Botulinum toxin type A as an adjunct to the surgical treatment of the medial rotation deformity of the shoulder in birth injuries of the brachial plexus

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We retrospectively reviewed 26 patients who underwent reconstruction of the shoulder for a medial rotation contracture after birth injury of the brachial plexus. Of these, 13 patients with a mean age of 5.8 years (2.8 to 12.9) received an injection of botulinum toxin type A into the pectoralis major as a surgical adjunct. They were matched with 13 patients with a mean age of 4.0 years (1.9 to 7.2) who underwent an identical operation before the introduction of botulinum toxin therapy to our unit.

Pre-operatively, there was no significant difference (p = 0.093) in the modified Gilbert shoulder scores for the two groups. Post-operatively, the patients who received the botulinum toxin had significantly better Gilbert shoulder scores (p = 0.012) at a mean follow-up of three years (1.5 to 9.8).

It appears that botulinum toxin type A produces benefits which are sustained beyond the period for which the toxin is recognised to be active. We suggest that by temporarily weakening some of the power of medial rotation, afferent signals to the brain are reduced and cortical recruitment for the injured nerves is improved.

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Botulinum toxin type A has many clinical uses, including the treatment of ophthalmic,[1] neurological, gastrointestinal, urological, dermatological and orthopaedic disorders.[2] These applications are based on the peripheral inhibition of the release of acetylcholine at the presynaptic neuromuscular junction, thereby temporarily weakening the muscle.

In children with neuromuscular disorders, botulinum toxin type A has been used for the treatment of spasticity since 1990.[3] The injections are given in combination with physiotherapy, casting, splinting and orthoses to prevent or reduce hypertonicity and contractures, improve function, facilitate the tolerance of orthoses, and to assist in pre-operative evaluation.[2] More recently, botulinum toxin type A has been used to treat contractures of the biceps and triceps in children with birth injuries of the brachial plexus.[4,5] It has also been used as an adjunct during primary neurosurgical repair of these injuries.[6] We present our experience using botulinum toxin type A as an intra-operative adjunct in the later surgical treatment of medial rotation deformity in children who sustained an injury to the upper brachial plexus at birth.

In these patients there is often considerable muscle imbalance between the paralysed lateral rotators of the shoulder and the less involved medial rotators, leading to contracture and even dislocation and bony deformity. The accepted treatment is to release any contracture and restore active lateral rotation by a muscle transfer, usually the latissimus dorsi/teres major complex, into the rotator cuff.[6-9] As an adjunct we began to inject the pectoralis major with botulinum toxin type A intra-operatively during such transfers.[6] We have reviewed the results of this strategy in a series of cases and compared them with a group of patients who underwent an identical operation without the use of botulinum toxin type A.

Patients and Methods

We retrospectively reviewed 26 patients who underwent reconstruction for a medial rotation deformity of the shoulder after a birth injury to the brachial plexus at either Miami Children's Hospital or the Hospital for Joint Diseases between 1998 and 2003. Surgical release of the contracture using a subscapularis slide and transfer of the latissimus dorsi and teres major into the rotator cuff was performed in all patients. In 13 consecutive patients (group A), we injected the pectoralis major with 100 units of botulinum toxin type A at the end of the operation. We compared the outcome with that of 13 patients (group B) who did not receive botulinum toxin type A,

Table I. The modified Gilbert shoulder evaluation scale[10,11]

Grade	Description	Results
0	Completely paralysed - shoulder or fixed deformity	
1	Abduction = 45°	No active external rotation
2	Abduction < 90°	Bi-active external rotation
3	Abduction = 90°	Active external rotation < 10°
4	Abduction < 120°	Active external rotation 10° to 30°
5	Abduction > 120°	Active external rotation 30° to 60°
6	Abduction > 150°	Active external rotation > 60°

but who underwent an identical operation. The latter group was selected as a consecutive series from our database immediately before the introduction of botulinum toxin type A therapy in our practice.

Group A included six male and seven female patients with a mean age of 5.8 years (2.8 to 12.9) and involved five right and eight left shoulders. Group B comprised four male and nine female patients with a mean age of 4.0 years (1.9 to 7.2) and involved ten right and three left shoulders. Both groups of patients were immobilised in a modified shoulder spica cast for six weeks after surgery, and then began physiotherapy for a minimum of three months. In all patients, the shoulders were evaluated pre-operatively and at mean of three years (1.5 to 9.8) after surgery using a modified Gilbert scale (Table I).[10,11] No complications occurred and no patient underwent additional surgery.

We used Student's *t*-test to compare the difference in the outcome, with a p-value ≤ 0.05 considered to be significant.

Results

The patients in group A improved from a mean of 3.38 (2 to 4) to a mean of 5.31 (5 to 6) and those in group B improved from a mean of 2.85 (2 to 4) to a mean of 4.85 (4 to 5) on the modified Gilbert scale. Using the *t*-test, the pre-operative difference in the scores was not significant ($p = 0.093$) but post-operatively the difference was significant ($p = 0.012$).

Discussion

Our retrospective review of two comparable groups of children with medial rotation deformity of the shoulder from birth injuries to the brachial plexus, showed that those who received injections of botulinum toxin type A into pectoralis major in addition to surgery had a better outcome than those who underwent the same procedure but did not receive botulinum toxin. We anticipated that injecting botulinum toxin would temporarily weaken the pectoralis major and would facilitate the maintenance of lateral rotation of the shoulder and aid in the rehabilitation of the transferred muscles. This was accomplished immediately after surgery. More importantly, a sustained improvement in shoulder function was noted at a mean follow-up of three years (1.5 to 9.8), well after the effects of the botulinum toxin would have worn off at between ten and 15 weeks.

The treatment of birth injuries to the brachial plexus has always focused on the nerve lesion and recovery of distal joints and muscles, but the central nervous system may have a greater role than previously thought. In a study of electromyographic data in patients with brachial plexus palsy,[12] significant weakness and clumsiness were observed in arms which were adequately innervated. It was concluded that these children had an underlying developmental apraxia and it was suggested that the motor cortex failed to completely construct the electrical pathways for the developing limb. Sundholm, Eliasson and Forssberg[13] reported poor grip strength in patients with a brachial plexus palsy which did not involve the hand. These data support our assumptions that the central nervous system is not merely a passive agent in the healing of such injuries. Our theory is that by temporarily weakening some of the power of medial rotation, afferent signals to the brain are reduced and cortical recruitment for the injured nerves is improved.

Curra et al[14] stated that "botulinum toxin type A affects the functional organisation of the central nervous system indirectly through peripheral mechanisms". Through its action on α and γ motor endings, botulinum toxin type A changes the afferent signals to various areas of the motor cortex of the brain, thereby altering mechanisms and spinal pathways. Because of the direct relationship between the muscle afferent input and the motor cortical output, more signals can be detected from the muscles affected by a birth lesion of the brachial plexus. Curra et al[14] also stated that "the botulinum toxin type A induced reduction in spindle signals could, therefore, alter the balance between afferent input and motor output, thereby changing cortical excitability".

We began to use botulinum toxin type A as a short-term measure to aid our post-operative therapy programme.[6,11] After reviewing our results, we discovered a difference in the final outcome of patients in whom botulinum toxin type A had been used, and we began routinely using it intra-operatively, to establish better muscle balance for the shoulder during early post-operative therapy. It appears that the use of botulinum toxin type A as an adjunct to these shoulder procedures in birth-related injuries to the brachial plexus is effective in improving the long-term results. We are currently undertaking a randomised, prospective outcome study.

Supplementary Material

A table detailing the pre- and post-operative modified Gilbert scores is available with the electronic version of this paper on our website at www.jbjs.org.uk

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