J Korean Neurosurg Soc 37 : 350-353, 2005

Clinical Article

# Selective Peripheral Denervation for the Treatment of Spasmodic Torticollis

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**Objective :** Various methods of treatment for idiopathic cervical dystonia have been tried in the past with unsatisfactory results. The authors report cases of five patients who underwent selective peripheral denervation for spasmodic torticollis.

**Methods :** Between July 2002 and December 2003, 5 patients underwent surgery at St. Mary's Hospital for spasmodic torticollis. Age of the patient at the onset of symptoms ranged from 29 to 56years (mean 43.75years). Selective peripheral denervation(SPD) was performed at 7 to 11months after the onset of symptoms (mean 8.75 months). A patient was considered to be the candidate for surgery if conservative methods were unsuccessful and symptoms persisted for longer than 7months. In addition, 2patients who refused treatment with botulinum toxin were also enrolled in this study.

**Results :** Although one patient underwent reoperation, all of the five patients' symptoms were improved after the operation. Clinically, patients with retrocollis showed better improvement than laterocollis patients.

**Conclusion :** Although injection of botulinum toxin is the first-choice in treatment modality, when surgery is required, selective peripheral denervation provides good results with minimum side effects.

**KEY WORDS :** Spasmodic torticollis · Cervical dystonia · Selective peripheral denervation · Botulinum toxin.

## Introduction

Spasmodic torticollis is the most common form of focal dystonia, characterized by deviation of the neck due to involuntary tonic or clonic contraction of cervical muscles[6]. The cause of spasmodic torticollis is unknown[5]. Spasmodic torticollis has been notoriously difficult to treat. Conservative methods such as pharmacotherapy, psychotherapy, and physotherapy have been highly unsuccessful, and a few symptomatic therapeutic options are available to date[5]. Recently, many authors reported good results after treating torticollis with SPD. We report 5patients with spasmodic torticollis who underwent selective peripheral denervation procedures.

## Materials and Methods

Between July 2002 and December 2003, 5patients (4 males and 1 female) underwent surgery at St. Mary's Hospital for spasmodic torticollis. The age of the patient at the onset of symptoms ranged from 29 to 56years (mean 43.75years).

Selective peripheral denervation was performed at 7 to 11 months after the onset of symptom (mean 8.75months). Patients with laterocollis had a history of botulinum toxin injection, but with unsuccessful results. Two retrocollis patients who refused botulinum toxin injection were enrolled in this study.

Preoperatively, the patients complained of head position, muscle spasms, and pain. The severity of cervical dystonia was assessed with The Toronto Western Spasmodic Torticollis Rating Scale(TWSTRS), Cervical spine Dystonia Severity Scale (CDSS) and, Visual Analogue Scale(VAS).

The involved muscles were identified by clinical examination and a multichannel electromyographic(EMG) recording(Table 1). Simultaneous recordings obtained from both *sterno-cleido-mastoid*(SCM) and splenius capitis muscles were mandatory, and when necessary, from both trapezius muscles as well. Dystonia was documented on videotape. A magnetic resonance imaging study of the brain was performed to rule out a intracranial lesions.

Bilateral posterior selective peripheral denervation was performed in the two retrocollis patients. Denervation of the involved SCM muscle and unilateral posterior selective denervation was performed in the three laterocollis patients.

• Received : September 10, 2004 • Accepted : November 10, 2004
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**Table 1.** Clinical data of five patients

| Case NO. | Sex/Age | Type of torticollis | Previous botulinum toxin injection | Involved muscles on EMG |
|---|---|---|---|---|
| 1 | F/56 | Retrocollis | Refuse | Bilateral SCM |
| | | | | Bilateral splenius capitis muscle |
| 2 | F/43 | Laterocollis, Lt | Yes | Rt. SCM and trapezius muscle |
| | | | | Lt. splenius and scalenus muscle |
| 3 | M/29 | Laterocollis, Rt | Yes | Lt. SCM |
| | | | | Rt. cervical PSM |
| | | | | Rt. Splenius muscle |
| 4 | M/47 | Laterocollis, Rt | Yes | Lt. SCM |
| | | | | Lt. upper trapezius muscle |
| | | | | Rt. Splenius capitis muscle |
| 5 | M/49 | Retrocollis | Refuse | Bilateral SCM |
| | | | | Bilateral splenius capitis muscle |

Rt : Right , Lt : Left , EMG : Electromyography , PSM : Paraspinal muscle, SCM : Sterno cleido mastoid Muscle

**Fig. 1.** The bilateral posterior branches of C1−5 are identified after meticulous dissection of posterior neck muscles.

## Surgical Technique

In all cases, surgery was performed after induction of general anesthesia with the patient in a prone position and his or her head fixed in a Mayfield clamp. The different nerve branches were identified using monopolar stimulation, and therefore we were able to refrain from administering muscle relaxants.

In laterocollis patients, surgery is initiated with the denervation of the SCM muscle. The exit point of the spinal accessory nerve from this muscle in the lateral neck triangle is estimated by applying a supramaximal transcutaneous electrical stimulation (2Hz). Using a 5-cm skin incision on the posterior muscle margin, the trapezius branch of the spinal accessory nerve is first identified and then followed proximally under amplification. Injury of the greater auricular nerve crossing the operative field should be carefully avoided. Once the main trunk of the spinal accessory nerve is reached, its branch or branches to the SCM muscle can be detected by electrical stimulation, severed, and then resected widely. The SCM muscle may be further innervated through anterior branches of the C-1 and C-2 nerve roots or through the recurrent nerves leaving the trapezius branch of the spinal accessory nerve. These nerves must be resected as well.

In the second step, the involved posterior neck muscles are denervated via a separate skin incision. Selective peripheral denervation of the neck muscles can be conducted superselectively, *ie*, with any of the following muscles denervated separately : splenius capitis, semispinalis capitis or cervicis, and inferior oblique muscles. We recommend not only the denervation of the involved splenius capitis muscle but also the complete denervation of the autochthonous muscle group on the ipsilateral side.

The posterior branches can be best identified extraspinally, lateral to the joint facets, leaving the anterior branches innervating the shoulder and the arm muscles intact. The posterior branches are approached via a midline skin incision in the neck, running from the external occipital protuberance down to the C-7 spinous process, allowing both the left and the right side to be surgically treated. Having reached the C2-6 spinous processes, the inferior oblique capitis muscle is detached from its origin at the C-2 spinous process. Using microsurgical technique, we then enter the cleavage plane between the semispinalis capitis and cervicis muscles on the involved side. Blunt dissection is performed down to the area lateral to the articular facets until the level of the multifidus muscle is reached. The large posterior branch of C-2 (greater occipital nerve) is easily localized beneath the inferior oblique muscle. The most difficult nerve to identify is the posterior branch of C-1 (suboccipital nerve). However, it is most often identified 1.5 to 2cm lateral off the midline above the arch of the atlas and below the vertebral artery within the vertebral sulcus.

The posterior branches of C3-6 nerves are found on a perpendicular plane between the lateral margin of the semispinalis cervicis muscle and the medial margin of the semispinalis

capitis muscle. Whereas the posterior branches of C3-4 are easily identified lateral to the facets, the tiny branches of C5-6 are found medially, running on the surface of the semispinalis cervicis muscle(Fig. 1). After resection of all the posterior branches of C1-6, the entire region lateral to the articular facets are stimulated and observed for any further muscle contractions. Often, additional posterior branches can be found within the multifidus layer, which also have to be resected. To prevent nerve regeneration, all the proximal and distal nerve stumps are extensively coagulated.

In retrocollis patients, only bilateral posterior selective denervation was carried out.

## Results

Of the five patients operated on, one patient underwent reoperation, because the symptoms of laterocollis recurred. Again, the involved muscles were identified using EMG recording. EMG recordings demonstrated reinnervation of previously denervated muscles. The surgical site was reopened, and a search was made for nerves that might have spontaneously regenerated.

The patients' symptoms were assessed by TWSTRS, CDSS, and VAS scores. All the patients who underwent surgery had improved scores(Table 2). The postoperative day 2 to 3 showed improvement in the retrocollis patients slightly less than that of laterocollis patients. Conversely, after 3 months, the abnormal positions and dystonia in retrocollis patients were more improved than in laterocollis.

In this study, complications related to selective peripheral denervation such as significant swallowing difficulties, neck muscle weakness, or sensory change were not observed.

## Discussion

Torticollis is an involuntary contracture of cervical muscles. The involved muscles can be identified by EMG test and abnormal posture of the head.

In this study, the contralateral SCM was involved in laterocollis and bilateral SCM in retrocollis patients. The ipsilateral scalenius and trapezius muscles were involved in laterocoliis patients. The musculature involved in torticollis includes the SCM muscle, which is active in more than 75% of all previous torticollis cases[12]. Posterior neck muscles such as the splenius capitis and cervicis, obliques inferior, and rectus capitis posterior also play a major role in producing torticollis. In the reoperation case of laterocollis in this study, reinnervation of contralateral SCM and ipsilateral scalenius muscles, were examined to play a major role in the development of laterocollis.

The most recent breakthrough in the treatment of spasmodic torticollis was the introduction of the botulinum toxin administration in the 1980s. Botulinum toxin injections have been widely accepted as a good therapeutic option since they unify low-risk and promise results.

A minority of reported patients, however, failed to respond and in some, the symptoms become refractory to botulinum toxin treatment with repeated injections, often due to the development of anti-botulinum toxin antibodies[1]. In these cases a more invasive treatment alternative need to be considered. However, stereotatic procedures such as cervical rhizotomy have also been largely abandoned due to subsequent severe side effects that sometimes developed[17].

The current choice of operation is selective peripheral denervation, which aims to denervate muscles responsible for the abnormal movement while preserving innervation to those that do not participate in dystonia[4].

Bertrand and colleagues[3,4] developed a selective peripheral denervation method, which has been shown to be highly successful and without major risks. In their hands, excellent or good results have been demonstrated in 88% of the patients[3,4]. They also reported that success depends on (1) a careful selection of patients and of the appropriate denervation; (2) a meticulous and complete denervation during operation; and (3) early and intensive postoperative physical theraphy.

The results reported by Arce and Russo[2], Davis, et al.[8], and Dieckmann[9] have been promising as well. Braun[5] reported that symptoms recurred in 11% of the patients. Fifteen patients among the 155 patients had to undergo second surgery. and nearly 80% of the patients in his study group can be effectively treated without major risks and with good long-term prognosis.

Cohen-Gadol[6] reported that 62% of the patients who did not respond to botulinum toxin were

**Table 2.** Surgical outcome of selective peripheral denervation

| Case NO. | Sex/Age | Type of torticollis | TWSTRS | | CDSS | | VAS | |
|---|---|---|---|---|---|---|---|---|
| | | | Preop | Postop | Preop | Postop | Preop | Postop |
| 1 | F/56 | Retrocollis | 17 | 5 | 12 | 4 | 4 | 2 |
| 2* | F/43 | Laterocollis, Lt | 15 | 2 | 8 | 6 | 10 | 2 |
| 3 | M/29 | Laterocollis, Rt | 18 | 4 | 14 | 6 | 10 | 0 |
| 4 | M/47 | Laterocollis, Rt | 20 | 3 | 15 | 7 | 9 | 2 |
| 5 | M/49 | Retrocollis | 53 | 20 | 17 | 10 | 10 | 2 |

TWSTRS : Toronto Western Spasmodic Torticollis Scale, CDSS : Cervical spine Dystonia Severity Scale, VAS : Visual Analogue Scale, Preop : Preoperation, Postop : Postoperation, Lt : Left , Rt : Right , * : Reoperation case

satisfied with the result of surgery. And nearly 80% of the patients in 168 cases were effectively treated by surgery without major risks and with good long-term prognosis.

Recently, there have been several preliminary reports on the use of pallidal stimulation in patients with generalized dystonia[18]. Islekel, et al[13] treated one patient and Krauss, et al[15], treated three patients with spasmodic torticollis using pallidal stimulation. It is widely known that the effect of stimulation tends to decrease within a few years. Thus, although these patients improved immediately after the implantation of the electrical device, the long-term effect of deep brain stimulation in patients with local cervical dystonia is not yet known.

## Conclusion

SPD is recommended for patients who do not respond or no longer respond to botulinum toxin therapy. SPD provides good results and minimum side effects.

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