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Research Article

Reducing Spasticity of Muscles that are Surgically Treated, with Simultaneous Botulinum Toxin Injections, Further Improves the Final Result, in Children Affected from Cerebral Palsy

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Abstract

Botulinum toxin injections are widely used in children affected from cerebral palsy (CP) to reduce spasticity. When short muscles develop, their lengthening with surgical procedures is required, in order to improve the motor function. We studied simultaneous use of BoNT-A on the surgically lengthened muscles, to reduce their spasticity.

We present a cohort of 52 patients, affected from spastic type of CP, ages 5 - 14 yrs old. Evaluation of function was performed with GMFCS, whereas for measurement of spasticity we used the modified Ashworth scale. Short muscles were evaluated clinically under anesthesia, in previous BoNT-A injections. The range of movement (ROM) affected joints was recorded. BoNT-A injections were performed at the end of the surgical intervention, in the proximal part of the lengthened muscle, using US guidance. Removable splints were used in the first 3 - 4 postoperative days.

Improvement of mobility in our children was achieved after surgery. Early mobilization of the treated limbs was allowed, as tolerated. Children returned early in their individual physiotherapy program. Reduced spasticity had a positive effect in the pain tolerance. Formation of adhesions was eliminated with the early mobilization and reduced spasticity.

We propose treatment of appropriately selected CP children, with simultaneous surgical lengthening and BoNT-A injections.

Keywords: *Cerebral Palsy; Children; Botulinum Toxin; Surgery; Muscle Lengthening*

Introduction

Multilevel botulinum toxin injections are an effective method to reduce abnormal increased muscle tone, for children affected from cerebral palsy. This will improve the range of movements of the affected joints, provide further action to weak antagonists muscles and improve the individual motor function. As children are growing, affected muscles become shortened, remaining always with spasticity. Lengthening of short muscles either as tendon lengthening, or musculotendinous release, is effectively improving the function of CP children. These procedures require immobilization with casts at the initial period, until healing of the surgically lengthened muscles is completed. At the initial postoperative period, pain increases the muscle tone acting as positive stimulation, further increasing spasticity and acting as a vicious circle, increasing pain and spasticity [1-6].

We are using simultaneous botulinum injections, in those muscles that are surgically treated with lengthening, in order to reduce their spasticity. Combining the lengthening of short muscles and reducing spasticity, we intend to maximize the functional improvement of children with CP.

Aim of the Study

The aim of our study is to evaluate the effectiveness of our simultaneous approach for management of children affected from CP that have developed short spastic muscles.

Patients and Methods

During the period 2005-2015 we have surgically treated 52 children with CP. All of them were affected from spastic type of cerebral palsy. There were 16 tetraplegic, 24 diplegic and 12 hemiplegic children. Their ages ranged from 5 to 14 yrs old, with mean age 11 yrs. Before surgery we evaluated the level of mobility using the GMFCS. For each patient a detailed examination for the range of movement (ROM) of the hip, knee and ankle joint was recorded, in the supine position. In the standing position, either active or assisted in a standing frame, we evaluated the presence of deformities and axis deviations for the lower limbs. We used visual assessment for gait and occasionally instrumented gait analysis.

Criteria for surgical intervention was the presence of short muscles that prevented the proper alignment of the lower limbs in standing position or in walking trials, either alone or in their physio program using appropriate aids. In the presence of severe spasticity, it is difficult to accurately recognize short muscles. We regularly perform BoNT-A injections under anesthesia, in order to evaluate the presence of short muscles. In this cohort, the vast majority had previously BoNT-A injections, simultaneous with physiotherapy.

Surgical treatment for the hip, with release of adductors, was performed in cases of reduced abduction of the hips with simultaneous flexion contracture, with severe scissoring during standing or laying. In 8 children with radiological signs of subluxation, or with severe increased anteversion, femoral osteotomies of varus and derotation were included. We have not performed any psoas release in our patients.

For the knee joint, surgical treatment was done in children with reduced popliteal angle, with fixed flexion of the knees in the standing position. We performed posterior thigh muscles lengthening (semitendinosus, semimembranosus, gracilis, biceps femoris) with minimal invasive surgery. We always tried to repair the tendon sheath. Lengthening of the semitendinosus tendon was done with Z plasty, never exceeding 4 cm lengthening, suturing end to end. We use aponeurosis release for semimembranosus and biceps femoris and tendon release at the musculotendinous junction for gracilis. Only two children had quadriceps lengthening, due to stiff knee gait, with absence of knee flexion during swing.

For the ankle joint, surgical intervention was done in fixed equinus or severe equinovarus deformity on the standing or the walking phase. In planovalgus deformities, ankle correction included subtalar fusion with achilles lengthening. When fixed equinus of the ankle joint is more than 20d, we prefer achilles lengthening with Z plasty, with 3 - 4 cm lengthening. We used gastrosoleus release in less equinus deformity. In severe varus we performed tibialis posterior lengthening. Meticulous repair of the sheath was always attempted.

Similar approach was followed for the upper limb, when hemiplegic and tetraplegic children were treated. Preoperative evaluation with BoNT-A injections performed under anesthesia, with accurate US guidance, revealed the presence of short muscles. This enabled us to select children that will benefit from surgical intervention in the upper limp. For the arm, lengthening procedures were performed for elongation of biceps brachi, FDP and FDS, FPL. In 7 children we performed FCU transfer to ECRB. BoNT-A were performed simultaneously for the elongated muscles only and not for the transferred one. All elongated tendons were sutured with end to end technique, with absence of any tension in the sutures.

Injections were performed at the end of the surgical elongation. Children were already anesthetized, so we used ultrasound guided injections in the proximal part of the elongated muscles. The absence of spasticity eliminates the feeling of injecting in a spastic muscle, so the use of US is very helpful for accurate injections. We used Abodotulinum toxin (Dysport) at a maximum dose of 1000 iu and with a dosage of 20 iu/kg body weight, with the same principles as those children regularly treated with BoNT-A alone.

After the procedure, none of our children was immobilized in a plaster cast. We have used for the first 3 - 4 days removable splints for the knee and the ankle joint as well as for the wrist. The casts were then removed, as we were expecting reduction of the spasticity. The usual analgesics (paracetamol) were prescribed in our patients.

Children remained for the night in the hospital when multiple procedures were performed, but in cases of few muscle releases, they were in the day clinic program.

Results

Children returned in their complete physiotherapy program after 3 - 4 weeks, waiting for the complete healing of elongated muscles. While staying home, they were able to be transferred around easily. In the absence of casts, while in bed, they could perform movements of the joints that were around the elongated muscles. Toilet actions were easy due to the absence of casts. Movements of the limbs without weight bearing were allowed as tolerated. Promoting early movements, we aim to eliminate the formation of adhesions at the level of the surgical procedure.

In children that had achilles tendon lengthening with a Z procedure, we started partial weight bearing at 3 weeks, using a removable cast below the knee. Children with tetraplegia were allocated in their standing frames since the first week postoperatively. Diplegic and hemiplegic children were in passive movements twice per day from parents or care givers, according to the physio program that was provided in the initial period. Each patient had an individual postoperative physio plan, in order to improve particular motor skills.

Early reduction of spasticity has a positive effect in the pain relief of our patients. Severely affected children are not able to express clearly the intensity and the site of pain, whether it is the surgical procedure or the uncomfortable brace that may create a pressure site in their limb. Relief of muscle spasm in any injured muscle reduces pain. In our children, that had multilevel procedures, none of them complained for pain that required further analgesics.

Improvement in the motor ability was achieved because of the surgical lengthening of short muscles. Restoring the anatomical axis of the body, when releasing joint contractures, contributed to the whole balance of the body. Occasionally diplegic children achieved their autonomous first steps, after correcting their standing balance. Hemiplegic children developed better gait, mainly by eliminating toe strike at the initial foot contact and by improving knee movements both in the contact and swing phase of the gait.

Tetraplegic children improved the hip, knee and ankle joint movements and alignment, increasing the time on the standing frame for GMFCM 4,5 or by advancing their motor level for those in the 3 and 2 grades. The ROM of joints of surgically elongated muscles became wider but simultaneously there was further reduction in spasticity. Measurement of spasticity using the Modified Ashworth scale showed better level of scaling. We feel that this is the most reliable muscle tone assessment method. It is difficult to measure the reduction in spasticity in elongated muscles. However children remained in proper alignment of their limbs, without the contractures that produced equinus, knee flexion, hip flexion and scissoring. This was also apparent in the position of the fingers, wrist and elbow in those treated for the upper limb.

Discussion

Use of BoNT-A has been established as a common method to reduce spasticity and improve the motor development of CP children. There is a great volume of references in the literature regarding the effectiveness of BoNT-A use, always as an adjuvant therapy combined

with intense physiotherapy and the use of casts. The key muscle concept and the selection of key muscle for injections is an appropriate treatment plan for each individual child [1,2,8]. In a systematic review of the effect of physiotherapeutic intervention after BoNT-A injections, in the lower limb, improvement in muscle tone is reported, with further improvement of motor and functional skills [9].

Adjuvant therapies consisted of casting, splints, resistance training electrical stimulation and rehabilitation programs. Casting associated with BoNT-A injection improved the range of passive and active motion and reduced spasticity better than did BoNT-A alone [10]. There is low to moderate improvement of function, from resistance training and rehabilitation program.

There are reported concerns for the value of BoNT-A injections for the final result, as compared to intensive physiotherapy alone [11]. They refer to a group of GMFCM 1-3, but they emphasize that there is a subgroup in the CP population that can benefit for injections. They comment also on the problems the children with casts have, during their intensive physiotherapy.

It has been reported that casting alone increases the muscle length unit, while BoNT-A affects only the muscle resting length [12]. When used in young children, usually in ages 4 - 8 years it is claimed that it reduces the development of contractures and the incidence of surgery for these children [13].

However, as children grow up, they develop short muscles, despite all our conservative methods with appropriate physiotherapy. It is difficult to recognize with accuracy short muscles when there is increased spasticity.

With repeated BoNT-A injections, muscle tone may reduce but the ROM is not affected, as development of contractures is not only the result of increased muscle tone [7]. They report a prospective clinical cohort, similar to our subtypes of CP, with mean age of 5 yrs. The children had a mean of 2 injections per muscle. There were up to 8 injections, with a range of age from 11 months to 17 years. Expectations to improve the GMFCS level are limited only to injections and physiotherapy program, requiring surgical intervention in carefully selected children, in their long term results. Development of contractures continues, despite the reduction in muscle tone, due to molecular changes and deposition of collagen [14].

We regularly perform BoNT-A injections under anesthesia, in the selected muscles, using US guidance. When anesthetized, there is accurate assessment for the presence of short muscle [1,15]. This is even more essential for the upper limb, when deciding whether surgical treatment will be of benefit [16,17]. Arm injections are less painful than in the lower limb, but the whole evaluation of the child's hand is more valid when anesthetized. Pain from BoNT-A injections is well tolerated. Using local anesthetic or even distraction child life specialists, local BoNT-A injections were reported as pain free in 84% of children [18].

Short muscle will not respond to BoNT-A injection, since it requires lengthening. Surgical management of CP children, addressing with a single event multilevel lengthening procedures, with realignment of the anatomical and mechanical axis of the limb, definitely improves the motor level of them. For tetraplegic children, in level GMFCM 3-5, we plan appropriate position in standing frame or supported walking using aids, considering the position of the hips, knees and ankle. When bony procedures as hip varus and derotation osteotomies are required, they are performed in the same surgical session. Same principles are applied for diplegic children that are usually in level GMFCM 1-3, where improvement of the gait is more obvious. In hemiplegic children, who are usually level 1 - 2, the gait improves both in metric and time parameters.

Surgically treated short muscles are always spastic. By lengthening them, we improve the ROM of the joints, the limb alignment, but not spasticity. Our physiotherapy team, postoperatively, is working for increasing the strength of antagonists muscles, improving the ROM and avoiding development of adhesions. Using simultaneously BoNT-A injections we reduce spasticity. Dosage depends on the body weight and the number of muscles that are planned to be injected. The use of US is facilitating the injection, since in the anesthetized children, with complete muscle relaxation, the feeling of entrance of the needle in a spastic muscle is reduced [1,19].

Appropriate immobilization with casts is used for tendon or muscle lengthening procedures. Approximately 3 - 4 weeks are required for healing and removal of casts. During this period, handling of the child, either at home or from caregivers, poses difficulties, with hygiene, sitting position or generally transfer. Often postoperative casts create pressure sores and pain in their edges.

The use of BoNT-A has been described preoperatively in order to reduce pain. It refers to two groups of 9 tetraplegic children that had orthopaedic surgery both with bony and muscular procedures. The second group had BoNT-A treatment before surgery, with multisite injections. Children did not take less analgesic drugs, but they had decrease of duration of postoperative pain. They report prevention of skin lesions from the casts [20]. We use removable splints for the first 3 - 4 postoperative days in order to maintain the corrected position of the limbs, after the surgical procedure. The immediate postoperative pain produces severe spasm that brings the limbs in the unpleasant position. When the muscle paralysis starts, we remove the splints as muscles are relaxing and pain reduces, with the limbs in the desired position.

Preoperative BoNT-A injections were first described from Rutz, Hofmann and Brunner [21] as a selection tool for those children that will benefit for surgery. The main concern for surgical tendon lengthening procedures is the weakening of the muscle that may lead to deterioration of the gait. Using instrumental gait analysis with kinematic, kinetic and EMG studies, they canceled scheduled procedures for those whose gait deteriorated after injections. We use a similar protocol, based on clinical observation, for these children that do not have a positive effect in their posture or gait after BoNT-A injections. Treatment of short muscles, appropriately selected, requires lengthening, since BoNT-A injections alone, will have no positive effect.

The analgesic effect of BoNT in children has been reported in detail [22]. We describe the analgesic effect on surgically treated spastic muscles. Immediate postoperative pain was easily managed with paracetamol, for the first 2-3 days. Application of splints for these first days was used to keep limbs in the new position, avoiding painful muscle spasms. The analgesic effect appeared with reduction of spasticity, allowing us to remove splints and to offer our patients mobilization of the limbs, as tolerated. There are studies that report that botulinum toxin may block neuropeptides that participate in the inflammation process. There may be a more central analgesic effect, by retrograde axonal transport to the dorsal root.

Improvement of function for CP children is a multidisciplinary work [23]. Physiotherapist has a detailed program for early intervention, according to each individual plan. Orthotics were used when starting the standing or walking program, after the procedures. In children older than 6 - 7 years, we promote gait without splints, by correcting their deformities that require orthotics.

Initial splints for the first 2 - 4 days were also used to protect the sutures of lengthened tendons. Excessive spasticity that is continuous in CP children may pose excessive tension for the tendons as well as the musculotendinous junction. In our children, all sutures were done without tension and with adequate but not excessive lengthening, thus eliminating the risk of breakage. With spasticity reduction, with the botulinum toxin effect, we permitted early movement, as tolerated, with parental and physiotherapy assistance.

Conclusion

This report describes the effective management of CP children, by combining both surgical treatment and simultaneous BoNT-A injections. Despite the lack of a control group of children, because of the reproducible results in our department, we propose the above approach.

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