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Case Report

Botulinum toxin-A in postoperative pediatric stiff hips

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الملخص

أهداف البحث: تهدف هذه الدراسة إلى إيضاح مدى تأثير المساهمة الوظيفية لذيفان الوشيقية (البوتولينوم) – أ في علاج تيبس الورك عند الأطفال بعد التدخل الجراحي.

طرق البحث: أُدرج أربعة أطفال في هذه الدراسة ثلاثة منهم يعانون من خلل التنسج النمائي للورك بعد إجراء جراحات عظمية متكررة لهم. والحالة الرابعة لطفلة تعاني من الشلل الدماغي وتيبس بعد خضوعها لعملية جراحية عظمية بالورك. تم حقن ذيفان الوشيقية في عضلات مختارة كعلاج إضافي للعلاج العظمي والتأهيلي.

النتائج: جميع الأطفال لاحظوا انخفاض كبير في الألم مع تحسن كبير في التوضع وفي مدى حركة المفاصل ومقدار التنقل.

الاستنتاجات: النتائج الأولية لحقن ذيفان الوشيقية في عضلات مختارة تبدو واعدة في تخفيف الآلام وتحسين نطاق الحركة لدى الأطفال المصابين بتيبس الورك بعد التدخل الجراحي، مما يجعلها خيارا لعلاج الحالات الصعبة وغير المستجيبة للعلاج الطبيعي.

الكلمات المفتاحية: ذيفان الوشيقية; الشلل الدماغي; خلل التنسج النمائي; الورك; بعد التدخل الجراحي; تيبس الورك

Abstract

Objective: The aim of the study is to show the functional contribution of botulinum toxin-A in the treatment of postoperative pediatric stiff hips.

Methods: Four pediatric patients with postoperative stiff hips were included. Three patients with complicated developmental dysplasia of the hip after multiple revision surgeries, and a case of cerebral palsy after hip surgery were given botulinum toxin injections in selected muscles as an adjunctive therapy to the standard orthopedic and rehabilitation management.

Results: All patients experienced a significant reduction in pain with a significant improvement in posture, range of motion and mobility.

Conclusion: The preliminary results of botulinum toxin-A injection when given to selected muscles seem to be promising in relieving pain and improving range of motion in pediatric patients with postoperative stiff hips. It may be considered as an option in the treatment of difficult cases of postoperative stiff hips refractory to physiotherapy.

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Keywords: Botulinum toxin; Cerebral palsy; Developmental dysplasia; Hip; Postoperative; Stiff hip

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Introduction

Acquired hip stiffness in pediatric age group can be quite a challenging task for rehabilitation. Its causes include trauma, surgery, immobilization, infections, Perthes' disease, cerebral palsy (CP), slipped capital femoral epiphysis, idiopathic chondrolysis and others.

Postoperative stiffness is a disabling and frustrating surgical complication for both patient and surgeon.[1] Persistent postoperative stiffness remains difficult to treat in some patients and require different interventions.

Temporary reduction in hip range of motion is frequently seen after surgical treatment of Developmental Dysplasia of the Hip (DDH), which usually responds to stretching exercises. Nevertheless, reduction in range of motion and stiffness is more severe after redo or revision surgery especially if surgery was repeated several times, which means prolonged casting as well.[2–4] Revision cases are difficult to treat and need an expert team for management.[3,4]

The reported hip problems in CP are relatively frequent (25–75%).[5] They include progressive subluxation, dislocation, stiffness and pain.[6,7] These occurred because of spastic muscle forces acting on the femoral head in the acetabular cavity.[5]

Botulinum toxin-A (BoNT-A) as a neuromuscular blocker is used widely in different medical specialties including orthopedics. Jóźwiak et al.[8] reported the usefulness of targeted intramuscular injection of BoNT-A to prevent neurogenic hip dislocation in children with spastic CP. In fact the use of BoNT-A was reported much earlier in 1988, but the first published report for orthopedic use was in 1993. However, it was licensed only in Switzerland until around 2007.[8–10] Now, BoNT-A injection has been established as an alternative therapy in cases of muscle hyperactivity to achieve early mobilization and to control pain.[5] It also helps in improving the personal hygiene and rehabilitation.[11]

Targeted BoNT-A is a well-tolerated anti-spasticity treatment. It produces short term chemical denervation, which reduces muscle contractility by inhibiting presynaptic release of acetylcholine in a dose dependent fashion.[12,13] Consequently, it relieves muscle spasm related to pain from musculoskeletal origin.[12,14,15] The effect of BoNT-A injection lasts for 3–6 months, which gives a long term improvement to allow other treatments to take place.[12]

The current study aims to present the results of the use of BoNT-A in the treatment of postoperative acquired hip stiffness in children that is refractory to standard physiotherapy.

Case 1

A 38-month-old girl noticed to have an abnormal gait and diagnosed as a case of bilateral DDH. One stage bilateral open reduction without acetapuloplasty was performed in another institute at the age of 26 months. Follow up X-ray after four weeks showed dislocated right hip in the spica cast (Figure 1). Revision surgery was done in our institute, twice for redislocation, which included acetabuloplasty and femoral osteotomy. She had a fracture distal femur in between. The total casting was around 28 weeks. A significant decrease in range of motion was noticed in both hips and the patient was walking with a limp and hyperlordosis. Hip flexion was only from 40 to 90 degrees and abduction, adduction, and rotation were not more than 20 degrees each. Extensive physiotherapy was started but did not show a significant improvement. Decision was taken to admit the patient for rehabilitation and BoNT-A injection (BOTOX^{R}, Allergan Inc., Irvine, CA 92623, USA) in the quadriceps, the hip adductors and the Sartorius muscle (100 units in each muscle). She showed a marked improvement with her range of movements, spasm, pain and limping that started at two weeks post injection and continued till in her last follow up (Table 1).

Figure 1: Case 1; Pelvic X-ray showing dislocation of the right hip in hip spica cast, four weeks postoperatively.

Case 2

A four-year-old female was diagnosed to have bilateral DDH at the age of 16 months. At the age of 2 years she had bilateral hip surgery in form of anterior open reduction and Pemberton's acetabuloplasty (Figure 2). She had two revision hip surgeries for redislocation of both hips at two settings with a total casting of 24 weeks. With the removal of the last cast, both hips were manipulated under anesthesia and the patient was referred for an extensive physical therapy program (Figure 3). The flexion in the right hip was from 30 to 90 and in the left 10–90 degrees with very limited rotation and abduction. At three months she was assessed again in the clinic for hip range of motion. She had only gained 110 degrees of flexion, and around 20 degrees of abduction and rotation on either side. BoNT-A injection therapy (100 units in the quadriceps, 100 units in the hip adductors and 100 units in the Sartorius muscle), which decreased stiffness in both hips; this facilitated physiotherapy and rehabilitation measures that improved her

Table 1: Pain and spasticity before and after injection.

Patient	Pain scale before injection	Pain scale after injection	MAS* before injection		MAS after injection	
			R	L	R	L
#1	9	1–2	4	4	1	1
#2	9	1–2	3	4	1	1
#3	8–9	1–2	N	4	N	1
#4	8	1–2	4	2	1	1

* MAS: Modified Ashworth Scale. 0 = No increase in muscle tone, 1 = Slight increase in the muscle tone, manifested by catch and release or minimal resistance at the end of the range of motion, 2 = more marked increase in the muscle tone through most of the range of motion, but affected parts are easily moved, 3 = considerable increase in the muscle tone, and passive movements are difficult, 4 = affected parts are rigid in flexion or extension.[16]

Figure 2: Case 2; Postoperative pelvic X-ray showing both hips after bilateral open reduction and Pemberton's acetabuloplasty.

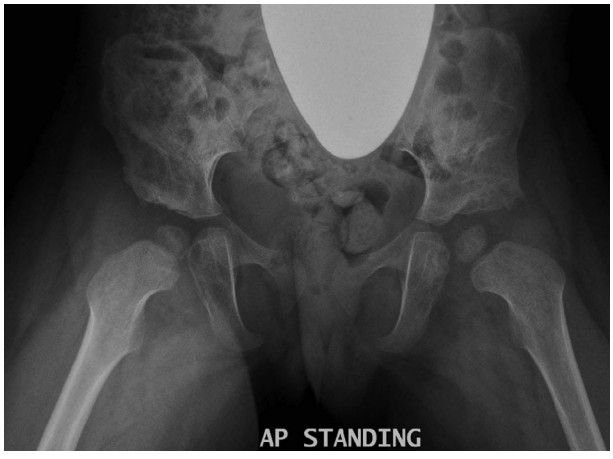

Figure 3: Case 2; Pelvic X-ray six weeks post cast removal showing bilateral concentric reduction of both hips but hips were stiff.

range of motion to near normal in the last follow up at the age of 4 years (Table 1).

Case 3

A seven-year-old female diagnosed with left DDH at an early age and was offered treatment but refused by parents. At 5 years of age parents agreed for surgical treatment, which was carried out in form of open reduction, acetabuloplasty and femoral shortening osteotomy. Three months post operation the hip spica cast was removed. Left hip was found in fixed flexion (80 degrees), abduction and external rotation deformity. She started to walk with a clear limp and a lordotic gait. She was subjected to extensive in-patient followed by outpatient physical therapy program. Due to failure in gaining significant range of motion, the left hip flexors and adductors were released surgically. However optimum results were not achieved. Subsequently BoNT-A injection was given (100 units in the quadriceps, 100 units in the hip adductors and 100 units in the Sartorius muscle). This was followed by extensive physical therapy that resulted in an improvement in range of motion. It was also proved to be beneficial in decreasing the limp, pelvic obliquity and spinal deviation (Table 1). Slight limitation of flexion and internal rotation (10–15 degrees) was observed at the left hip in the last follow up.

Case 4

A 17-year-old female; known to have diplegic CP presented with right hip pain and limping. She was seen at the movement disorders clinic for spasticity management, she had leg length discrepancy and pain in the right hip. She was diagnosed with right hip severe subluxation, for which she had right hip Chiari osteotomy. After surgery she had severe muscle spam, pain and spasticity in the right lower extremity much more than the left side and more than before surgery. BoNT-A injection was given twice, six months apart (100 units in the quadriceps, 100 units in the adductors, and 100 units in the hamstring muscles). She had an improvement in spasticity, pain and in her gait. In the last follow up visit she was using a cane to assist her in long distance walking but she was pain free with equal hip range of motion (Table 1).

Methods

Four patients with postoperative hip stiffness that was refractory to extensive physiotherapy program were included in the study after obtaining informed consent from the parents to go through the trial of BoNT-A treatment. The four patients included were two patients with post revision open hip reduction for DDH and prolonged casting, one older patient with DDH that had three simultaneous procedures and one older patient with CP post hip surgery for subluxation. They all had an extensive physiotherapy program that was for more than six weeks at

least. The decision of which muscles to be injected was reached between the pediatric neurologist and the physiotherapist. Modified Ashworth Scale (MAS) in children with spasticity was used to assess muscle tightness before and after injection (Table 1).[16]

All injections were given by an expert in the treatment with BoNT-A (the first author). Patients were examined after the injections for signs of adverse reaction to the toxin. Parents were asked to report back if any of the complications occurred later. Patients were referred back to physiotherapy department to continue their rehabilitation program. The pain severity was assessed on a standardized Color Analog Scale (in case of children below 6 years the mother was asked to answer), which is a validated scale (Table 1).[17,18]

Results

All four patients experienced a significant reduction in pain with a significant improvement in posture, range of motion and mobility. All patients showed an improvement at 2 weeks period (max effect of the Botox). No serious complications were reported that were directly related to the use of BoNT-A treatment. The three DDH cases needed single dose injection to get the desired outcome whereas the fourth CP patient needed repeated injections (two doses) to get the desired outcome, probably due to her original spasticity. She showed an improvement after 2 weeks, but she has to be injected after 6 months because the wearing off of the effect of the Botox. All muscles injected matched the clinical presentation (Table 1).

Discussion

There are multiple options for treatment of a postoperative stiffness: physiotherapy, manipulation under anesthesia and surgical debridement or releases,[19] the gain in the range of motion with physiotherapy is often modest. The surgeon can forcefully overcome adhesions with manipulation under anesthesia, while moving the joint through the desired range of motion paying attention to exert enough force to move the joint and not enough to fracture the already weakened bone by long immobilization and reduced mobility. In refractory cases surgical release of adhesions often is used.[19] In all our cases we started with an extensive physiotherapy program, case 2 had manipulation under anesthesia and case 3 had surgical releases. These measures did not help and they had refractory postoperative stiffness that led us to explore more options for treatment. As a multiple disciplinary team in our spasticity clinic, which includes pediatric neurologist, pediatric orthopedic surgeon, pediatric rehabilitation medicine, pediatric physical therapist and others, and as the experience with BoNT-A injection is well established in our center; the idea of using BoNT-A injection came out, knowing its safety and effectiveness even after repeated injections.[13,20,21]

BoNT-A has been used to decrease postoperative spasticity related pain[22] and in postoperative contractures in cases post total knee and total hip arthroplasty.[21,23] It has been shown that the chemical denervation effect of BoNT-A leads to temporary paralysis that relieves muscle over activity, which is a direct cause of muscle shortening and the decrease in range of motion.[24]

The presented cases showed that BoNT-A injection helped to reduce pain and increase range of motion in young patients with pain and stiffness after surgery for difficult and complex complicated DDH cases, and in CP patients post hip surgery. These findings appear to confirm previous reports of the role of BoNT-A as a potentially effective adjunct therapy, which could stabilize the condition and improve the quality of life in CP and sport injury cases.[12,14] The choice of muscles for BoNT-A administration should be determined by clinical examination looking for the painful muscles affected with spasm and decrease range of motion. To increase the effectiveness of BoNT-A it should be injected inside the fascial compartment in an appropriate dose and volume so the diffusion occurs with minimizing unwanted spread.[22]

Following BoNT-A injection, a measurable goal should be set to reach through multidisciplinary team; consisting of physiotherapy, neurology and orthopedic surgery. This may include tight soft tissue stretching, activation of weak muscles and re-educating the affected muscles reaching to functional exercises that leads to return to normal function.[12]

To our knowledge, no previous study has shown the results of BoNT-A in postoperative stiff hips in children. The limitation of this study is the small number of the patients and the lack of a control group.

Conclusion

The presented cases demonstrate that the preliminary results of using BoNT-A are promising and it is a potentially rewarding management option that could be considered in the management of postoperative stiff hips in the pediatric age group, especially in difficult cases that are refractory to standard treatment. It is recommended to recruit more patients to establish this line of treating postoperative hip stiffness in children. Muscle selection, injection techniques and participation of all concerned subspecialties utilizing appropriate modalities of treatment will result in well-defined treatment goals and outcome.

Disclosure

The authors have no conflicts of interest whatsoever.

Author contributions

All authors contributed equally to this manuscript. TR wrote the first draft.

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