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Case Report

Modified McKenzie-Dandy operation for a cervical dystonia patient who failed selective peripheral denervation: A case report and literature review

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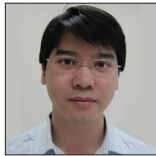
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ABSTRACT

Background: Cervical dystonia (CD) is a rare and difficult-to-treat disorder. Various neurosurgical options are available, each with its own set of advantages and disadvantages. We investigated using the modified McKenzie-Dandy operation for a patient with CD who failed selective peripheral denervation (SPD).

Case Description: A 42-year-old man presented left-sided rotational torticollis for 3 years. He was referred for surgery after treating with a variety of oral medications and repeated botulinum toxin injections that became ineffective. For the first operation, the patient underwent SPD (modified Bertrand's operation); unfortunately, the postoperative outcome was unsatisfactory, and the operation was considered a failure. After his symptoms did not improve after 6 months, the modified McKenzie-Dandy operation was performed. Immediately following surgery, he experienced satisfactory outcomes. He was able to resume his normal activities and employment after 1 month after recovering from his temporary swallowing difficulties. He only complained of minor neck pain and no recurrence was observed after 3 years follow-up.

Conclusion: For patients who have failed SPD, a modified McKenzie-Dandy procedure is a feasible and effective option. The procedure is relatively safe when performed properly, and the long-term effects can be maintained.

Keywords: Cervical dystonia, Denervation, Rhizotomy, Spasmodic torticollis

INTRODUCTION

Cervical dystonia (CD) is an uncommon movement disorder characterized by involuntary intermittent twisting or sustained contractions of many cervical muscle groups. The various forms of disfiguring head-and-neck postures that are caused by imbalanced action of several paired cervical muscles include rotational torticollis (also known as spasmodic torticollis; ST), laterocollis, anterocollis, retrocollis, or their various combinations.[7] The patient may experience pain, discomfort, and limitations in functional or occupational activities, and the deformity may result in low self-esteem, embarrassment, depression, or social isolation.[14]

Among the various surgical options, most neurosurgeons prefer extraspinal selective peripheral denervation (SPD), widely known as Bertrand's operation, or its modifications.[6,8,10,12] Even though it has been proven to be highly effective, a significant number of patients are unable to attain satisfactory results.[20]

The authors have described a case in which SPD failed to provide significant symptomatic relief, necessitating an intradural rhizotomy procedure (modified McKenzie-Dandy operation)[14,15,22,27] as a last resort. We also examined the literature to determine what other options are available to those who failed SPD.

CASE DESCRIPTION

Case history and physical examination

A 42-year-old man presented to our department after 3 years after receiving a diagnosis of the left-sided rotational torticollis. He began to exhibit symptoms 2 months after a motorcycle accident in which he temporarily lost consciousness and experienced minor neck pain. Initially, he exhibited a paroxysmal involuntarily head rotation toward the left without elevation of the shoulder. No other symptoms were evident such as blepharospasm, oromandibular dyskinesia, or tremor. The symptom was exacerbated by anxiety or mental stress. Later, the head rotation became more tonic, resulting in a constant fixed rotational position except when sleeping. He had to touch his chin to keep his head in the midline. No history of torticollis or dystonia was reported among his family members. Treatments with a variety of oral medications as well as repeated botulinum toxin injections were no longer effective. His symptoms deteriorated and became more uncomfortable, limiting his daily activities and ability to work. He was then referred to our department for surgical consultation.

The neurological examination results were normal at the time of admission, except for palpable abnormal sustained contractions of the right sternocleidomastoid (SCM) and left posterior neck muscles (splenius capitis and semi-spinalis capitis). A sensory tick (*geste antagonistique)* was noticed when he touched his chin. The Toronto Western ST Rating Scale (TWSTRS) gave a score of 24 on the torticollis severity scale, 23 on the disability scale, and 11 on the pain scale (total score = 58). An electromyography (EMG) was not used to record aberrant muscle activation. His magnetic resonance imaging (MRI) brain revealed no abnormalities, but his MRI spine indicated cervical spondylosis with mild narrowing of the left neural foramen at C3 to C4 and C6 to C7 levels.

Following a thorough discussion of the risks and benefits of surgical treatment, the patient opted to undergo the operation in February 2018. We used the modified Bertrand's procedure to denervate the left posterior neck muscles, in which we strictly followed the technique described by Taira *et al.*[29] In brief, the anterior rootlets of the left C1 and C2 spinal nerves were sectioned intradurally through a C1 hemilaminectomy, whereas the posterior branches of the left C3 to C6 spinal nerves were sectioned extradurally. A separate incision was made along the posterior border of

the right SCM muscle to section all branches of the spinal accessory nerve (SAN) supplying the muscle. During the procedure, we used monopolar electrical stimulation to identify each nerve branch and then confirmed total denervation using a strong voltage stimulation (5 Hz, 1 ms pulse width, 2 volts).

Unfortunately, after the operation, the patient had no meaningful relief, with a TWSTRS score of 22, 23, and 9 on the torticollis severity scale, disability scale, and pain scale, respectively (total score = 54). After 6 months of intensive exercise and physiotherapy, the symptoms appeared to be unchanged. A second stage operation with modified McKenzie-Dandy operation was considered and performed in September 2018.

Surgical procedure

The patient was placed in the prone position. We used spinal somatosensory evoked potential and motor evoked potential for neuromonitoring. An EMG was used to monitor the SCM and trapezius muscle activities. A left-sided Bertrand's hockey stick incision was made. The trapezius, splenius capitis, and semi-spinalis muscles were dissected and sectioned from their insertion just below the nuchal line of the occipital bone. Subperiosteal dissection was then performed bilaterally. At the C1 and C2 spinous processes, the left inferior oblique capitis and rectus capitis major and minor were detached and severed from their origin. A laminectomy of C1 to C3 was carried out and the posterior rim of the foramen magnum was removed. The dura was then opened to expose the lower brainstem and upper cervical spinal cord. Using the microsurgical technique, an arachnoid dissection was performed. After cutting the denticulate ligaments, we used bipolar electrode stimulation (0.1–0.3 volts) to detect the anterior cervical nerve rootlets. The first cervical motor root was identified as it runs transversely just below the proximal intradural part of the vertebral artery. Some of the left anterior C1 and C2 rootlets were already cut by the previous operation. The left anterior C1 to C4 rhizotomies were carried out using bipolar and microscissors. The right anterior C1 to C3 rootlets, on the other hand, were sectioned less aggressively to minimize the risk of swallowing difficulty and bilateral phrenic nerve impairment. Any tiny blood vessels were carefully preserved. The spinal root of the right SAN was tracked upward until where it crosses the vertebral artery. Neither abnormal vascular loop nor the connection between the SAN and the C1 root (McKenzie branch) was found. Three to four spinal rootlets of the SAN that produced a strong contraction of the SCM muscle on electrical stimulation were sectioned, while the upper medullary rootlets were preserved to minimize the risk to the pharynx or vocal
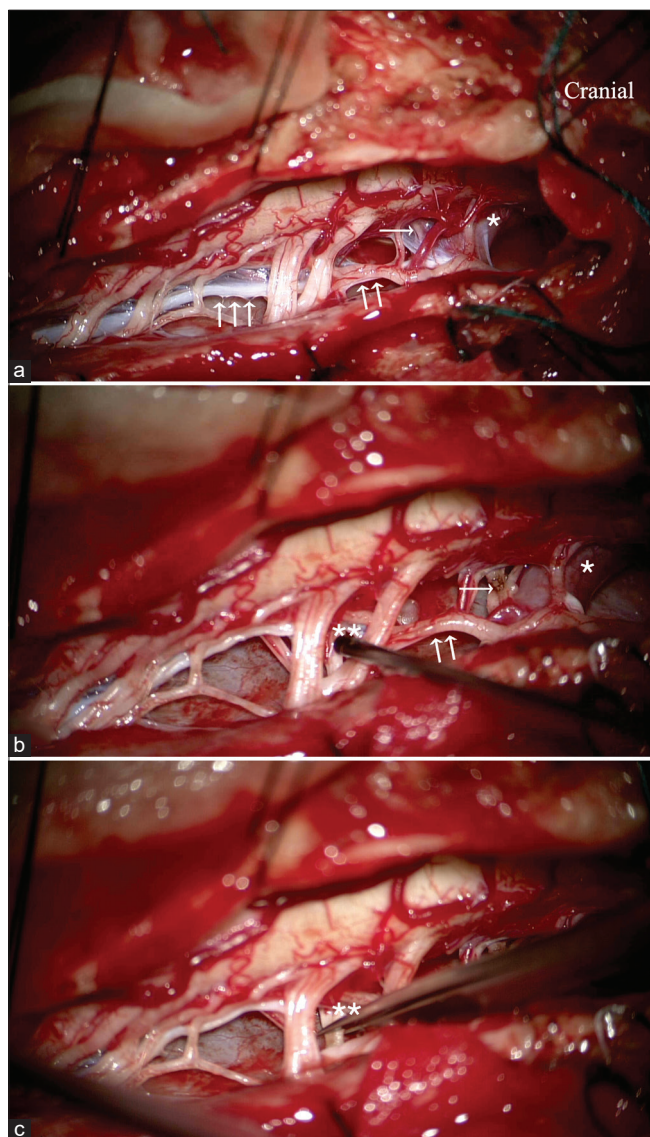
Figure 1: Intraoperative photograph showing the anatomy after opening the dura (a and b). The right-sided spinal accessory nerve; SAN (double arrow) runs parallel to the spinal cord. The denticulate ligaments (triple arrow) were carefully dissected. The anterior root of C-1 (single arrow) is often hidden just beneath the vertebral artery (asterisk). The McKenzie nerve was not identified in this case. The C1-C3 and SAN motor rootlets (double asterisk) were sectioned selectively, using electrical stimulation (c).

cord impairment [Figure 1]. The dura was closed. Then down to C6, dissection was performed between the plane of the semi-spinalis capitis and semi-spinalis cervicis. The insertion of the semi-spinalis capitis to the vertebral body was severed, and extensive coagulation was performed in the area lateral to the facet joints. A strong voltage electrical stimulation (2–5 volts) was used to confirm total denervation. A suction drain was then placed, and the incision was closed.

Postoperative course

After the operation, the patient's head and neck returned to normal posture. He stated that he had a marked improvement. He complained of a mild degree of swallow difficulty. He did not have choking or dysphagia. The symptoms were completely resolved 1 month after surgery. The TWSTRS at 3 months gave a score of 3 on the torticollis severity scale, 4 on the disability scale, and 2 on the pain Scale (total score = 9). He could return to his previous job and social activities. He experienced no numbness or dysesthesia in the occipital area. At 3 years follow-up, he was free of symptoms, and the effect of treatment was still sustained [Figure 2]. A follow-up flexion-extension radiograph of the cervical spine revealed a mild degree of segmental kyphosis at the C3 to C4 level, but no signs of spinal instability [Figure 3].

DISCUSSION

Nonoperative treatments for CD, including oral medications, botulinum toxin injections, and physiotherapies, have been used as first-line treatments, albeit they typically only provide minor and temporary symptom relief. In addition, a significant number of patients failed to respond or became refractory to botulinum toxin after repeated injections, due to developing anti-botulinum toxin antibodies. In such cases, a neurosurgical procedure should be considered.

Bertrand *et al.* described an extraspinal SPD in the early 1980s, and it has since become the most preferred surgical treatment for CD among neurosurgeons.[12] The aims of the procedure for rotational torticollis, as in this case, are to denervate the ipsilateral posterior cervical muscles by extradural section of the C1 and C2 roots, as well as an extradural section of the posterior primary rami of C3 to C6 (posterior ramisectomy), and section of all peripheral branches of the SAN that innervates the contralateral SCM muscle.[4,5,23,24] Taira *et al.* described a modification of Bertrand's procedure by sectioning the ventral C1 and C2 roots intradurally through a C1 hemilaminectomy to reduce numbness or dysesthesia in the area of the great occipital nerve and avoid troublesome bleeding from venous plexuses, as well as the danger of vertebral artery injury.[28,29] According to several case series of patients with CD treated with SPD (Bertrand's or its modifications), 70–90% of patients can receive total or marked relief.[3,5,6,8,10,13,28,30,34] In contrast, a considerable number of patients (12–27%) did not obtain a satisfactory result following surgery, whether immediate or delayed [Table 1]. In recent studies, a TWSTRS severity score improvement of <25% after SPD is defined as an unsatisfactory result.[20] Inappropriate patient selection, incorrect identification, or inadequate denervation of involved muscles, as well as reinnervation of the denervated muscles, are all possible causes of failed or ineffective SPD.

Figure 2: (a) The patient's posture at the time of presentation. (b) When he touched his chin, sensory trick was observed. (c) After the patient underwent the selective peripheral denervation operation, his symptoms persisted. (d) After reoperation with the modified McKenzie-Dandy operation, a favorable result was obtained.
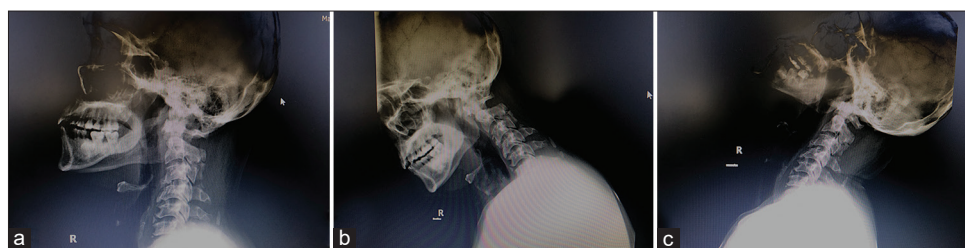

Figure 3: Cervical spine X-ray. (a) Lateral, (b) flexion, and (c) extension.

Table 1: The rate of failed SPD (Bertrand's operation) among surgical case series.				
Surgical series	Number of patients (n)	Percentage of satisfactory result (n)	Percentage of unsatisfactory result (n)	Percentage of recurrence or required second operation
Bertrand et al.,1993[14]	260	88 (230)	12 (30)	2 (5)
Chen et al., 2000[8]	207	87.9 (182)	12.1 (25)	NA
Braun et al., 2002[6]	140	73 (102)	27 (38)	11 (15)
Cohen-Gadol et al., 2003[10]	162	77 (125)	23 (37)	10 (17)
SPD: Selective peripheral denervation				

Various steps in SPD can be mistaken or overlooked. Thus, reoperation using the same surgical approach or modifications can be considered as the first option. However, reoperation has several disadvantages, including the difficulty of searching for small residual nerve branches in fibrotic or scar tissue, as well as, the loss of the original anatomical plane, which makes dissection difficult, both of which lead to reoperation failure. Furthermore, an extensive dissection can also cause unintentional damage to the arteries or nerves that supply other muscles in the neck or shoulder, particularly the branches to the trapezius.

McKenzie originally performed intradural sectioning of the anterior and posterior roots of the upper cervical spine, as well as partial sectioning of the SAN, in treating patients with CD.[22] Dandy improved the technique in 1930 by performing a bilateral upper three anterior rhizotomies with SAN denervation on the periphery to preserve the trapezius function. Because the surgical techniques and the validated rating scale were not yet standardized, the efficacy and safety of the procedure varied among subsequent studies.[15,27] Friedman et al. again refined the technique, using electrical stimulation to selectively section the intradural upper three cervical anterior roots and the rootlets of the SAN, mediating SCM muscle contraction. Among the 58 consecutive patients, he reported a significant improvement result and restoration of the head posture in 85% and 59% of the patients, respectively, although 45% experienced a mild transient swallowing difficulty that mostly resolved in a few months.[14] Krauss et al. later proposed a two-stage procedure, with the first stage being bilateral rhizotomy or ramisectomy and the second stage being peripheral denervation or section of the SCM muscle. However, in his series, excellent or marked outcomes were achieved in only 48% of cases.[19] The McKenzie-Dandy operation has been abandoned following a report associating it with an increased risk of cerebrospinal fluid leakage, swallowing difficulties (dysphagia), vascular injuries, phrenic or trapezius paralysis, and neck instability.[6-8,12,27]

In this case, we modified the McKenzie-Dandy operation by performing an asymmetrical upper anterior rhizotomy (C1 to C3 ± C4) and selective sectioning of the SAN's rootlets, intradurally. The aim was to interrupt all motor supplies to the muscles that controlled head rotation while minimizing the risk of swallowing difficulties and trapezius paralysis. We used Bertrand's hockey stick incision to increase surgical exposure and created a myotomy effect by sectioning and dissecting the trapezius and splenius capitis from their insertion directly below the occipital bone.[10] We also severed the semi-spinalis capitis insertion to expose the area of the lateral branches of the posterior rami of C3 to C6 and performed extensive coagulation to eliminate any residual nerve branches in that location. This strategy offers the advantage of avoiding the difficulties associated with hunting for small residual nerve branches in fibrotic or scar tissue, as well as the risk of inadvertent injury to the nerve to the trapezius. The authors believe that using high-magnification microscopy combined with meticulous microsurgical techniques and intraoperative electrical stimulation, earlier documented complications can be reduced. Furthermore, it necessitates less technical demand than Bertrand's reoperation. Recently, similar to this present study, Aljuboori et al. published a clear and concise review with a video illustration of the modified McKenzie procedure to treat fixed painful torticollis.[1]

Several issues in this operation should be considered. The caudal limit of intradural anterior rhizotomy involves unilateral C4 to prevent the risk of bilateral phrenic nerve paralysis and the motor roots that supply the brachial plexus. The superior rootlets, or cranial root, of intradural SAN should always be preserved because they form a part of the inferior vagal nerve rootlets that are responsible for pharyngeal and vocal cord function.[31] Because it carries motor fibers from the anterior root of the C1 to the intradural SAN, the McKenzie nerve, which can be obscured by the denticulate ligament or vertebral artery and has been observed in 3.4–50% of patients, should always be sought and severed if it exists to provide complete denervation of the SCM muscle. The McKenzie nerve can be confirmed by the strong contraction of the SCM muscle when it is electrically stimulated.[7,14,25,26,32,33] Finally, full functional recovery of the patient necessitates extensive exercise and rehabilitation.

There have been reports of other surgical procedures for patients who have failed SPD. Globus pallidus deep brain stimulation (GPi-DBS) has become more commonly used in recent years, and some surgeons now advocate it as a first-line treatment for CD rather than denervation surgery, especially in complex forms of CD.[2,9,11,18] Our patient could not afford the expense of implantation, which is one of the most limiting factors to consider. Unilateral or bilateral pallidotomy has also been reported as rescue therapy for CD after SPD failure.[16,17,20,21] The obvious advantages of this procedure over GPi-DBS include its much lower cost, lack of hardware-related problems, and suitability for young patients. Horisawa et al. reported a case in which patients, who had failed SPD, underwent simultaneous bilateral pallidotomy with successful outcomes and no permanent adverse effects. According to his report, the Tsui score and TWSTRS severity score improved by 88.2% and 91.7%, respectively, after surgery.[17] In the authors' opinion, this procedure seems promising and appropriate for this patient. However, we did not have a radiofrequency generator to perform this procedure at the time we treated the patient.

CONCLUSION

The modified McKenzie-Dandy surgery was a feasible and effective alternative for patients who have failed SPD or Bertrand's operation. When performed properly, it remains relatively safe, and its long-term effects can be maintained.

Declaration of patient consent

Institutional Review Board permission obtained for the study.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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