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Selective endoscopic neurotomy of the upper and lower subscapular nerves in a patient with dyskinetic cerebral palsy: a case report

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Cerebral palsy (CP) is the dominant cause of motor deficiency in young children, occurring in 2-3 per 1000 live births.[10] CP constitutes a heterogenous neurological condition with various etiologies. Three main groups of CP have been described by Surveillance of Cerebral Palsy in Europe, based on clear neurological signs indicating pathology in the cerebral motor systems (eg, spastic, ataxic, dyskinetic, but also mixed CP forms).[3]

Dyskinetic CP is challenging to manage and often causes the patient significant functional impairment and pain. Regardless of which subtype of dyskinetic CP is the subject of discussion—dystonic, athetoid, or a combination of both—it is difficult to achieve a balance and synchrony in the highly complex interplay between agonist and antagonist musculature. Dyskinetic CP is conventionally treated with botulinum toxin type A injections with the aim to suppress dyskinesia. Occasionally, this is not sufficient and surgical options need to be considered. However, conventional surgical options for shoulder dyskinesia such as humeroscapular joint arthrodesis, tenotomies, tendon transfers, or tendon elongations are often associated with risk of further functional impairment.

Case report

Oral informed consent was obtained from the patient and her mother to write this case report.

A female patient who had suffered severe perinatal asphyxia and developed postasphyxial CP was referred to our clinic for assessment by a hand surgeon at 4 years of age. Although not immediately obvious, she was later assessed as having a right-sided dyskinetic CP. The presenting situation was one of intermittent internal shoulder rotation, elbow extension, wrist flexion, and hand (intrinsic) dystonia. Other than in the right upper extremity, the patient also suffered manifestations of the dyskinetic CP in the ipsilateral leg, as well as neuromuscular scoliosis and epilepsy. She also has a mild degree of intellectual disability.

The upper extremity dyskinesia persisted and worsened over the years despite repeated administrations of botulinum toxin type A. In total, botulinum toxin type A was administered to a multitude of muscles in the right upper extremity on 17 occasions between ages 4 and 19 years (December 2007 through March 2022). Furthermore, the dearth of results from the treatments was noted already in 2010. For wont of other feasible options, the treatment was carried on with inconsistent results until the autumn of 2018, when it was interrupted. Deep brain stimulation has also not been successful in controlling the dyskinesia.

At around 2019 (when the patient was aged 16 years), the shoulder dyskinesia started to become painful. An orthopedic surgeon specialized in shoulder conditions was consulted and shoulder arthrodesis as well as subacromial decompression were considered but dismissed.

The patient appeared for a 30 minutes of follow-up visit with a hand surgeon in March 2021, during which she suffered numerous episodes of extremely painful shoulder dyskinesia, leaving her exhausted. The internal rotations were forceful enough to make a subacromial gap appear, indicating subluxation of the humeroscapular joint with each dyskinetic episode. The patient rated the

Institutional review board approval was not required for this case report.

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pain a 10 on the Visual Analog Scale (VAS). At this point in time, she had already been started on strong opioids and gabapentin for pain relief and had also developed a depression. Furthermore, the pain had forced her to reduce attendance at school. Her mother also informed us that she was now actively searching the internet for clinics offering euthanasia to help her get out of her suffering.

The subscapularis muscle, which had not been treated earlier, was identified as the main generator of the dystonia, and targeted in 3 subsequent administrations of botulinum toxin type A. This would alleviate the dyskinesia and pain to the extent that the patient would be able to manage without opioids for a few weeks following the treatments. But soon, the pain would relapse unabated.

In November 2021, the patient was presented with the suggestion of selective denervation of m. subscapularis. To better predict the effects of such an intervention, she was scheduled for a test-block of the upper and lower subscapular nerves. To do that, the patient was subjected to general anesthesia and the subscapular muscle was localized using ultrasonography. Using the same injection technique[2] with an anterior approach that we had earlier used for treatment with botulinum toxin type A, 30 mL of Ropivacaine 7.5 mg/mL were infiltrated into the lateral and central portions of the subscapular muscle. Anatomic studies have shown that these are the regions in which the upper and lower subscapular nerves enter and branch out into the subscapular muscle.[4-6,8] The patient was admitted to our ward to be monitored for inadvertent spillover of local anesthetic into other parts of the brachial plexus. No such spillover effect was noted in the form of loss of sensation or voluntary movement, other than internal rotation. She remained relieved of shoulder dystonia for 12 full hours, whereafter the dyskinetic movements along with the pain, gradually returned as the effect of the local anesthetic waned.

The patient was much satisfied with the effect of the test-block and reported she had not noticed any negative side effects such as unmasking of other involuntary movements. This was also confirmed by her mother who had observed her for the duration of the test-block. The patient now wanted to proceed with a complete denervation of the subscapular muscle.

In late March 2023, she was operated under general anesthesia in the beach-chair position, with her right arm resting on a side table. A routine joint arthroscopy using a 30-degree angle scope was performed through a posterior portal at the soft spot. Except for a somewhat distended joint capsule, no joint pathology was found. The scope was then shifted to the subacromial space. Bursectomy was performed using a shaver introduced through the lateral portal, until the coracoacromial ligament was clearly visualized and could be followed down to the coracoid process. Next, the scope was moved to an antero-lateral portal at the level of the coracoid process. Another portal, for the introduction of a standard radiofrequency ablation tool, was established 3 cm inferior to the first antero-lateral portal (Fig. 1).

The loose connective tissue between the conjoined tendon and the subscapular muscle was gently pried open as well as vaporized using a standard radiofrequency ablation tool, until the axillary nerve (Fig. 2) was visualized. The axillary nerve, itself a branch of the posterior cord, was used as a reference point to localize the lower subscapular nerve as it branched off from the posterior cord some 20 mm cephalad at about the level of the medial border of the base of the coracoid process. Once the nerve was found, gentle and momentaneous activation of the radiofrequency ablation tool induced muscular contraction of the subscapular muscle, confirming that the correct nerve had been identified. Thereupon it was severed using the same radiofrequency ablation tool. Following the posterior cord further medially, the thoraco-dorsal nerve was seen branching off. Then, by a slight external rotation of the arm

Figure 1 Scars showing the positioning of the portals used for the procedure.

and gentle dissection further medially and cranially, the upper suprascapular nerve was encountered and neurotomized after testing as previously described. The procedure was performed under continuous irrigation. Upon completing the procedure, the arm was immobilized with a sling for a few days.

The patient recovered quickly after surgery. On the 15th postoperative day, she reported pain at level 4 according to the VAS scale and was already making plans to return to school full-time. At 1 month after surgery, she had managed to discontinue all opioid

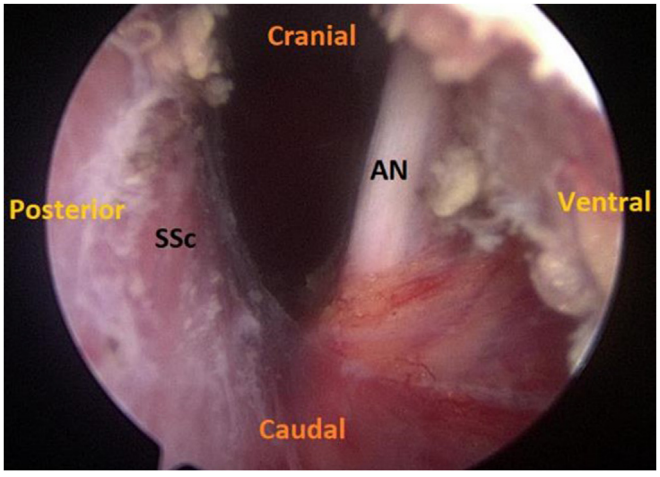

Figure 2 Intraoperative image depicting the axillary nerve (*AN*) in the center and the subscapular muscle (*SSc*) to the left.

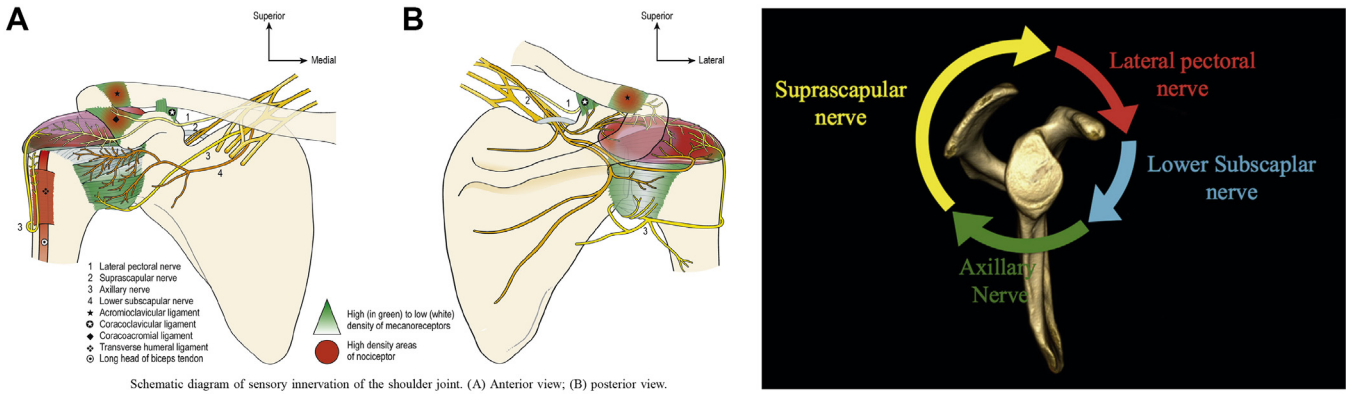

Figure 3 The sensory innervation of the human shoulder joint. (With permission from Laumonerie et al.[7] Reprinted with permission from the Journal of Shoulder and Elbow Surgery Board of Trustees (C) 2020).

pain killers. At follow-up 3½ months after surgery, the arm was calm, and we did not observe a single dyskinetic episode during the 1 hour we observed her at our outpatient department. The patient now rated her pain level at 3 according to the VAS scale. She appeared for the annual CPUP follow-up[1] some 4½ months after surgery and it was found that no shoulder movement had been lost while the passive shoulder range of motion was also still preserved. As this case report is being written, 10 months after the surgery, the patient has managed to graduate from upper secondary school and is now in part-time employment at a newspaper office and considering moving to her own apartment.

Discussion

When treating patients with dyskinetic CP, it is usually quite difficult to predict the results of the intervention beforehand. Not seldom, treating the predominant muscles in a dyskinetic pattern with botulinum toxin type A will unmask underlying dyskinetic movements of lesser magnitude. On other occasions, we might be targeting the right muscles, but the dose may be either inadequately low or excessive. Against that backdrop, performing selective neurotomies on patients with this specific type of CP can be a daring decision to make. Therefore, performing a selective local anesthetic block that mimics the effects of the intended neurotomy can help both in predicting the effects of such a procedure, as well as help the surgeon observe underlying but less prominent patterns of dyskinesia as they are unmasked under the effect of the local anesthetic block.

Several approaches have been described for the brachial plexus, but not all are ideal for accessing the upper and lower subscapular nerves for selective neurotomies. Power et al[9] have devised an open infraclavicular approach for neurectomy of the spastic shoulder, which makes it possible to perform neurectomies for multiple spastic muscles using the same surgical approach. It also has the advantage that the surgeon can perform intraoperative nerve stimulation to confirm which nerve is being examined. This approach could have been used for our case, but we opted for an endoscopic approach due to previous familiarity with endoscopic equipment and interventions. Similar accesses to ours have been described earlier, although not with the goal to denervate the subscapular muscle.

The described method of performing selective neurotomies by endoscopic access provides both a close view of pertinent nerves, as well as a great overview of local anatomical structures. Yet, it is recommended that the procedure be performed by well-experienced surgeons with good knowledge of shoulder anatomy and prowess in arthroscopic techniques. The procedure, while affording the surgeon a method of high precision, is admittedly also quite demanding to perform.

As was mentioned above, not only was the patient relieved of her shoulder dyskinesia but her pain was also significantly reduced. The explanation to that lies partly in the fact that the shoulder no longer goes into extreme internal rotation—thereby putting strain on local stabilizing structures, and partly in the fact that the lower subscapular nerve courses on to contribute a significant amount of nociceptive articular branches to the anteroinferior portion of the glenohumeral joint capsule[7] (Fig. 3). By severing this nerve, part of the glenohumeral joint capsule is consequently denervated.

As is obvious from her own previous medical history, dyskinesia has a natural tendency to change over time. It remains to be seen if the performed selective neurotomies will stand the test of time and suffice in keeping the shoulder dyskinesia, as well as the associated pain, at bay. Although too early to say, the presented case inspires the suggestion that selective neurotomies around the shoulder, when planned such that they result in partial denervation of the glenohumeral joint, constitute a good strategy in the management of the dyskinetic and painful shoulder where more conventional modes of treatment have failed to yield satisfactory results.

Seeing the positive outcome for this patient has been very encouraging and we now have a new tool to address severely dyskinetic subscapular muscles. To our knowledge, there are no previous publications on performing test-blocks for the dyskinetic subscapular muscle or on performing endoscopic selective neurotomies of the upper and lower subscapular nerves.

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