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Case Report

INNOVATIVE METHOD TO TREAT PATELLA ALTA IN A PATIENT WITH SPASTIC CEREBRAL PALSY: A CASE REPORT WITH LITERATURE REVIEW

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SUMMARY – Patella alta is a common condition in ambulatory patients with cerebral palsy, especially those with crouch gait. The treatment of symptomatic patella alta in such patients is complex and conservative methods seldom provide satisfactory results. Thus, patellar tendon advancement surgery is often required and can be, if necessary, combined with other surgical procedures. Here, we report a case of combining both conservative method and surgical procedure in a 26-year-old female patient with cerebral palsy and bilateral patella alta. One month prior to surgical intervention we administered a botulinum toxin type A (BTX-A) in the ipsilateral quadriceps. Reduced muscle spasticity of the knee extensor mechanism facilitated the patellar tendon shortening procedure and yielded excellent postoperative results. After a six-month postoperative rehabilitation course, the patient had no pain and the knee range of motion improved from 5-50° preoperatively to 0-140° postoperatively in both knees. To our knowledge, we are the first to report a use of preoperative BTX-A in the treatment of patella alta in a patient with cerebral palsy. The efficacy and safety of this treatment modality may give encouragement to a wider use of preoperative BTX-A in order to facilitate surgical intervention and postoperative rehabilitation in cases of muscle spasticity.

Key words: *patella, patellar ligament, cerebral palsy, spasticity, botulinum toxins*

INTRODUCTION

Patella alta, also called the high-riding patella, is a condition in which the patella lies superior to the trochlear groove of the femur. It is often found as one of the four major factors in patellofemoral joint instability. Along with trochlear dysplasia, excessive TT-TG, and patellar tilt, it can contribute to subluxation and dislocation of the patella[1]. Patella alta is also frequently found in ambulatory patients with cerebral palsy, with a reported prevalence of 61-93% in these patients[2,3]. It is often associated with crouch gait, i.e. the pattern of walking in cerebral palsy patients where a knee flexion contracture is present during the stance phase of walking. It is considered that subsequent overloading of the knee extensors combined with spasticity of the rectus femoris muscle leads to overstretching of the patellar tendon fibres causing development of patella alta[4]. Due to patella alta, contact areas between the patella and the femoral trochlea are reduced, therefore patellofemoral joint pressure stress is increased. This leads to patellofemoral dysfunction, weakness of extensor mechanism of the knee and eventual development of articular cartilage degeneration and anterior knee pain[5].

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Patella alta in cerebral palsy can be treated conservatively and surgically. Some of the non-operative methods found in literature are taping of the patella, application of a patella stabilising knee brace, and injections of botulinum toxin type A (BTX-A) into the quadriceps muscle[6]. Surgery is advised in symptomatic patients and those refractory to conservative treatment. Patellar tendon advancement (PTA) surgery is used for the correction of patella alta by distalising the patella and restoring the patellofemoral joint congruency[7]. This can be achieved either by shortening or plication of the patellar tendon or by transposing and fixing the tendon insertion or tibial tubercle distally. For the treatment of crouch gait and knee flexion contractures in patients with cerebral palsy, the PTA is frequently combined with other surgical procedures, i.e. as a part of single-event multilevel surgery (SEMLS) or with distal femoral extension osteotomy (DFEO)[8,9].

Here, we present the first case to our knowledge, in which a patient with cerebral palsy and bilateral patella alta was treated by coupling both a conservative and surgical approach. One month prior to the PTA procedure we administered a BTX-A in ipsilateral quadriceps. As the extensor apparatus of the knee had relaxed, we performed a surgical plication of the patellar tendon securing it with a trans quadriceps tendon encircle wiring. Due to the reduced tension of the quadriceps muscle achieved by a local injection of BTX-A, the surgical intervention of distalizing the patella was easier to perform and excellent short-term postoperative clinical results were noted.

CASE REPORT

Our patient was a 26-year-old woman with cerebral palsy in the form of spastic quadriplegia with bilateral anterior knee pain of 3 years duration. Prior to the development of knee pain, she underwent multiple surgeries on her lower extremities. Regarding the knee, at the age of 12, a bilateral elongation of knee flexors was performed. Her last surgery was at the age of 23 when she underwent a total left hip arthroplasty (THA). During the postoperative rehabilitation following the THA, she started to feel pain predominantly in her right knee. She also complained of intermittent right knee swelling after walking and a limited range of motion. Despite physical therapy, the symptoms aggravated.

At the time of the presentation, she walked with the aid of a walking frame. Clinically, both quadriceps muscles were atrophic and spastic. There was no knee effusion present, the patellae were situated proximal to the normal position and the right patella was extremely painful on palpation and compression. The right knee range of motion (ROM) was 5-50° of flexion with severe pain during the whole arc of motion. Left knee ROM was 5-90° of flexion with pain on terminal movements.

The plain radiographs of both knees, including weight-bearing anteroposterior (AP), laterolateral (LL), and axial patellar radiographs with knees bent to 45° were obtained (Figure 1a-1b, Figure 2a-2b). Patellar height was measured using the Insall-Salvati index (ISI) and the Caton-Deschamps index (CDI)[10,11]. Pre-

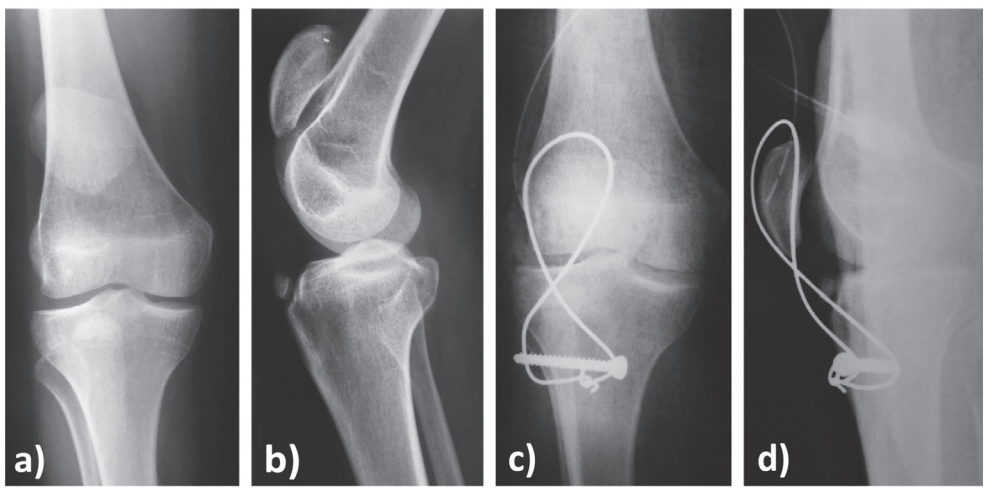

Figure 1. Preoperative weight-bearing anteroposterior (a) and laterolateral (b) radiographs of the right knee. Postoperative anteroposterior (c) and laterolateral (d) radiographs of the right knee.

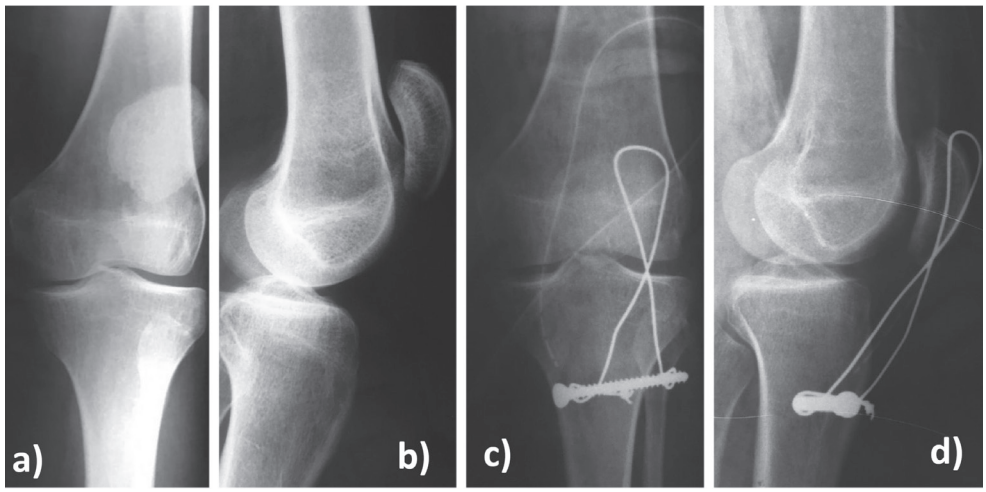

Figure 2. Preoperative weight-bearing anteroposterior (a) and laterolateral (b) radiographs of the left knee. Postoperative anteroposterior (c) and laterolateral (d) radiographs of the left knee.

operative right knee ISI was 1,46 and CDI 1,83, and for the left knee ISI was 1,33 and CDI was 1,43.

After evaluation and confirmation of the diagnosis of symptomatic bilateral patella alta, relief of the symptoms was attempted by administering a BTX-A in the distal right quadriceps. Two weeks after the injection, the spasticity of the right quadriceps muscle was attenuated but the pain and limited ROM persisted. One month after the injection, while the quadriceps muscle was still relaxed due to the BTX-A administration, we performed a patellar tendon shortening procedure. With the patient in spinal anaesthesia, a midline anterior prepatellar incision of the skin and subcutaneous tissue was performed and the extensor apparatus of the knee was visualised, together with the cranial position of the patella clearly notable. Firstly, the cortical screw was inserted in the coronal plane through the tibial tubercle from medial to lateral. Then, just proximal to the base of the patella, a cerclage wire was passed through the quadriceps tendon and then pulled distally around the patella crossing the wires in the figure-of-eight manner. Using the wire, repositioning of the patella was attempted, but still, the tension in the quadriceps was too high for the complete reduction of the patellofemoral joint. Consequently, a V-to-Y plasty of the quadriceps tendon was performed. Then, the patella was positioned so that the distal end of the patella was in line with the tibiofemoral joint line, close to its normal anatomical position. The plication of the patellar tendon was achieved with Ethibond 2 and Vicryl 2 sutures. The cerclage wire was then secured around the cortical screw in the proximal tibia, thus completing the augmentation of the patellar tendon using the McLaughlin wiring technique[12]. Postoperative ISI and CDI were 0,64 and 0,74, respectively (Figure 1c-1d). After surgery, the leg cylinder cast with the knee in full extension was applied for a period of 6 weeks, and afterward cautious physical therapy commenced. The surgery and postoperative rehabilitation period went uneventful. Eight months after the surgery, the cerclage wire and the screw were extracted from the right knee.

Simultaneously with development of right-sided anterior knee pain and limited ROM, similar symptoms but to a lesser extent occurred in the left knee. After the right knee surgery, during the rehabilitation period, the left knee succumbed to an increased load and symptoms affecting the left knee aggravated. Left knee ROM decreased to 5-50° of flexion and the pain intensified. Thus, the identical procedure, with a BTX-A injection administered one month preoperatively, was performed on the left knee, fifteen months after the surgery on the right knee, with the same postoperative rehabilitation protocol. Postoperative ISI and CDI were 0,69 and 0,62, respectively (Figure 2c-2d). The cerclage wire and the screw were extracted from the left knee six months after the surgery.

On the final follow-up, 21 months after the right knee surgery, and 6 months after the left knee surgery, the patient was very satisfied with the outcome. She didn't experience any pain in her knees. There was no joint effusion, and the patellae were in a normal (an-

atomical) position, stable, and painless on palpation and compression. The ROM in both knees increased to 0-140° of flexion.

DISCUSSION

The main finding of our study is that the administration of BTX-A, coupled with PTA using the McLoughlin wiring technique, V-Y plasty of the quadriceps tendon and patellar ligament plication, represents a feasible option for treating patients with cerebral palsy and symptomatic patella alta. The main effect of BTX-A is a chemical denervation that takes place at the neuromuscular junction where BTX-A binds to presynaptic nerve terminals and in a biochemical cascade inhibits the release of acetylcholine. Intramuscular administration of BTX-A thus leads to a reversible flaccid paralysis that lasts for an average of 12 weeks with its maximum effect at 4-6 weeks after the injection[13]. Due to its well-known mechanism of action and benefits in the initial treatment of neurological disorders characterised by overactive muscle movement, indications for its use in modern medicine are ever-increasing, especially in treating various musculoskeletal disorders such as cerebral palsy, talipes equinovarus, torticollis, plantar fasciitis, lateral epicondylitis, and others[14].

For the aforementioned indications, BTX-A is commonly used as a single modality therapy. However, a few reports also describe the application of BTX-A in a perioperative setting with various positive results. For example, Barwood et al. compared two groups of children with spastic cerebral palsy undergoing adductor-release surgery to treat or prevent hip dislocation with one group receiving an intramuscular injection of BTX-A five to ten days before the scheduled date of surgery and the second group receiving normal saline. They found that the group of patients with a preoperative administration of BTX-A compared to that receiving placebo had a reduction in mean pain scores, reduction in mean analgesic requirements and a reduction in mean length of hospital admission, concluding that muscle spasticity as a significant cause of postoperative pain can be managed effectively by preoperative injection of BTX-A[15]. Eibach et al. report the use of BTX-A injections in hip flexor and adductor muscles ten days prior to total hip arthroplasty in a patient with tetra-spasticity and presumably a high risk of postoperative luxation. They found an improvement of position from 45° to 20° of hip flexion and from 20° to 10° of hip adduction along with decreased muscle tone in hip flexor and adductor muscles that lasted for eight weeks, thus concluding that preoperative administration of BTX-A can play an important role in reducing the risk of postoperative hip luxation[16]. Racette et al. report on two patients undergoing cervical spine fixation procedures due to severe cervical dystonia and developed progressive cervical myelopathy. Prior to surgery, they received a high-dose BTX-A for muscle relaxation and limitation of involuntary neck movements intending to facilitate cervical decompression and stabilisation, promote postoperative healing and permit the patients to better tolerate halo fixation[17]. The most recent study depicts perioperative BTX-A injections in three patients with three distinct causes of muscle spasticity and three different surgical procedures. The first patient sustained a C6 incomplete hemicord spinal cord injury with subsequent right hip adductor spasticity. Due to preexisting right hip osteoarthritis and worsening pain, the patient underwent a total hip replacement. Two weeks before surgery she received BTX-A injections to the hip adductors and tensor fasciae latae muscle in order to prevent hip dislocation, minimise postoperative increase of spasticity and enable early rehabilitation. The next patient suffered from a spasticity in her left hand following a stroke and due to progressive hand deformity she underwent extensor carpi radialis brevis tendon transfer, however, severe postoperative wrist extension and finger flexion overactivity caused tendon failure. During the emergency repair surgery of the ruptured tendons, she received BTX-A injections directly into her extensor digitorum communis and flexor digitorum superficialis muscles. Following the salvage surgery, she was able to wear the postoperative splint, she had only mild dystonia and spasticity and her tendons healed well. The third patient had progressive multiple sclerosis, subsequent spasticity in her hip adductors, and a chronic sacral wound with underlying osteomyelitis. The wound was covered using V-Y plasty and 12 days postoperatively she received BTX-A injections in her hip flexors and adductors as to help prevent dehiscence of the recent flap surgery and to facilitate pericare[18].

Regarding the surgical intervention on tendons, one animal model study showed that preoperative intramuscular injection of BTX-A reduces the stiffness of the injected muscle-tendon unit thus facilitating its surgical manipulation, while another study on surgically bisected Achilles tendons in rats proved that in-

traoperative BTX-A produces a reduction in muscle force, twitch and tetanic contractions of gastrocnemius leading to significantly higher tendon rupture force and consequently decreased rate of tendon rupture after the surgical repair of tendon[19,20].

To this date, there is no reported case of combining intramuscular administration of BTX-A and surgical procedure of patellar tendon advancement in treating patients with patella alta and spastic cerebral palsy. Based on the excellent outcome of our patient and benefits of preoperative BTX-A application demonstrated in aforementioned reports, we can conclude that such use of BTX-A is safe and effective in certain cases with a great potential to expand its utilisation. However, further research and well-designed clinical trials are needed to demonstrate efficacy and establish indications for perioperative BTX-A administration.

Disclosure of conflict of interest

There is no conflict of interest regarding the publication of this paper.

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Sažetak

NOVA METODA LIJEČENJA PATELE ALTE KOD BOLESNICE S CEREBRALNOM PARALIZOM: PRIKAZ SLUČAJA S PREGLEDOM LITERATURE

I. Levaj, I. Bohaček, T. Đapić i *D. Delimar*

Patela alta relativno je često stanje u ambulatornih bolesnika s cerebralnom paralizom, osobito onih koji se prezentiraju čučećim hodom. Liječenje simptomatske patele alte u takvih je bolesnika složeno i konzervativne metode liječenja rijetko daju zadovoljavajuće rezultate. Stoga je u većini slučajeva indicirano operacijsko liječenje u smislu izolirane distalizacije patele ili udružene s drugim kirurškim zahvatima. U ovom radu prikazan je slučaj udružene primjene konzervativne metode liječenja i kirurškog zahvata kod 26-godišnje bolesnice s cerebralnom paralizom i obostranom patelom altom. Mjesec dana prije kirurške intervencije apliciran je botulinum toksin tip A (BTX-A) u ipsilateralni mišić kvadricepsa. Posljedično smanjena spastičnost mišića ekstenzora koljena olakšala je postupak skraćivanja ligamenta patele i omogućila je sigurniju rehabilitaciju. Šest mjeseci nakon operacije pojedinog koljena bolesnica se nije žalila na bolove, a opseg pokreta oba koljena poboljšan je s 5-50° prije operacije na 0-140° poslije operacije. Ovim radom prvi smo izvjestili o uporabi prijeoperacijskog BTX-A u liječenju patele alte kod bolesnika sa cerebralnom paralizom. Dokaz o učinkovitosti i sigurnosti ove metode liječenja mogao bi potaknuti širu prijeoperacijsku primjenu BTX-A kako bi se, u slučajevima povišenog tonusa mišića, olakšala kirurška intervencija i unaprijedila poslijeoperacijska rehabilitacija.

Ključne riječi: *patela, patelarni ligament, cerebralna paraliza, spastičnost, botulinum toksini*