was 23% after skeletal maturity if the migration percentage was $\geq 30\%$ ¹⁸. Bishay et al. treated 50 patients with spastic-type CP (100 hips) and a mean age of 3.6 years with open adductor longus, proximal gracilis, and proximal rectus femoris myotomy and iliopsoas lengthening with immediate postoperative immobilization in an abduction bar for three weeks followed by physiotherapy. Their findings indicated that the final outcome depended on age at the time of surgery, limited hip abduction, pre-surgical percentage migration, surgical technique, and postoperative physiotherapy¹⁹. Shore et al. stated that the need for subsequent surgeries following hip adductor surgery was related to walking ability of the patients²⁰. The results of the present study have revealed that a significant short-term improvement can be obtained in the hip abduction range of motion as well as in RMP by performing Adductor Tenotomy alone followed by 3 weeks of abduction casting and then by 3 months of night-time abduction orthosis if the patient has a GMFCS Level between II and IV, the RMP is less than 50% and no other hip muscle contractures or no coexisting acetabular dysplasia. Adductor Tenotomy provides better hip abduction range of motion than adductor muscle BTX-A injection in the overall patient population in the short-term follow-up (Table 3). Although we do not have a radiological data comparing AT and BTX-A outcomes, since the hip adduction contracture is one of the main causes of hip instability in CP patients, we certainly believe that a better range of motion in hip abduction may also have positive effects on the radiological outcomes.

The surgical success rate is closely related to the follow-up duration. High surgical success rates of approximately 70% have been found in studies with less than five years of follow-up. Turker and Lee reported eight-year results of 45 children with quadriplegic CP treated with soft tissue surgery for hip subluxation. They observed a 58% failure rate defined as progressive subluxation or dislocation eventually resulting in the need for a revision surgery²¹. Presedo et al reported 11 year results of 65 spastic children (mostly quadriplegic) treated with soft tissue surgery including adductor and iliopsoas releases for hip subluxation and found that the long-term hip dislocation could be prevented in 67% and a favorable outcome was more common in patients

who could walk²². So, based on the results of these two long-term studies, soft tissue release can be considered as a useful surgical alternative for preventing hip dislocation in diplegic patients but not in quadriplegic patients. The mean follow-up duration of patients who have undergone AT is about 2.5 years in the present study. Although current findings suggest good outcomes, long-term outcomes may not be as good as the short-term. Therefore, we plan to re-assess the patient data in the future to report the long-term follow-up outcomes.

Indications for BTX-A injections include deformities interfering with function, pain, conditions contributing to progressive deformities, painful spasticity with or without fixed muscle contracture, postoperative or post-treatment pain control, and symptomatic focal dystonia. In case of fixed contracture, toxin injections are ineffective. BTX-A has an excellent safety profile with a low incidence of side effects. Adverse events appear related to the total dose administered rather than the dose calculated based on the body weight of patients¹¹. We did not observe any side effects in patients with BTX-A application in the present study. The total BTX-A dose has never exceeded 22U/kg in the patients treated at our department, and the BTX-A dose for adductor muscles ranged between 1 and 2.5 U/kg.

The results of a randomized controlled trial revealed that BTX-A significantly reduced spasticity in the adductor muscles²³. Combined BTX-A injections and abduction bracing were investigated in a randomized controlled trial, which included 91 children with CP. The results of the group including patients with six monthly BTX-A injections over three years combined with the use of the Sitting, Walking and Standing Hip Orthosis was compared with the one including patients receiving standard care and surveillance but no bracing or BTX-A injections. The results indicated that fewer children in the treatment group required surgery and that the rate of hip displacement was lower in the treatment group. The clinical significance of these findings was found to be unclear because of the short follow-up²⁴. The findings of the present study have revealed that adductor muscle BTX-A injection followed by abduction casting for 10-14 days and then by nighttime abduction orthosis for 3 months improves the range of hip abduction. We think that BTX-A injection may be an alternative for preventing and treating hip instability in which the RMP has not exceeded 25%, a coexisting acetabular dysplasia is not

present and only adductor muscle contracture is present in spastic type CP. However, the clinical improvement obtained following BTX-A injection is not as much as the one obtained following the open AT (Table 3).

In the present study, both AT and BTX-A group were evaluated according to GMFCS levels as well. In the study by Shore et al., GMFCS levels were suggested to be the most important predictive factor in the success of adductor surgery²⁰. In the present study, the improvement in hip abduction range of motion as well as in the RMP does not seem to be dependent on the GMFCS level. Choi et al. stated that the patients with non-ambulatory GMFCS (IV-V) levels obtained lower functional scores after lower limb BTX-A injection²⁵. In the present study, GMFCS level of the patient does not seem to be correlated with the obtained range of hip abduction following BTX-A injection.

The shortcomings of the present study include a limited number of patients treated in a single institution, a short follow-up duration especially in the BTX-A Group, the absence of comparative radiological data in the BTX-A Group, and the retrospective study design. Besides, we have not assessed the rate of subsequent surgery, which is another significant determinant of the index intervention. However, the effects of two different hip adductor muscle interventions on the hip instability were assessed individually by excluding any kind of interventions on other muscles or bones around the hip joint. This was the main powerful side of the study.

Finally, we can conclude that although significant improvements have been observed in both AT and BTX-A groups regarding hip abduction range, the improvement in the mentioned variable was significantly higher in the AT group than in the BTX-A group. Besides, significant improvement in the RMP can be obtained in the hips treated by AT. The beneficial effects of AT and BTX-A on hip instability does not seem to be correlated with the functional status of the patient. We recommend the use of isolated AT treatment if the RMP is less than 50% and only adductor muscle contracture around the hip joint is present without coexisting acetabular dysplasia to prevent and treat hip instability in spastic type CP patients with GMFCS Levels between II and IV.

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