Management of unilateral idiopathic masseter muscle hypertrophy with botulinum toxin type A

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SUMMARY

Masseter hypertrophy (MH) is an uncommon disorder which can cause both aesthetic and functional problems. The most common aetiological factors associated with MH are habit of chewing gum, clenching and/or bruxism. The treatment of MH includes conservative management as well as surgical resection of the enlarged muscle and/or bone. Injection of botulinum toxin type A is a relatively new and minimally invasive method for management of masseter muscle hypertrophy, which offers many advantages over conventional surgical management. This paper reports a case of unilateral MH of unknown origin which was treated with injection of botulinum toxin type A, resulting in satisfactory reduction in the volume of muscle and improvement of facial aesthetics.

BACKGROUND

Masseter hypertrophy (MH) is an uncommon disorder, characterised by benign enlargement of the masseter muscle volume. It has the highest incidence in the second and third decades of life, with no sex predilection.[1] MH may result in aesthetic and functional disturbances. It causes prominence of muscle, leading to square or rectangular facial form. Functional problems associated with MH are due to the altered tone of the hypertrophied muscle, which may lead to protrusion of jaw, pain and headache.[2] The aetiology of MH is obscure and is most commonly related to the grinding habits of jaw that includes excessive gum chewing, clenching and bruxism. When no aetiological factor is identified, MH is considered to be idiopathic in nature.[3] Idiopathic masseter muscle hypertrophy (IMMH) was first described by Legg.[4] People suffering from psychological disorders or emotional disturbances that impact the proprioception and ability to maintain the tone of the masseter muscle are at a higher risk of developing IMMH.[2] MH can involve either one or both of the masseter muscles; however, bilateral MH is more common; accounting for 60% of the cases.[5 6] The present paper describes a case of idiopathic unilateral masseter muscle hypertrophy which was successfully treated non-surgically by injecting botulinum toxin type A.

CASE PRESENTATION

A 22-year-old female patient reported to the oral and maxillofacial surgery clinic with the primary symptom of painless swelling over left lower third of the face, since past 2 years. The primary symptom of the patient was due to the aesthetic concerns and there was no functional deficit. The swelling was gradual in onset and was not associated with pain or fever. The patient reported no deleterious or parafunctional oral habit. Also, there was no history of trauma or psychological distress. On extraoral examination, the swelling was localised to the left angle region of the mandible and was non-tender on palpation (figure 1). When the patient was asked to clench her teeth the bulk of the masseter muscle was visible and the swelling was firm in consistency. On intraoral examination, there was no sign of odontogenic infection or occlusal prematurity. Mouth opening was adequate. No growth was seen in the left posterior region of oral cavity.

INVESTIGATIONS

Panoramic radiograph was done to rule out any bony lesion in the angle and ramus region of the mandible. The orthopantomogram revealed mild broadening of the mandible in the angle region, bilaterally (figure 2). Patient was subjected to ultrasonography (USG), which did not reveal any soft tissue pathology involving the parotid gland or the masseter muscle. Beside USG, advanced imaging tools namely MRI and CT with contrast could be used to rule out soft tissue pathology causing the masseter swelling. However, in the present case they were not done due to financial constraint. Based on the clinical examination and radiological findings, diagnosis of unilateral IMMH of left side was made.

TREATMENT

Options for management of MH included, surgical excision of part of the hypertrophied left masseter muscle with or without osseous recontouring or conservative management with intramuscular botulinum toxin injection. After discussing the treatment options with patient and her family members, conservative management with botulinum toxin type A injection was chosen, to avoid the risks and complications of surgery. The botulinum toxin used was purified *Clostridium botulinum* toxin type A, dispensed as freeze-dried powder containing 100 units per vial. It was reconstituted with 2 mL of sterile saline, forming a solution with concentration of 50 units of botulinum toxin type A of per mL solution (5 units per 0.1 mL).

Patient was prepared for local injections of botulinum toxin in the left hypertrophied masseter muscle. A local subdermal skin test was done to rule of any allergy to botulinum toxin. The skin over the lower face was prepared with betadine solution. The injection protocol and dose used for the present case was based on the guidelines reported by Kim *et al* and Ayhan *et al*.[7 8] Skin

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Figure 1 Extraoral photographs showing swelling over the left angle of mandible region on (A) frontal view, (B) right and (C) left profile view.

markings for injection were drawn with surgical marking pen. A line was drawn from the corner of the mouth to the tragus of ear. Below this line, anterior and posterior limits of masseter muscle were marked till the inferior border of mandible. A second marking was made with its periphery 1 cm inside the previous markings. The boundary of this margin denoted the area of injection of botulinum toxin. The rationale was to keep the injection area 1 cm within the boundaries of masseter muscle, to ensure that the solution was injected into the bulk of masseter muscle and not too anteriorly near the corner of mouth or too superiorly near the zygomatic bone. Patient was then asked to firmly clench her teeth, making the masseter muscle prominent. A central point was marked where maximum thickness of the muscle was palpated. From this point, four points in all the directions were marked at a distance of 1 cm (figure 3). The reconstituted solution of botulinum toxin was injected at the centre and the four peripheral areas using 1 mL syringe and 30G needle. Total of 1.5 mL of the solution was deposited at these five points, accounting for 75 units of botulinum toxin.

OUTCOME AND FOLLOW-UP

The patient had mild pain at the site of injection on the first postoperative day, which subsided thereafter. On extraoral examination there was decrease in the bulk of the masseter muscle at first month follow-up. At 4 months recall, the MH was significantly resolved on visual inspection by the operating maxillofacial surgeon and the patient herself (figure 4). Standardised and quantitative assessment of the reduction in MH could be measured using CT or MRI study. However, the patient was satisfied with her improved appearance and was not willing for further intervention. Patient's satisfaction was rated using a 5-grade scale by Lee *et al*, with scores ranging from 0 (completely dissatisfied) to 4 (completely satisfied).[9] The patient gave a score of 4 at the end of 4 months follow-up.

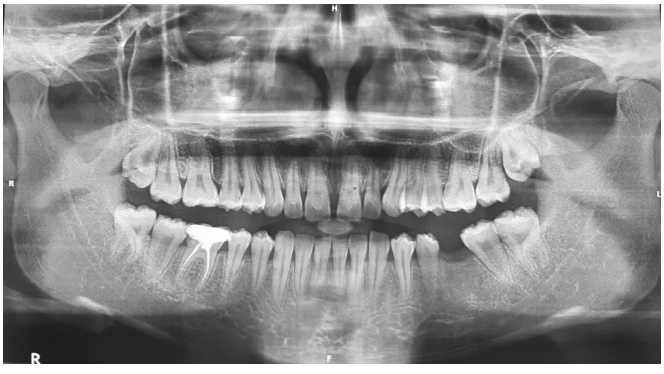

Figure 2 Orthopantomogram showing mild broadening of the mandible in the angle region.

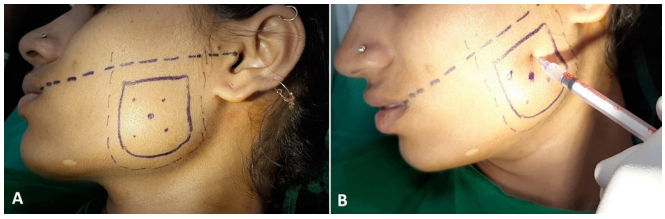

Figure 3 Intraoperative photographs showing (A) skin markings and (B) injection at the marked site.

DISCUSSION

MH is a rare disorder characterised by benign enlargement of the masseter muscle, unilaterally or bilaterally. Clinical findings of MH include asymmetric swelling at the angle region of mandible which is usually painless and firm on palpation when the patient clenches his/her teeth. Clinically, MH can be mistaken for other pathologies and hence should be accurately evaluated by clinical examination and radiographic findings.[2] Differential diagnosis of MH include submasseteric space infection, soft-tissue or intrabony lesion at the angle and ramus region of mandible, and salivary gland tumours related to the parotid gland.[10] Trismus and signs of odontogenic infection (ie, fever, tenderness, pus discharge) should be ruled out to eliminate submasseteric space infection. Orthopantomogram should be taken to rule out intrabony pathologies. USG assists in excluding any soft tissue or salivary gland tumours. CT and posteroanterior skull view radiograph reveals irregularities in the bone mediolaterally which manifests as 'bony flaring', 'bone spur' or 'hyperostosis' at the insertion of masseter muscle at the inferior border at angle region and lateral surface of ramus of mandible.[11] In the present case, clinical examination did not show any signs of odontogenic infection involving the submasseteric space (ie, trismus, oral pus discharge, tooth pain, carious tooth, localised raise in temperature over the angle of mandible with tenderness and fever). No soft tissue swelling or growth was appreciated on intraoral assessment. Use of USG further ruled out any soft tissue or parotid tumour, which can present as swelling over angle of mandible. The orthopantomogram showed mild broadening over the angle of mandible, which is possibly due to the hypertrophied masseter muscle, supplementing the diagnosis of MH.

The treatment for MH is either conservative, surgical or a combination of both, depending on the severity.[12] Conservative treatment includes counselling of patients with psychological disorders and use of mouth-guards in patients having habit of clenching or bruxism.[2 13] Antispasmotic, anxiolytic drugs and analgesics are used to relieve the symptoms of MH. Surgical management reportedly used for MH includes removal of the vertical band of the internal half of masseter muscle belly, from below zygomatic arch up to its insertion at the inferior border and lateral surface of ramus of mandible.[14] Depending on the severity of MH, upto two-third thickness of the muscle bulk can be resected, either through intraoral or extraoral approach.

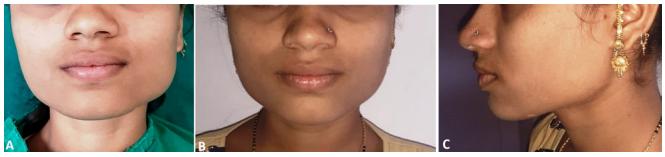

Figure 4 Photographs showing (A) preoperative extraoral view and (B) postoperative extraoral view at 4 months follow-up. Left profile view (C) showing reduction in the bulk of the masseter muscle.

Extraoral approach was first proposed by Gurney in 1947, where only the lateral part of the muscle was removed.[5] Extraoral approach has a risk of injury to the marginal mandibular branch of the facial nerve and the lateral part of muscle removal includes risk of damage to the parotid gland. Extraoral approach also results in scar from incision placed in submandibular region of the neck. However, when reduction osteoplasty is to be performed in cases of bony hyperplasia, extraoral approach provides a better access. Mandibular angle osteotomy along with the removal of the medial part of the masseter muscle was reported by Adams.[15] Converse reported an intraoral approach for muscle volume and bone removal, thereby eliminating risk of nerve damage and scar.[16] The surgical treatment; however, involves complications like facial nerve injury, masseteric nerve or inferior alveolar nerve damage, trauma to masseteric artery. Postoperative complications include pain, swelling, trismus, haematoma formation and presence of scar. Asymmetric resection in bilateral cases giving rise to more unesthetic appearance, which may further leads to patient dissatisfaction.

Injection of botulinum toxin type A is relatively new and conservative treatment modalities for MH.[13] Botulinum toxin is a lethal toxin produced by anaerobe *C. botulinum* which is responsible for producing disease of botulism. There are seven types of botulinum toxins out of which only type A and B are commercially available for pharmacological uses. Botulinum toxin type A is a potent neurotoxin available as type-A haemagglutinin complex. It binds permanently to the motor end-plate at neuromuscular junction and blocks the release of acetylcholine from presynaptic vesicles. The reuptake of acetylcholine from the neuromuscular cleft is blocked and the storage vesicles of acetylcholine are used-up. This mechanism is responsible for the delayed onset of action of toxin which is usually after 2–4 days of administration.[1] Local administration of botulinum toxin in small doses in a muscle produces paralysis of the muscle which in turn weakens the muscle leading to its subsequent atrophy. Though the toxin binds permanently to the motor end-plate, presynaptic axonal sprouting occurs which leads to the formation of new neuromuscular junctions and the function of the muscle is returned. This explains why the effect of botulinum toxin is not permanent and this process by which the muscle function returns was first demonstrated by Duchen.[17]

The first clinical application of botulinum toxin was for the treatment of strabismus in 1980.[18] Smyth first reported good clinical results with the use of Botox-A to treat MH.[1] The merits of injecting botulinum toxin over surgical management include avoidance of exposure to GA, intraoperative and postoperative complications and it is a relatively less invasive technique without the requirement of local anaesthesia. However, in cases of severe hypertrophy with hyperostosis, surgical resection remains the treatment of choice. The dosage of the botulinum toxin can be decided based on the clinical experience depending on the individual case. The toxin is dispensed in freeze-dried powdered form in a vial as 100 units. This is diluted in 1 or 2 mL of sterile saline and the reconstituted solution should be used within 4 hours of its formation. Kim *et al* evaluated the effects of botulinum toxin on 11 patients with bilateral MH using computed tomographic measurements. 100 units was diluted in 1 mL saline and about 30 units of the toxin was injected on each side. They stated that the injection of toxin induced muscular atrophy up to 22% varying from 8.1% to 35.4%.[7] But the bulk of muscle was regained to some extent within 6 months. The peak effect of the toxin that is responsible to cause muscular paralysis is seen at 1–2 weeks. However, the peak changes in lower facial contour are obtained at 12 weeks. This is because the muscular atrophy is secondary to muscular paralysis. To *et al* studied the quantitative effect of botulinum toxin for treatment of MH on five patients and reported a median reduction of 30.9% of masseter bulk.[19] The mean masseteric muscle thickness was measured by USG which is however, limited due to possible inter-measurement variations. Ayhan *et al* reported a case of unilateral MH of right side which was treated by botulinum toxin.[8] Hundred units of the toxin was diluted in 2 mL saline and about 75 units were injected at first visit. As the patient was not satisfied with the results, another 60 units were injected at second visit 1 month later.[8] The longevity of the effect of the toxin depends on the number of injections. Repeated injections at 12 weeks interval in divided dosage increases the longevity of muscular atrophy. Also, the muscular paralysis brings about cessation of the clenching and grinding habits, as it is not possible to clench the teeth firmly due to functional denervation of jaw closing muscles.[1] The bony changes in the mandible are secondary to the muscular hypertrophy in the region of insertion of muscle. It is presumed that these bony abnormalities will remodel back to normal once the hypertrophy is corrected by the use of toxin. In the present case botulinum toxin was injected for a single sitting. A total dose of 75 units divided over five sites over the master muscle yielded a satisfactory result. The effect on the muscle bulk reduction was stable at 9 months follow-up.

There are no permanent complications reported with the use of botulinum toxin. Few transient complications include change in the facial smiling expression, sunken cheek, facial muscle paralysis, pain at the site of injection or pain during mastication.[7 8] Alteration in the facial smiling expression is due to the injecting the toxin far too anteriorly near the corner of mouth into zygomaticus major muscle which is responsible for elevation of the corner of mouth. Sunken cheek appearance is caused as a result of injecting the toxin far too superiorly below the zygomatic arch or zygomatic buttress which leads to atrophy of the masseter muscle near its origin. Facial paralysis may be caused due to injecting the toxin into the parotid gland close to the branches of the facial nerve. Injection into the parotid gland may also lead to reduced salivary flow. However, these complications recover with time once the effects of the toxin are reversed. In the present case transient mild pain was reported at site of injections which was not significant. No permanent complications were seen with the treatment.

Learning points

- Botulinum toxin injection is a conservative method for the treatment of masseter hypertrophy, which is minimally invasive and can offer many advantages over conventional surgical treatment.
- The present report highlights a case of unilateral left masseter muscle hypertrophy of unknown origin. The disorder presented with purely aesthetic disturbance due to unilateral swelling over the left angle of mandible. No functional disturbance was present.
- The masseter muscle hypertrophy was successfully treated non-surgically by injection of botulinum toxin type A. Seventy-five units of botulinum toxin injected over five sites of the affected masseter muscle in a single sitting provided satisfactory reduction in muscle bulk, restoring the facial symmetry and appearance in the young female patient.

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