

Surgical Therapy for Pure Trigeminal Motor Neuropathy Accompanied by Limited Mouth Opening: A Retrospective Study

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Abstract

Purpose: This study aims to find out if there is any new manifestation of pure trigeminal motor neuropathy and evaluated the long-term results of surgery for its treatment.

Patients and methods: The present study includes 10 patients referred from December 2003 to June 2014. The clinical manifestation investigations were recorded and 3 patients with limited mouth-opening were operated. The long-term follow-up result was performed.

Results: Among 10 patients, 6 suffered from limited mouth opening, the other 4 jaw deviation. Among patients who had difficulty in mouth-opening, 3 patients chose surgery to improve the situation, the others just follow-up. At the final follow-up, the average maximum mouth opening of operated patients totally increased by 180%. For non-operated patients, no obvious improvement of mouth opening was observed.

Conclusion: Limitation on mouth-opening is another manifestation of pure trigeminal motor neuropathy and surgery can effectively improve the situation.

Keywords: Pure trigeminal motor neuropathy; Limited mouth opening; Surgical therapy

Introduction

Pure trigeminal motor neuropathy was first described by China in 1988 [1]. It is a kind of trigeminal motor paralysis disease, which is unaccompanied by trigeminal sensory or any other cranial nerve disturbances. Generally, patients complain about pain and masticatory disability. The morbidity presents low rate. According to literature, less than 20 cases of pure trigeminal motor neuropathy have been reported [1-9]. The primary cause of the pain is not very clear. Autoimmune-inflammatory reactions, infectious, and genetic causes had been considered [5,10], but none of them confirmed. Patients also suffer from jaw deviation [1,5,7,9,11]. After Computed Tomography (CT) or Magnetic Resonance Imaging (MRI) examination, atrophy of the masticatory muscles and smaller ramus in the lesion side could be found. At the same time, electromyography can confirm paresis of muscles. Also, there is no effective treatment for this disease. Except those who had clear secondary pathogenesis, such as tumor, cerebral infarction etc., no one had been completely cured [3,4,6,8,12].

However, according to our study which is based on some cases, severe limitation of mouth opening seems to be the most observed symptom. Aiming at this new symptom, surgery was used. In this study, different clinical manifestations and surgical therapy results are also discussed.

Materials and Methods

Patients

The present series included 10 patients referred from December 2003 to June 2014 with pure trigeminal motor neuropathy (6 women and 4 men; age range of 8-60 years; mean age, 39.3 years). Patients presented to our hospital with the complaints of weakness in chewing, limitation of mouth opening, progressive asymmetry of the face, and so on. The ethical approval was given by Shanghai Jiao Tong University's Ethics Committee. We have read the Helsinki Declaration and have strictly followed the guidelines in this investigation.

Diagnosis standard

The diagnosis standard of pure trigeminal motor neuropathy included three aspects:

Clinical examination: It showed severe atrophy of the masticatory muscles on the lesion side, jaw deviation, or/and limitation of mouth opening. Sensation to light touch in the distributions of bilateral trigeminal nerve, swallowing function, voice, and tongue motions were normal (Figures 1A and 1B) [1-9].

Radiographic investigation: Ramus of the lesion side mandible was vertically and horizontally smaller than the other side. Lesion side trigeminal motor nerve-innervated muscles showed severe atrophy and fat replacement. No space-occupying, artery occlusion lesions or degenerative changes in the brain or brainstem (including the pons) (Figures 2A, 2B and 2C) [3-9].

Electromyography examination: Needle electromyography of the masseter and temporalis muscle showed sparse fibrillations and positive sharp waves and scanty motor units with a fast firing rate, while the volitional activity was markedly reduced. The masseter reflex test revealed loss of waveform from the lesion side (Figure 3) [1,2,7,9].

Surgical technique

3 patients with limited mouth opening chose surgical therapy to improve jaw-opening symptom. All of these patients were operated on

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Figure 1A: Muscle atrophy of patient. Atrophic masseter (arrowhead) and temporalis (arrow) are shown.



Figure 1B: Severe limitation of mouth-opening.



Figure 2A: Panoramic radiography shows that the ramus of the left mandible is vertically and horizontally smaller than the ramus of the right mandible.

under general anesthesia and by the same senior maxillofacial surgeon. Bilateral mucosal incision was made over the external oblique line, and the anterolateral portion of the masseter muscle was exposed. Masseter muscle attachment was loosened from the ramus of the mandible and coronoid process was also resected. To reduce the incidence of reattachment of muscle, we put part of buccal pad between mandible and masseter muscle. Part of masseter muscle was resected for histopathology examination. 1 week after surgery, mouth opening and closing training was initiated, which included passive mouth opening training.

Histopathological examinations

Resected muscle was embedded in paraffin, stained with hematoxylin and eosin, and examined microscopically.

Results

Among 10 patients, 6 (60%) suffered from limited mouth opening, the other 4 (40%) jaw deviation, all patients (100%) complained weakness in chewing and muscle atrophy. Only one patient had head trauma history (Patient 1), and one had viral infection (Patient 2) before they get ill. The possible cause is still unclear (Table 1).

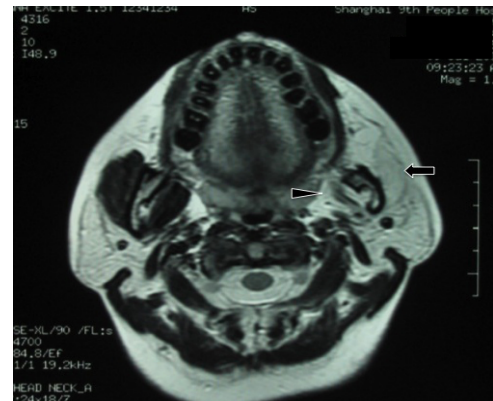


Figure 2B: T1-weighted facial MRI showing atrophy and fat instead of the left masseter (arrow) and medial pterygoid muscle (arrowhead).



Figure 2C: CT showing atrophy of the left masseter (arrow) and lateral pterygoid muscle (arrowhead).



Figure 3: Needle electromyography of right masseter showing sparse fibrillation and positive sharp wave.

Among the 6 patients who had difficulty in mouth-opening, 3 patients chose surgery to improve the situation, the other just follow-up, all patients accepted mouth opening and closing training. Maximum mouth opening of patients (including operative and non-operative) was measured, and showed in Table 2.

One patient was lost for follow-up. The mean follow-up duration of limited mouth-opening patients was 5.35 years (in arrange of 1 year 3 months to 10 years 3 months, except the lost patient). At final follow-up, among 3 operated patients, the average increase of maximum mouth opening was 21 mm, and compared to preoperative average mouth opening (11.7 mm), it improved by 180%. Patient 2 is an 8 year old boy who presented mouth-opening limitation when he was born. After the surgery, his mouth opening was excellent with 28 mm maximum mouth opening. For the other 3 patients who chose to follow up, no obvious improvement of mouth opening was observed and no patient showed a maximum mouth opening more than 25 mm. Two non-operative patients both complained still severe face atrophy and no improvement of mouth opening after training.

Histopathological examination of resected muscle showed degenerative changes of muscle fiber, like hyalinization and fibroplasia. Hyperplasia of fat was existed in the part of muscle region (Figure 4).

Discussion

Pure trigeminal motor neuropathy is characterized by mandibular branch motor weakness without any signs of trigeminal sensory or

other cranial nerve involvement. The etiology of this disease remains obscure. Suggested causes include neurofibromatosis, viral infection, multiple sclerosis, trauma, or unknown factors [2-4,6,8,11]. However, histologic evidence of lesion and therapies has not been reported yet.

The clinical manifestation varies with the chronicity of the process, like facial asymmetry, pain, weakness on chewing. Opening deviation is one of the important symptoms of pure trigeminal motor neuropathy. According to medical literature reports [3-9] and this study, we found that patients' jaw always deviated to the lesion side on mouth opening, and could not move to the normal side against resistance. However, in this study, except deviation, there was another important symptom, limited mouth opening which was not mentioned in literatures. Among 6 patients suffered from limited mouth opening, 3 of them had bilateral masticatory muscle atrophy. The reason of deviation and limitation on opening is not very clear. We suppose that denervation muscles become fibrillation and lose their elasticity [10], which leads to the inter-synechia of lateral pterygoid muscles, masseter muscles and temporalis muscles. Degeneration lateral pterygoid muscles cause to deviation and degeneration masseter and temporalis muscles cause to limitation. Histopathology examination which showed hyalinization and fibroplasia demonstrated our opinion partly.

Normally, according to literature review [1,5,7,9], because long-standing trigeminal motor denervation is irreversible, no effective therapy is available for patients with pure trigeminal motor neuropathy. For those patients who had difficulty in mouth-opening, it influenced

S.No	Sex/Age (Years)	Clinical Examination			Imaging Examination		EMG	Surgery	Follow-up
		Masticatory Muscle Weakness and Atrophy	Opening Deviation (L/R)*	Opening Limitation	X-Ray (Smaller Ramus)	CT/MRI (Head/Face)			
1	F/46	+	-	+	+	normal/ left atrophy	Spontaneous activities, decreased number of motor unit	+	10y 3m
2	M/8	+	-	+	+	normal/ bilateral atrophy	Spontaneous activities, decreased number of motor unit	+	7y 8m
3	F/60	+	-	+	+	normal/ right atrophy	Spontaneous activities, chronic neurogenic MUP	+	2y 1m
4	F/36	+	-	+	+	Normal/ left atrophy	Spontaneous activities, chronic neurogenic MUP	-	5y 6m
5	M/38	+	-	+	+	normal/ bilateral atrophy	Spontaneous activities, chronic neurogenic MUP	-	7y 8m
6	F/42	+	-	+	+	normal/ bilateral atrophy	Spontaneous activities, scanty or no MUP	-	lost
7	F/49	+	+(L)	-	+	normal/ left atrophy	Spontaneous activities, chronic neurogenic MUP	-	4y 1m
8	F/43	+	+(R)	sometimes	+	normal/ right atrophy	Spontaneous activities, chronic neurogenic MUP	-	3y 8m
9	M/17	+	+(L)	-	+	normal/ left atrophy	Spontaneous activities, chronic neurogenic MUP	-	1y 7m
10	M/54	+	+(R)	-	+	normal/ left atrophy	Spontaneous activities, decreased number of motor unit	-	4y 1m

*Deviation direction, L = left, R = means right; MUP = motor unit potential.

Table 1: The general characteristics of the patients.

Patient Follow-up	Mouth opening(mm)			Duration of Follow-up
	Preoperative	During operation	Postoperative	
1 (operative)	10	42	30	10y 3m
2 (operative)	15	45	30	7y 8m
3 (operative)	10	42	33	2y 1m
4 (non-operative)	15	-	18	5y 6m
5 (non-operative)	23	-	20	1y 3m
6 (non-operative)	20	-	-	lost

Table 2: Maximum mouth opening of patients.

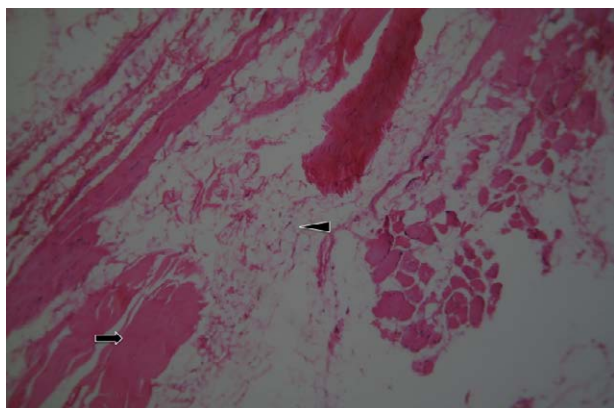


Figure 4: Histopathology examination of resected masseter. Fibrosis (arrow) and fat replacement (arrowhead) is showed.

their daily life severely and nutrition uptake. So we tried to solve this problem by surgery. The possible reason of difficulty in mouth-opening might be muscle fibrillation, we chose loosening the masseter and coronoid resection surgery to improve the situation. Loosening the muscle can reduce the stretch from masseter to mandible, on the other hand, after resection of coronoid, antagonism from temporalis and lateral pterygoid muscle has also decreased when opening mouth. No matter during or post operation, maximum mouth opening obviously increased compared to before, and after years of follow-up, operative patients still keep a better mouth-opening situation in contrast of non-operative patients. The long-term results of the surgery were mainly satisfactory, although facial atrophy not had any improvement.

Conclusion

With the results of the study we can conclude that the limitation on mouth-opening is another manifestation of pure trigeminal motor neuropathy which can be improved by surgery (masticatory muscle loosen and coronoid resection).

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Dr. Chai Ying and Dr. Wenbin Wei contributed equally to the manuscript.

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