Short report

Jaw closing spasm—a form of focal dystonia? An electrophysiological study

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SUMMARY The case of a 36 year old man suffering from unilateral right jaw closing spasms over two years is reported. Permanent spasm with trismus severely impeding mouth-opening was combined with paroxysms triggered by various sensory stimuli. The diagnosis of temporo-mandibular joint syndrome was considered but treatment failed to improve the symptoms. Neurological investigation two years after onset of the spasms showed by electrophysiological studies excessive co-contraction of the antagonistic jaw-closers, mainly the right masseter during attempts at jaw opening and absence of the silent period in the right masseter and anterior temporalis following jaw tap and trigeminal exteroceptive stimulation. Jaw dystonia was therefore considered and Botulinum A toxin was injected into the right masseter and temporalis which dramatically improved the patient's condition.

Jaw spasms are involuntary contractions of masticatory muscles. Paroxysmal spasms last from a few seconds to a few minutes bringing on a sudden closure, opening, or deviation of the jaw. Permanent spasms usually result in a jaw closure with trismus. Electrophysiological studies may be useful in an attempt of understanding the mechanisms of the spasm.

We report the case of a patient suffering from unilateral jaw closing spasms over two years. Permanent spasm with trismus and paroxysms were associated. Injections of Botulinum A toxin in the right masseter and right temporalis substantially improved the patient's condition.

Case report

First symptoms appeared in March 1984. This patient, a 36 year old man, complained of difficulty opening his mouth wide. He had a sensation of stiffness in the right mandibular region with tenderness during and just after eating. Three months before the beginning of his trouble, he had a motorcycle accident, but without evident head injury. An orthopantomogram and tomography of the temporomandibular joints ruled out gross abnormality of teeth, maxilla, or

joints. A temporomandibular joint syndrome secondary to dental malocclusion was considered, and a maxillary acrylic occlusal splint was placed. This procedure did not relieve the symptoms. During the next six months his condition worsened. He experienced more and more difficulty in opening his mouth. Tiredness, emotion, cold or prolonged talking reinforced the trismus. Brief paroxysms with abrupt jaw closure, occasionally causing tongue biting, were triggered by yawning, sudden startle or knocking against something. He was given different drugs, mainly sedative, but without result. Only diazepam (30 mg per day) was slightly effective, reducing the frequency of paroxysms. Partial detachment of the fibres of the right lateral pterygoid from the zygomatic arch was performed to reduce a possible muscular dysfunction (January 1985). Under general anaesthesia, trismus was relieved with complete passive jaw opening, proving that there was no limitation of condylar movements. But a few days after the intervention, the pre-operative status returned. Magnetic resonance imaging (MRI) of the brain (April 1985) showed no abnormality. He was referred to us in February 1986. He had a severe trismus. The widest interincisor distance was 1.5 cm. Right masseter and right temporalis muscles were firm and hypertrophic compared with those on the left. There was no sensory loss in the right trigeminal area. Neurological examination was normal. We noticed that cold reinforced the trismus. Routine biological investigations, including those for blood and urine levels of calcium phosphate, magnesium and serum creatine kinase, were normal. Titration of antibodies to tetanus toxin was considered protective. A second MRI examination 9 months after the first, showed no abnormalities in the brainstem and confirmed the hypertrophy of the right masseter. Diazepam (10 mg i.v.) did not improve the

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Received 31 May 1988 and in revised form 12 December 1988.
Accepted 19 December 1988

Fig Before injection of botulinum A toxin in the right masseter. (a) Strong EMG activity in the right masseter (RM) on attempt of maximal jaw opening. The left (LM) remains silent. (Horizontal bar 500 ms—vertical bar 1 mV.) (b) Silent period recorded from the right masseter (RM) and the left (LM) during maximal clenching: silent periods absent in the right masseter following (from top to bottom) jaw tap, right mental nerve stimulation, left mental nerve stimulation. (Horizontal bars 20 ms.) (c) After injection of botulinum A toxin into the right masseter: Abrupt discharge of EMG activity in the right anterior temporalis (RAT) on yawning, no noticeable activity in the right masseter (RM). LAT: left anterior temporalis, RD: right digastric, LD: left digastric. (Horizontal bar 1 s, vertical bar 500 μ V).

symptoms. A transient improvement was obtained by injection of xylocaine 1% (5 ml) in the right masseter, the widest interincisor distance temporarily reaching over 3 cm.

Electrophysiological study

Coaxial needle recording showed activity of normal MUPs at rest in the right masseter. Their firing was below 10 Hz and irregular. Progressive opening of the mouth increased their discharge-frequency and new potentials of larger amplitude were recruited. At maximal opening of the mouth, limited to 1.5 cm, a strong EMG activity of full interference was recorded in the right masseter while the left masseter remained silent (fig a). In the right internal pterygoid, EMG activity was present at rest, increasing during opening, however clearly less marked than in the right masseter. At

maximal teeth clenching, normal interferential EMG patterns were recorded in jaw-closers (right internal pterygoid, right and left masseters). In the right lateral pterygoid (jaw opener), approached through the semi lunar notch of the mandible, a pattern of reduced interference was recorded at maximal jaw opening, without signs of neurogenic or myogenic involvement. In this muscle there was no activity at rest and slight activity during maximal clenching. Latencies of the jaw-reflex recorded from masseters were normal. Silent period (SP) following jaw tap, and SPs following electrical stimulation of the right or left mental nerves (exteroceptive suppression¹), were absent in the right masseter and the right anterior temporalis, whereas these SPs were present in the left masseter and the left anterior temporalis (fig b). No abnormalities were found in the parameters of the H reflex in the right masseter studied as

described by Godeaux and Desmedt.² A SP of 20 ms was found in the right masseter on supramaximal stimulation of the right masseteric nerve. Blink reflex, direct responses of the orbicularis oculi following stimulation of the facial nerves and trigeminal SEPs were normal.

After electrophysiological tests, diagnosis of jaw dystonia was considered. Diluted clostridium botulinum toxin type A (3 ng of neurotoxin) was injected at four points in the right masseter. One month later, maximal interincisor distance reached 3 cm. Electrophysiological tests were once more performed. The strong EMG activity, observed in the right masseter during mouth opening before infiltration, was no longer present. However, in this muscle SPs following jaw-reflex and stimulation of the mental nerves remained abolished. This demonstrated that the injection improved jaw function, but did not modify the physiopathological mechanisms of the trouble. Opening was then apparently slightly limited by the right anterior temporalis shown by EMG activity. In this muscle, during yawning a discharge of EMG activity was recorded (fig c) without paroxysmal spasm. Diluted clostridium botulinum toxin (2 ng of neurotoxin) was injected in the right anterior temporalis. Three months later, the patient considered he had recovered normal jaw function.

Discussion

Two main points emerge from our electrophysiological study. The first was the limitation of mouth opening because of an excessive co-contraction of the antagonistic jaw-closers predominating in the right masseter. The second was the abolition of the SPs following jaw jerk and exteroceptive stimulations, in the masseter and anterior temporalis on the right. Another electrophysiological finding of interest is that stimulation of the right masseteric nerve was followed by a SP of 20 ms in the right masseter. If autogenic inhibition by Ib fibres plays a part in this silent period, it would not play a part in the SP following jaw-jerk which was absent in this muscle.

The first diagnosis considered for our patient was a temporomandibular joint syndrome. The aetiology of this syndrome is obscure and probably multifactorial.³ Muscle dysfunction is most likely an important factor, usually connected with malocclusion. Prolonged SP following jaw jerk has been reported.⁴ Correction of malocclusion improves the symptoms and decreases the duration of the SP. In our patient absence of SP following jaw-jerk and the failure of treatment for malocclusion were unlike the results usually reported in this syndrome. Nothing in our patient supported the hypothesis of a peripheral lesion of the trigeminal nerve, inducing unilateral paroxysmal jaw closing spasms.⁵⁻⁶ A strictly unilateral masticatory spasm has been exceptionally reported in a case of cephalic tetanus,⁷ but an isolated masticatory spasm over two years is inconsistent with this diagnosis. Absence of other symptoms since the beginning of the troubles, and normal MRI examinations at 9 months interval,

excluded trismus due to a structural lesion in the pons.⁸ It is also unlikely that isolated jaw-closing spasm, worsening progressively over two years, would be the first manifestation of multiple sclerosis. Moreover demyelination tends to increase the SP following mixed nerve stimulation.⁹

Excessive co-contraction with loss of reciprocal inhibition between agonist and antagonist during voluntary activity and posture is a fundamental component of dystonia.¹⁰ Exteroceptive suppression in the masseters following mental nerve stimulation was absent in nearly half of the cases of blepharospasm and oromandibular dystonia in the study of Berardelli *et al.*¹¹ Focal dystonia after trauma have been reported. In our patient symptoms appeared three months after trauma. Pharmacological modifications induced by traumas might be responsible for dystonia.¹²⁻¹³ The trigeminal motor nucleus, where receptors to many pharmacological agents have been identified,¹⁴ could be particularly sensitive to such modifications. Thus in our patient a form of focal dystonia, perhaps secondary to trauma, is possible. However, the two cases of jaw dystonia reported by Thomson *et al.*¹⁵ were different from our case. They consisted of jaw deviation, the lateral pterygoid was the dystonic muscle and signs of dystonia were present elsewhere. In our patient no signs of dystonia were present elsewhere.

The physiopathology of dystonia remains unclear. One hypothesis is the reduction in the inhibition normally exerted on the agonists by the Ia inhibitory interneurons from the antagonists.¹⁰ The mechanism is necessarily different for jaw-closers because jaw-openers which contain no or few spindle afferents, are unable to originate an inhibition from Ia fibres.¹

Finally we believe that some cases of the so-called temporomandibular joint syndrome are in fact jaw dystonia. They could be improved by local injection of botulinum A toxin as reported for other focal dystonia.¹⁶ Electrophysiological studies are able to define the abnormalities in jaw muscles, allowing a better understanding of individual cases and therefore a better adjustment of the treatment.

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