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# RESEARCH PAPER

# Selective peripheral denervation for cervical dystonia: long-term follow-up

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# ABSTRACT

**Objective** 61 procedures with selective peripheral denervation for cervical dystonia were retrospectively analysed concerning surgical results, pain, quality of life (QoL) and recurrences.

**Methods** The patients were assessed with the Tsui torticollis scale, Visual Analogue Scale (VAS) for pain and Fugl-Meyer scale for QoL. Evaluations were performed preoperatively, early postoperatively, at 6 months, then at a mean of 42 (13–165) months. All patients underwent electromyogram at baseline, which was repeated in cases who presented with recurrence of symptoms after surgery.

Results Six months of follow-up was available for 55 (90%) of the procedures and late follow-up for 34 (56%). The mean score of the Tsui scale was 10 preoperatively. It improved to 4.5 (p<0.001) at 6 months, and 5.3 (p<0.001) at late follow-up. VAS for pain improved from 6.5 preoperatively to 4.2 (p<0.001) at 6 months and 4 (p<0.01) at late follow-up. The Fugl-Meyer score for QoL improved from 43.3 to 46.6 (p<0.05) at 6 months, and to 51.1 (p<0.05) at late follow-up. Major reinnervation and/or change in the dystonic pattern occurred following 29% of the procedures, and led in 26% of patients to reoperation with either additional denervation or pallidal stimulation. **Conclusions** Selective peripheral denervation remains a surgical option in the treatment of cervical dystonia when conservative measures fail. Although the majority of patients experience a significant relief of symptoms, there is a substantial risk of reinnervation and/or change in the pattern of the cervical dystonia.

# INTRODUCTION

Spasmodic torticollis is a condition characterised by focal dystonia involving the cervical muscles with an incidence of 1.2/100 000.<sup>1</sup>

The mainstay of treatment is chemical denervation by means of intramuscular injections of botulinum toxin.<sup>2 3</sup> However, not infrequently, patients either do not respond to botulinum injections, or develop secondary resistance.<sup>4–6</sup> In those cases surgery may be considered. Historically, several surgical techniques have been tried aiming at symptom relief, and keeping in mind that no cure from the disease is to be expected.

Today, there are three main surgical approaches used: two approaches involve extradural selective peripheral denervation (SPD), as suggested by Bertrand, or a modification of the Bertrand technique, which includes intradural C1-2 ventral rhizotomy as suggested by Taira and coauthors.<sup>7 8</sup> The third approach is posteroventral pallidal deep brain

stimulation (DBS), which has emerged following the positive effect of posteroventral pallidotomy on Parkinsonian dystonia-dyskinesias<sup>9–13</sup> or, as has recently been suggested, subthalamic nucleus DBS.<sup>14</sup> The optimal indication, and the proper selection of patients for the different surgical approaches, are still to be established.

Pallidal DBS for cervical dystonia is relatively new, and encouraging long-term results are emerging.<sup>13 15 16</sup> Although SPD has been available for a longer period of time, it has not been widely practiced, and the published reports are few compared with the number of reports dealing with botulinum injections, or with DBS. Also, the last publication of a large series of patients undergoing SPD is 10 years old,<sup>17</sup> and in some of the previous studies, the results were vaguely described using semiguantitative evaluation scales. Other reports have used torticollis scales such as the Tsui scale, the Toronto Western Spasmodic Torticollis Rating Scale (TWSTRS), or the modified TWSTRS.<sup>18–20</sup> Finally, the results in terms of pain relief and quality of life (QoL), and analysis of observed recurrences, have seldom been reported.

At our institution we have performed SPD for cervical dystonia since 1993. In this paper we report on the long-term results of 61 procedures in 54 patients with a mean follow-up time of 42 months (range: 12–165).

# METHODS

# Patients

All except three patients selected for surgery were secondary non-responders to botulinum toxin injections as defined by lack of benefit by the referring neurologist. Preoperatively, all patients underwent MR imaging of the cervicocranial region to exclude pathological conditions that could induce a cervical dystonic state. Preoperative investigations included bilateral simultaneous needleelectromyogram (EMG) of the muscles of the neck during habitual position of the head, to define the muscles involved; EMG targeted the sternocleidomastoideus, the splenius capitis, the semispinalis, the levator scapulae and the upper trapezius muscles. The EMG was performed by the neurophysiologist in the presence of the neurosurgeon(s). Since the denervation surgery would preferably be performed with the patient in a sitting position, with risk for air embolism, an echocardiography was performed to exclude cardiac abnormalities such as a patent oval foramen. Patients with pathological hyperactivity in muscles accessible for denervation were selected for peripheral denervation, while those with involvement in muscles not accessible for denervation, such as the scalene muscles, were selected for pallidal stimulation. Table 1 shows the patients' demographic data and those muscles that were selectively denervated.

# Surgery

Peripheral denervation of the splenius, the semispinal, and the sternocleidomastoid (SCM) muscles was performed according to the method of Bertrand, and denervation of the levator scapulae as described by Taira *et al* and Anderson *et al.*<sup>21–23</sup> In addition, a section of the accessory nerve to the trapezius muscle was performed at the posterior border of the SCM muscle in six patients. Before the sectioning of any nerve, bipolar electrical stimulation at 4 Hz was performed to confirm its selective innervation of the affected muscle. The different denervation procedures are presented in table 1.

All except one patient were operated on in the sitting position with monitoring of eventual air-embolism using transoesophageal echocardiography. One patient was operated on in the prone position due to a cardiac patent oval foramen. For safety reasons, all patients had a central venous catheter installed to allow evacuation of an eventual air embolus, if needed.

## **Evaluation**

All patients were videotaped and evaluated by an experienced physiotherapist (EL) preoperatively and early postoperatively, usually on day 5 after surgery, and at all follow-up occasions, that is, at 6 months postoperatively and onwards until the last follow-up. The Tsui scale for cervical dystonia was used for assessment.<sup>18</sup> Since 1999, a Visual Analogue Scale (VAS) was administered to rate pain and the Fugl-Meyer life-satisfaction questionnaire<sup>24</sup> was used to rate QoL. The Tsui scale has a

Table 1 Characteristics of 54 patients with spasmodic torticollis
treated with a total of 61 procedures of peripheral selective
denervation

Age (mean and range)	53.3 (30–69) years
Sex	21 male/33 female
Duration of disease (mean and range)	15.96 (4–49) years
Muscles selectively denervated (n (%) of patients)	
Sternocleido and contralat splenius/semispinal	22 (36)
Sternocleido and bilat splenius/semispinal	8 (13)
Bilat splenius/semispinal	7 (11)
Unilat splenius/semispinal	6 (11)
Unilat sternocleido	5 (8)
Sternocleido and ipsilat splenius/semispinal	2 (3)
Sternocleido and ipsilat splenius/semispinal and upper trapezius	2 (3)
Lev scapulae and ipsilat upper trapezius	2 (3)
Sternocleido and ipsilat splenius/semispinal and lev scapulae	1 (2)
Sternocleido and contral splenius/semispinal and upper trapezius	1 (2)
Sternocleido bilat and bilat splenius/semispinal	1 (2)
Sternocleido and ipsilat upper trapezius	1 (2)
Unilat splenius/semispinal and sternocleido and lev scapulae	1 (2)
Unilat splenius/semispinal and lev scapulae	1 (2)
Lev scapulae and upper trapezius	1 (2)

The muscles denervated in each patient are presented.

contralat, contralateral; ipsilat, ipsilateral; lev scapulae, levator scapulae; splenius, splenius capitis; Sternocleido, sternocleidomastoid; unilat, unilateral.

range of 0-25, where 25 is the worst possible state of the cervical dystonia. The VAS is a subjective rating by the patient on a scale from 0 to 10, where 10 indicates maximal pain. The Fugl-Meyer questionnaire rates 12 different aspects of life quality on a scale from 1 to 6, where 6 represents maximal satisfaction with life.

In patients with recurrence of their torticollis or with a changed pattern of their cervical dystonia, a neurophysiological examination was performed at follow-up.

# RESULTS

A total of 54 patients with cervical dystonia underwent SPD. Demographic data are presented in table 1. In 15 patients the surgery was staged in two sessions, one for denervation of the SCM muscle and the other for denervation of posterior cervical muscles. The two-staged procedures were performed within a few months of each other and were analysed as one surgery. In three patients cervical dystonia was the dominant symptom in a context of a generalised DYT-1 negative dystonia. Six patients with recurrence of torticollis and EMG-verified reinnervation of previously denervated muscles were reoperated on with selective denervation more than 15 months after the initial SPD procedure, and one of them was reoperated on twice. These patients were included in the follow-up, which resulted in a total of 61 surgical procedures.

Since our centre is the only one in Scandinavia performing the Bertrand/Taira procedure, and several patients were referred from other Scandinavian countries, we encountered logistical issues that affected our ability to conduct a long-term follow-up. The 6 months follow-up was available for 55 of the 61 surgical procedures (90%), and evaluation at long-term follow-up at a mean of 42 (13–165) months postoperatively could be completed for 34 procedures (56%). One patient died due to an unrelated cause before long-term follow-up could take place.

# **Surgical results**

The surgical results at different time points are presented in table 2. An immediate improvement of the dystonic state was achieved in all but one patient. The positive effects were still

			Mean improvement	
	Preoperative	Follow-up	in per cent	p Value
Cervical dystonia scale (Tsui)				
5 days	10.0 (4.0)	3.9 (2.1)	61.0	<0.0001
postoperative (n=61)				
6 months postoperative (n=55)	10.4 (3.9)	4.5 (2.7)	56.7	<0.000
Long-term follow-up (n=34)	9.7 (4.0)	5.3 (3.3)	45.3	< 0.000
VAS for pain				
6 months postoperative (n=38)	6.5 (2.6)	4.2 (2.6)	35.3	<0.000
Long-term follow-up (n=27)	6.1 (2.5)	4.0 (2.9)	34.4	<0.01

Mean values and (SD) of Tsui's Cervical Dystonia Scale and of a VAS for pain are shown. Wilcoxon signed rank test was used for statistical analysis. VAS, Visual Analogue Scale

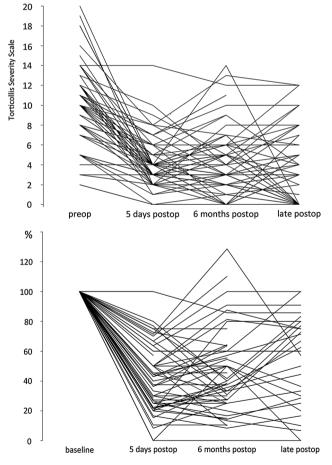
present at further follow-ups in a majority of the patients. Individual results are presented in figure 1. Despite the mean improvement in the Tsui scale ratings, the individual results showed a considerable variability.

# Pain and QoL

The evaluation of pain by VAS was performed in the last 48 procedures. Thirty-eight (79%) procedures were evaluated at 6 months and 27 (56%) at long-term follow-up, showing a significant improvement of pain at both follow-up time points (table 2). QoL was assessed in the last 50 procedures, at 6 months in 37 patients (74%) and at long-term follow-up in 27 patients (54%). For life-quality in general, most patients reported a modest but significant and sustained improvement. When analysing the different domains individually, only somatic health and psychological health were rated as improved at 6 months follow-up, but not at longer term. However, the total score was significantly improved at 6-month and also at late follow-up (table 3).

# **Complications/side-effects**

The surgical complications and neurological side-effects were few with exception of the expected sensory loss in the C2



**Figure 1** Surgical results following selective peripheral denervation for cervical dystonia in 54 patients/61 procedures. Assessment performed using the Tsui torticollis severity scale preoperatively, 5 days after surgery, at 6 months and late postoperative at mean of 42 (range: 13–165) months after surgery. Upper figure: individual scores of the Tsui scale. Lower figure: percentage changes in severity of dystonia in individual patients.

 Table 3
 Life satisfaction according to Fugl-Meyer scale before and after selective peripheral denervation

	Preoperative	Follow-up	p Value
Life satisfaction, in general			
6 months postoperative	2.9 (1.4)	3.7 (1.3)	<0.01
Long-term follow-up	3.6 (1.1)	4.1 (1.8)	<0.05
Occupation			
6 months postoperative	2.8 (1.8)	3.0 (1.9)	0.301
Long-term follow-up	3.5 (2.0)	3.4 (1.9)	0.745
Economy			
6 months postoperative	4.0 (1.3)	4.0 (1.4)	0.827
Long-term follow-up	4.0 (1.3)	4.3 (1.4)	0.059
Leisure			
6 months postoperative	3.0 (1.2)	3.6 (1.1)	0.055
Long-term follow-up	3.7 (1.1)	4.0 (1.2)	0.158
Social contacts			
6 months postoperative	4.3 (1.3)	4.3 (1.0)	0.896
Long-term follow-up	4.5 (1.4)	4.7 (1.1)	0.693
Family life			
6 months postoperative	4.6 (1.2)	4.9 (0.9)	0.263
Long-term follow-up	4.5 (1.1)	4.1 (1.4)	0.073
Partner relation			
6 months postoperative	4.1 (1.8)	4.0 (1.9)	0.543
Long-term follow-up	4.1 (1.7)	4.3 (1.4)	0.537
Sexual life			
6 months postoperative	3.4 (1.6)	3.5 (1.6)	0.783
Long-term follow-up	3.8 (1.5)	3.8 (1.5)	0.765
Personal ADL			
6 months postoperative	4.8 (1.3)	5.1 (0.9)	0.071
Long-term follow-up	4.9 (1.2)	5.1 (1.2)	0.429
Instrumental ADL			
6 months postoperative	4.1 (1.3)	4.3 (1.2)	0.274
Long-term follow-up	4.3 (1.4)	4.5 (1.4)	0.446
Somatic health			
6 months postoperative	3.0 (1.5)	3.6 (1.1)	0.05
Long-term follow-up	3.3 (1.4)	3.8 (1.1)	0.054
Psychological health			
6 months postoperative	3.5 (1.4)	4.0 (1.2)	<0.05
Long-term follow-up	3.9 (1.2)	4.3 (1.5)	0.321
Life satisfaction, total score			
6 months postoperative	43.3 (9.5)	46.6 (9.7)	<0.05
Long-term follow-up	47.3 (9.2)	51.5 (12.7)	0.05

Six months follow-up in 37 patients and long-term follow-up in 27 patients. Wilcoxon signed rank test was used for statistical analysis.

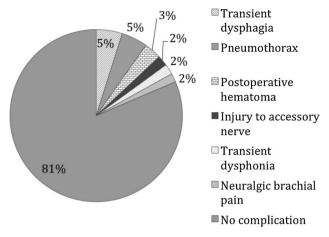
ADL, activities of daily living.

Bold figures represent statistically significant results.

region, which occurred in all patients who had denervation of the posterior C2 branch. A summary is presented in figure 2. Three patients had a transient dysphagia after posterior ramisectomy, and one of those also experienced a transient dysphonia. There were no complications affecting head position. In one patient, the accessory nerve was accidently injured, resulting in a weakness of the upper trapezius muscle, which had partially improved at late follow-up.

The rate of surgical complications was low, with the most frequent complication being a pneumothorax in three patients (5%), caused by the insertion of the central venous catheter through the subclavian vein. Therefore, the subclavian route for central venous line was abandoned in favour of a peripherally inserted central catheter (PIC-catheter). No case of significant air embolism was encountered in this series.

# **Movement disorders**



**Figure 2** Postoperative complications following 61 selective denervation procedures in patients with cervical dystonia.

In two patients, a postoperative haematoma in the SCM region required surgical evacuation. In one patient, the central line inserted through the jugular vein resulted in a postoperative transient severe neuralgic pain in the shoulder region.

# Recurrences and changes in pattern of muscular activity

Postoperative recurrences and/or change in the pattern of dystonia are presented in table 4. The major postoperative issue was a change in the pattern of pathological neck muscle activity. Following 31 procedures (51%) there was a change in pattern of dystonia with pathological muscle activity in preoperatively nonactive muscles. However, this was mild in most cases (18 patients, 29%). A reinnervation of denervated splenius was

Table 4The most noticeable findings regarding reinnervation or<br/>postoperative progression of disease (change in pattern) following<br/>61 procedures with selective peripheral denervation in 54 patients<br/>with cervical dystonia

	Degree of	Degree of affection		Reoperation	
	Minor degree	Major degree	Repeated or additional denervation	Pallidal stimulation	
Muscles reinnervate	d without cha	nge in patter	n		
Splenius	2 (3%)	4 (7%)	2 (3%)	1 (2%)	
Semispinal					
Sternocleido	3 (5%)	1 (2%)	1 (2%)		
Trapezius					
Muscles reinnervate	d and change	in pattern			
Splenius	5 (8%)	9 (15%)	4 (7%)	5 (8%)	
Semispinal					
Sternocleido		2 (3%)	1 (2%)	1 (2%)	
Trapezius					
Progressive disease/change in pattern without reinnervation	13 (21%)	2 (3%)		1 (2%)	
Total	23 (38%)	18 (29%)	8 (13%)	8 (13%)	

In some patients the reinnervation may be bilateral. A pathologic muscular activity that significantly affected the normal cervical posture was defined as 'major degree' while an activity that did not imply any major impact was defined as 'minor degree'.

found following 20 procedures (33%), and six patients (10%) had a reinnervated SCM muscle. In most patients the change in pattern and/or the reinnervation was mild to moderate as shown in figure 1 and tables 4 and 5, and the patients still showed a significant improvement in their initial torticollis. In total, after 41 (67%) procedures there was a change in muscular pattern, a reinnervation, or both. However, clinically relevant symptoms of major concern, due either to recurrences or changed pattern, occurred after 18 (29%) procedures. The timing of reinnervation and change in muscular pattern was highly individual as demonstrated in figure 1.

In some patients the recurred symptoms became severe later on and a reoperation was indicated. Eight patients (13%) were reoperated with repeated denervations. Five of those were operated on more than 15 months after primary surgery; they are included in the series and in the follow-up protocol. One of them was reoperated on twice due to aggressive reinnervation of the splenius. After reoperation, three patients (37%) were considerably improved and five were unchanged in their head position at a mean of 29.4 months follow-up (range 12–46). Their mean preoperative Tsui score was 7.4 (range 3–11) and the late postoperative score 5.7 (range 3–11; p=0.104). Their mean total score on the QoL scale improved from 52.8 to 59.3 (p=0.028).

In eight patients (13%) the cervical dystonia progressed and showed a widespread activity involving neck muscles available for denervation. These patients therefore underwent bilateral pallidal stimulation with, in most cases, a positive result as reported in a previous publication.<sup>25</sup>

# DISCUSSION

# Surgical results

SPD is a procedure that has been in the neurosurgical armamentarium for dystonia for many years. Lately, pallidal DBS has almost completely supplanted SPD for cervical dystonia. However, DBS is an invasive intracerebral operation and severe complications may occur.<sup>26</sup> On the other hand, SPD is a procedure completely outside the central nervous system, being an extradural procedure. Therefore, we believe that a peripheral procedure such as SPD may still be an option to consider for cervical dystonia affecting muscles readily accessible for denervation. However, as with all functional neurosurgery, the selection of patients is important. Almost all patients in our series had strict local pathologic muscle activity in a few muscles accessible for denervation. In patients with widespread cervical activity, that is, with clinically significant muscular activity

Table 5Numbers and percentages of minor and majorre-innervations according to the denervated muscles, following61 denervation procedures in 54 patients with cervical dystonia

Muscle	Denervated (n)	Minor reinnervation (n)	Major reinnervation (n)
Sternocleidomastoid	45	3 (7%)	3 (7%)
Splenius	70	19 (27%)	15 (25%)
Semispinal	70	6 (9%)	0
Upper trapezius	6	0	0
Levator scapulae	4	0	0
Total	190	28 (15%)	18 (9%)

outside of the muscles accessible for denervation, pallidal DBS should be the surgical procedure of choice.

Owing to the fact that our patients were referred from a very wide geographical area encompassing the Scandinavian countries, we had difficulties in achieving a proper long-term follow-up on all patients. However, as shown in tables 2 and 3, the subgroup of patients who we were able to follow for a longer time had a preoperative status comparable to the overall group, indicating that the patients with long-term follow-up should be representative for the whole series of patients.

In this series of treatment, which included refractory patients in whom surgery was the only remaining option, the outcome can be considered as satisfying for the majority of patients. The long-term results, as assessed by the Tsui torticollis scale, amounted to 45.3% improvement and were as good as, or even better than, what has been shown in studies of patients treated by botulinum injections, in whom improvements were reported to range from 19% to 40%.<sup>27 28</sup> However, one important drawback with our study is that the evaluations were not blinded.

In many old series of SPD the outcome had been assessed in terms of excellent, marked, moderate, mild and no improvement.<sup>17 21</sup> Other studies have utilised scales such as TWSTRS<sup>29</sup> or the modified TWSTRS that includes functional disability and pain.<sup>30–33</sup> We have used the Tsui torticollis scale, which was, at the start of our series, the most frequently used scale in studies on botulinum toxin. The Tsui scale has shown a high degree of congruence with the TWSTRS.<sup>34</sup> The mean reduction on the Tsui score in our patients is comparable with the reduction on the TWSTRS severity score in previous studies, which has been reported to range from 22% to 59%.<sup>29 30 35 36</sup>

Recently, Huh *et al*<sup>37</sup> published a report comparing SPD to pallidal DBS in cervical dystonia, showing no major difference in the results. However, we believe that the surgical treatment should be tailored according to the individual patient, and that the patient groups suitable for either of the two surgical options may differ.

#### **Quality of life**

A major goal in all functional neurosurgery is to improve the QoL for patients. The modified TWSTRS includes one section assessing the functional outcome in daily tasks, and studies reporting on this issue indicate a significant improvement in many of the practical tasks of daily life.<sup>30 36</sup> An improvement in functional outcome may not necessarily imply a better life quality as assessed by patients, and no previous study has specifically addressed this issue. In our study, the patients rated their life quality in general as significantly better postoperatively. Although the outcome according to each specific domain of the QoL scale was not significantly changed at long-term follow-up, the total score encompassing all different domains of life quality was significantly improved. The Fugl-Meyer QoL scale, being a generic scale, may not be ideal for this group of patients, and may fail to capture specific aspects relevant to patients with cervical dystonia.

## **Complications and side effects**

In earlier studies, postoperative dysphagia has been reported to occur in 4-33% of patients.<sup>31 32</sup> In two of our three patients who postoperatively had dysphagia, we believe the reason for it may have been due to a dramatic change in head position: both patients had a marked retrocollis preoperatively while postoperatively their heads were slightly anteflexed, resulting in changes in the anatomical conditions for swallowing. Another explanation could be a delayed initiation of swallowing, a

symptom recognised by Munchau *et al.*<sup>36</sup> The third patient had dysphagia as well as dysphonia. This patient had a retrocollis and underwent a bilateral dorsal ramisectomy. In this patient we believe that strain to the upper cervical nerve roots during surgery may have resulted in oedema of the medulla. Fortunately, all three patients recovered with time.

After sectioning of the posterior branch of C2 all patients developed a sensory loss within the corresponding area. One possible way to avoid this side-effect is to perform intradural posterior rhizotomy according to the description by Taira and Hori.<sup>8</sup> However, since in our experience the sensory loss very seldom raises any complaints from patients, we have chosen to remain extradural and thus less invasive.

#### Recurrences and changes in pattern of dystonia

Reinnervation is known to occur after SPD, possibly caused by incomplete denervation of small nerve branches that may facilitate reinnervation. Munchau et al performed a detailed evaluation of their series and reported a high incidence of 25% for reinnervated SCM muscle and 20% for the splenii. They believed that this problem was related to incomplete denervation of small branches from the cervical plexus, which were overlooked at surgery.<sup>38</sup> We have been aware of this problem and carefully looked for such branches by electrical stimulation. In order to find those branches, the SCM muscle has to be sectioned and its medial aspect dissected down to its most caudal part. In all patients where SCM was denervated we also found one or several small branches entering the muscle medially in its lower part. However, in two out of three patients with reinnervation of the SCM, we could nevertheless find a clear reinnervation through one or several pathological thick branches originating from the accessory nerve, indicating that reinnervation does not always depend on incomplete primary denervation.

The splenius seems to be the muscle where most recurrences appear. Munchau et al had reinnervation of the splenii in 20% of patients, a figure close to what we and others have found. This raises the question of the anatomy of the innervation. According to anatomical textbooks the splenius is innervated by dorsal branches from C1 and C2, together with dorsal rami from C3-C7. However, at surgery we have sometimes found nerve branches innervating the splenius from the level of C2-C4, and having a more lateral course than the dorsal rami. We suggest that these may arise from the upper cervical plexus and hence may often be overlooked at surgery. Thus, a double innervation similar to what has been described for the trapezius may be present.<sup>39</sup> This could explain the difference in recurrence rate when comparing it with the semispinal muscle, where we had only six cases (9%) with minor reinnervation. In 12 patients the reinnervation of splenii was severe and further surgery was indicated. Nine of those 12 patients also showed an altered pattern in their dystonia. Six patients with reinnervation of splenii were reoperated with repeated denervation and six underwent pallidal DBS due to a simultaneous change in the pattern of their dystonia.

At follow-up, 31 patients showed a change in their pattern of pathological muscle activity with a subsequent change in resting head position, compared to the preoperative position. Although most patients only had minor discomfort out of this, 13 (21%) had major problems, leading to repeated denervation in five patients (8%) and pallidal stimulation in seven (11%). Similar changes after SPD have also been reported previously.<sup>32</sup> Indeed, our incidence is high, but on the other hand we have been liberal with performing a postoperative EMG at follow-up, recognising even minor changes. In addition, most of our

# **Movement disorders**

patients had mild or moderate symptoms. It is difficult to appreciate whether this change in muscular activation pattern is caused by a progress of the disease itself or by a postoperatively induced modulation of the central circuits responsible for the dystonic state.<sup>40</sup> A change in pattern has also been reported after successful long-term treatment with botulinum injections, which may suggest common underlying pathophysiological mechanisms in the two treatment situations.<sup>41</sup> Since both treatments can be regarded as peripheral denervation by chemical or by surgical means, it is logical that the central response to those treatments can appear rather similar. Additionally, similar changes in dystonic patterns have been reported following longterm pallidal DBS for generalised dystonia, sometimes necessitating a new operation for insertion of additional DBS electro-des in the globi pallidii.<sup>42 43</sup> However, the eight patients that we reoperated with pallidal DBS due to a more generalised cervical activity did in most cases show a satisfactory outcome, as reported in a previous paper.<sup>2</sup>

# CONCLUSIONS

Our results show that SPD may still have a role in treatment of cervical dystonia. A significant number of patients showed satisfactory long-term results. Although no specific domain in ratings of life quality was significantly improved, the patients rated their life quality in general as improved and reported an alleviation of their pain at long-term follow-up. It is noteworthy that our patients all had a severe disease without any relief from botulinum injections or other conservative measures. With this in mind, and considering the documented safety of this procedure, the results of SPD can be considered as satisfactory.

The high incidence of reinnervation of the splenii muscles suggests that special attention to the innervation of this muscle is warranted at surgery.

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#### REFERENCES

- Claypool DW, Duane DD, Ilstrup DM, et al. Epidemiology and outcome of cervical dystonia (spasmodic torticollis) in Rochester, Minnesota. *Mov Disord* 1995:10:608–14.
- 2 Ferreira JJ, Costa J, Coelho M, et al. The management of cervical dystonia. Expert Opin Pharmacother 2007;8:129–40.
- Brashear A. The botulinum toxins in the treatment of cervical dystonia. Semin Neurol 2001;21:85–90.
- 4 Moore AP, Naumann M. Handbook of botulinum toxin treatment. Oxford, UK: Blackwell Science Ltd, 2003.
- 5 Berman B, Seeberger L, Kumar R. Long-term safety, efficacy, dosing, and development of resistance with botulinum toxin type B in cervical dystonia. *Mov Disord* 2005;20:233–7.

- 6 Ruiz PJ, Castrillo JC, Burguera JA, et al. Evolution of dose and response to botulinum toxin A in cervical dystonia: a multicenter study. J Neurol 2011;258:1055–7.
- 7 Bertrand C, Molina Negro P, Martinez SN. Technical aspects of selective peripheral denervation for spasmodic torticollis. *Appl Neurophysiol* 1982;45:326–30.
- 8 Taira T, Hori T. Peripheral neurotomy for torticollis: a new approach. Stereotact Funct Neurosurg 2001;77:40–3.
- 9 Laitinen LV, Bergenheim AT, Hariz MI. Leksell's posteroventral pallidotomy in the treatment of Parkinson's disease. *J Neurosurg* 1992;76:53–61.
- 10 Cacciola F, Farah JO, Eldridge PR, et al. Bilateral deep brain stimulation for cervical dystonia: long-term outcome in a series of 10 patients. *Neurosurgery* 2010;67:957–63.
- 11 Islekel S, Zileli M, Zileli B. Unilateral pallidal stimulation in cervical dystonia. Stereotact Funct Neurosurg 1999;72:248–52.
- 12 Hung SW, Hamani C, Lozano AM, et al. Long-term outcome of bilateral pallidal deep brain stimulation for primary cervical dystonia. *Neurology* 2007;68:457–9.
- 13 Skogseid IM, Ramm-Pettersen J, Volkmann J, et al. Good long-term efficacy of pallidal stimulation in cervical dystonia: a prospective, observer-blinded study. Eur J Neurol 2012;19:610–15.
- 14 Ostrem JL, Racine CA, Glass GA, et al. Subthalamic nucleus deep brain stimulation in primary cervical dystonia. *Neurology* 2011;76:870–8.
- 15 Sarubbo S, Latini F, Quatrale R, et al. Five-year follow-up of 10 patients treated with globus pallidus internus deep brain stimulation for segmental or multisegmental dystonia. Stereotact Funct Neurosurg 2012;90:84–91.
- 16 Loher TJ, Capelle HH, Kaelin-Lang A, et al. Deep brain stimulation for dystonia: outcome at long-term follow-up. J Neurol 2008;255:881–4.
- 17 Cohen-Gadol AA, Ahlskog JE, Matsumoto JY, et al. Selective peripheral denervation for the treatment of intractable spasmodic torticollis: experience with 168 patients at the Mayo Clinic. J Neurosurg 2003;98:1247–54.
- 18 Tsui JK, Eisen A, Stoessl AJ, et al. Double-blind study of botulinum toxin in spasmodic torticollis. Lancet 1986;2:245–7.
- 19 Consky ES, Lang AE. Clinical assessments of patients with cervical dystonia. In Jankovic J, Hallett M, eds. New York: Marcel Dekker, 1994:211–37.
- 20 Jankovic J, Hallett M, eds. Therapy with botulinum toxin. New York: Dekker, 1994:211–37.
- 21 Bertrand CM. Selective peripheral denervation for spasmodic torticollis: surgical technique, results, and observations in 260 cases. Surg Neurol 1993;40:96–103.
- 22 Anderson WS, Lawson HC, Belzberg AJ, et al. Selective denervation of the levator scapulae muscle: an amendment to the Bertrand procedure for the treatment of spasmodic torticollis. J Neurosurg 2008;108:757–63.
- 23 Taira T, Mitsuyama T, Okami N, et al. [Selective peripheral denervation for spasmodic torticollis involving the levator scapulae muscle]. No Shinkei Geka 1999;27:25–31.
- 24 Fugl-Meyer AR, Melin R, Fugl-Meyer KS. Life satisfaction in 18- to 64-year-old Swedes: in relation to gender, age, partner and immigrant status. J Rehabil Med 2002;34:239–46.
- 25 Capelle HH, Blahak C, Schrader C, et al. Bilateral deep brain stimulation for cervical dystonia in patients with previous peripheral surgery. *Mov Disord* 2012; 27:301–4.
- 26 Zrinzo L, Foltynie T, Limousin P, et al. Reducing hemorrhagic complications in functional neurosurgery: a large case series and systematic literature review. J Neurosurg 2012;116:84–94.
- 27 Nijmeijer SW, Koelman JH, Standaar TS, *et al*. Cervical dystonia: improved treatment response to botulinum toxin after referral to a tertiary centre and the use of polymyography. *Parkinsonism Relat Disord* 2013;19:533–8.
- 28 Poewe W, Deuschl G, Nebe A, et al. What is the optimal dose of botulinum toxin A in the treatment of cervical dystonia? Results of a double blind, placebo controlled, dose ranging study using Dysport. German Dystonia Study Group. J Neurol Neurosurg Psychiatry 1998;64:13–17.
- 29 Meyer CH. Outcome of selective peripheral denervation for cervical dystonia. Stereotact Funct Neurosurg 2001;77:44–7.
- 30 Krauss JK, Toups EG, Jankovic J, et al. Symptomatic and functional outcome of surgical treatment of cervical dystonia. J Neurol Neurosurg Psychiatry 1997;63:642–8.
- 31 Braun V, Richter HP. Selective peripheral denervation for spasmodic torticollis: 13-year experience with 155 patients. J Neurosurg 2002;97(2 Suppl):207–12.
- 32 Munchau A, Palmer JD, Dressler D, et al. Prospective study of selective peripheral denervation for botulinum-toxin resistant patients with cervical dystonia. Brain 2001;124(Pt 4):769–83.
- 33 Ford B, Louis ED, Greene P, et al. Outcome of selective ramisectomy for botulinum toxin resistant torticollis. J Neurol Neurosurg Psychiatry 1998;65:472–8.
- 34 Tarsy D. Comparison of clinical rating scales in treatment of cervical dystonia with botulinum toxin. *Mov Disord* 1997;12:100–2.
- 35 Ford B, Louis ED, Greene P, *et al*. Outcome of selective ramisectomy for botulinum toxin resistant torticollis. *J Neurol Neurosurg Psychiatr* 1998;65:472–8.
- 36 Munchau A, Good CD, McGowan S, et al. Prospective study of swallowing function in patients with cervical dystonia undergoing selective peripheral denervation. J Neurol Neurosurg Psychiatry 2001;71:67–72.

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- 37 Huh R, Han IB, Chung M, et al. Comparison of treatment results between selective peripheral denervation and deep brain stimulation in patients with cervical dystonia. *Stereotact Funct Neurosurg* 2010;88:234–8.
- 38 MacKenzie J. The morphology of the sternomastoid and trapezius muscles. J Anat 1955;89:526–31.
- Tubbs RS, Shoja MM, Loukas M, et al. Study of the cervical plexus innervation of the trapezius muscle. J Neurosurg Spine 2011;14:626–9.
- 40 Munchau A, Filipovic SR, Oester-Barkey A, et al. Spontaneously changing muscular activation pattern in patients with cervical dystonia. Mov Disord 2001;16:1091–7.
- 41 Maia FM, Kanashiro AK, Chien HF, *et al.* Clinical changes of cervical dystonia pattern in long-term botulinum toxin treated patients. *Parkinsonism Relat Disord* 2010;16:8–11.
- 42 Cif L, Vasques X, Gonzalez V, et al. Long-term follow-up of DYT1 dystonia patients treated by deep brain stimulation: an open-label study. *Mov Disord* 2010;25:289–99.
- 43 Cif L, Gonzalez-Martinez V, Vasques X, *et al.* Staged implantation of multiple electrodes in the internal globus pallidus in the treatment of primary generalized dystonia. *J Neurosurg* 2012;116:1144–52.