Case Report

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MANDIBULAR ANGLE AND CORONOID PROCESS FRACTURE SECONDARY TO OROFACIAL DYSTONIA: REPORT OF A CASE

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ABSTRACT

As the angle is a weak region in the continuity of mandible, so it is more prone to fracture. It has been proven time and again that coronoid fracture results from a strong sudden contraction of temporalis. Muscular forces influence the remodeling of bones. Orofacial dystonia is a centrally mediated disease in which there is an uncontrolled spasmodic contraction of facial and masticatory muscles. This continuous force applied over a long period of time has the potential to unfavorably remodel or weaken bone. A case is presented in which the dystonic action of facial musculature gradually resorbed the bone to such an extent that there was spontaneous fracture at the right angle of mandible as well as the contra lateral coronoid. Management of this fracture posed a challenge at every step eventually leading to resection of the ramus-condyle unit. No case has been reported so far in the literature where dystonic movements have resulted in fracture of the mandible.

KEY WORDS: Orofacial dystonia; Angle fracture; Reconstruction plate; Osteomyelitis.

INTRODUCTION

Orofacial dystonia is a neuromuscular disorder of central origin which causes involuntary, inharmonious, heavy, intermittent or continuous spasmodic and painful contraction of facial, lingual and masticatory musculature (Tolosa E, Marti MJ. 1988). The etiology of dystonia is varied. According to a study by Tan and Jancovic (1999), most of the cases are idiopathic (63%). Other causes outlined are drug-induced (22.8%), peripheral-induced (9.3%), post anoxic (2.5%), neurodegenerative disorder associated (1.8%) and head injury associated (0.8%). Triggering factors include emotional or physical stress, depression and orofacial surgical procedures/trauma. There is no specific outline for diagnosis and treatment of dystonia. Clinical symptoms and patient's presentation govern the choice of treatment to be rendered. The options reported in literature are medical management, chemodenervation and CNS procedures. In this case, we came

across dystonia that had been chronic but intermittent, increasing in duration and intensity with every episode. It gradually led to yielding of the hard tissue not only in form of bone in mandibular angle region but also in loosening and extraction of the mandibular molars.

CASE REPORT

A 50 year old female reported with a complaint of swelling and severe pain over the right mandibular angle region for the past ten days. The patient had been diagnosed with right angle fracture by an attending dental surgeon and referred to the center for management. On examination, the swelling was diffusely distributed over the right angle and was soft and extremely tender on palpation. The skin color and temperature was normal. Occlusion was disturbed and there was a step palpable at right angle. Mouth opening was decreased with an active opening of 15 mm between centrals.

Orthopantomogram (OPG) revealed fracture of the right angle and left coronoid process. Oral hygiene was poor with severe halitosis.

FIGURE 1: Initial presentation of the patient.

The patient appeared debilitated and depressed and was having continuous painful contractions of muscles of right half of the face. Ipsilateral muscles of mastication were also active. While collecting the case history information, it was revealed that these spasms had started 2 years ago with subtle intensity and intermittent occurrence. Neurological abnormality was detected and the patient had been kept on antidepressants and antiparkinsonian drugs which patient discontinued due to excessive dizziness. The episodes became stronger and long-lasting over the past 2 years. An episode of severe contraction had occurred about 10 days prior to the initial appointment. This severe contraction was followed by excruciating pain at the right angle region. This forced the patient to visit a dental surgeon who clinically and radiographically diagnosed right angle fracture. Since the patient belonged to a sub-urban city, she was referred to the center in Delhi. However, the family took about 8 days to report to the center. During this period, pain had increased

remarkably and the sleep was disturbed. Ingestion of liquids had become progressively difficult leading to dehydration. OPG revealed a fracture line each at right angle and left coronoid process of the mandible.

FIGURE 2: Orthopantomogram (OPG) showing right angle and left coronoid fracture (Pre-op).

An immediate decision was made to stabilize the fracture segments by intermaxillary fixation (IMF) and consult a neurologist again for the facial spasms. The neurologists ordered a few investigations including routine blood investigations, serum electrolytes, liver and kidney function tests and magnetic resonance imaging (MRI) of brain to rule out any central lesion. The patient was kept on baclofen and pacitane. The very next day patient reported to us with increased pain and inability to sleep. The patient had taken injectable diclofenac sodium from local physician at night resulting in mild pain relief. On examination, the tie wires used for intermaxillary fixation had loosened and the arch bar stability had also been compromised which was attributed to an episode of muscle spasm. Arch bars were reinforced and once again intermaxillary fixation was done in occlusion. The patient was asked to follow the neurologist's instructions. After a couple of days she reported to the neurologist with severe pain and loosened IMF. The investigation results were within normal range. No abnormality was detected on MRI. The patient was again advised to consult the maxillofacial surgeon for rigid fixation of the fracture segments as the

neurologists speculated that the intermaxillary fixation was aggravating the condition. The situation was explained to the patient that the muscular forces were beyond normal limits and the miniplate fixation would not be adequate. It was decided that the fixation with a reconstruction plate would be more appropriate. The patient was advised to continue with the medication and was on close observation. Within a few days her condition deteriorated. Carbamezipine was added to her medications with a suspicion of neuralgia. However, there was no significant relief.

The patient was placed under general anesthesia for open reduction and fixation of the right angle fracture with locking reconstruction plate (Synthes, 2.4 mm) via submandibular approach. The procedure was carried out safely. Intraoperatively, a lot of granulation tissue and enlarged submandibular lymph nodes were found at the fracture site. Nodes were excised and unhealthy granulation tissue removed. The mandibular molars were extracted as they had become periodontally compromised to an extent of grade III mobility. The surrounding mucosa was inflamed and fragile intraorally.

Primary closure could not be obtained. The wound was partially closed and iodoform gauze was packed intraorally. Extraoral site was closed in layers. Subsequently, on the third post-operative day, she developed lesions of herpes zoster. Local and systemic acyclovir was administered and steroids stopped. As the inflammation gradually subsided, the intraoral wound was primarily closed on the seventh post-operative day. She was discharged on ninth post-operative day and instructed to continue with the medications prescribed by the neurologist.

The patient continued weekly follow-up and reported marked relief in terms of pain but dystonic episodes persisted.

FIGURE 3: Herpes zoster.

During the first month, spasms decreased in intensity and duration. After about 2 months, she again reported with a swelling in right angle region that was mildly tender. The dystonic movements had amplified again but were intermittent. OPG was advised and revealed multiple fractures of the angle and ramus region with formation of sequestrum.

FIGURE 4: OPG showing multiple fractures and sequestrum formation.

The patient was again placed under general anesthesia for resection of the fractured segment. Upon exposure, it was noticed that the lytic process had involved whole of the proximal segment extending up to the condyle. Fractured segments and sequestrum were resected and sent for histopathology.

Microscopic examination confirmed osteomyelitis. The patient reported significant relief from spasmodic episodes within 15 days of surgery.

FIGURE 5: OPG after segmental mandibulectomy.

On 6 months follow-up, she was found to be symptom free and is still continuing with drugs as per neurologist's recommendation. During this period she had been kept on a changing combination of antiepileptics and benzodiazepines (Carbamezipine, gabapentin, valproic acid, chlordiazepoxide, clonazepam, pacitane), and was currently taking valproic acid, escitalopram and clonazepam.

FIGURE 6: Final follow-up photograph

DISCUSSION

Meige was the first person to describe oro-facial dystonia in 1910 (Tolosa ES 1981). This disease has also been termed Meige's syndrome when it occurs in conjunction with blepharospasm. This condition is not completely understood and has variable presentations. The disease can affect a single, some, or all the muscles of one or both sides of face. This leads to variations in presentation of the disease viz. painful jaw opening or closing, repetitive dislocations and jaw deflection (Jankovic, 1988; Tolosa, 1981; Tolosa & Marti, 1988). All kinds of jaw movements including protrusion, retrusion, lateral movements, either alone or in combination may be affected. These mandibular movements may be repetitive or sustained. When the muscles of facial expressions and lingual musculature are affected by the spasmodic contractions, there is combination of facial twitching, abnormal lip movements, nasal contractions, clenching/grinding of teeth, retractions of oral commissures and/or abnormal movements of tongue (Jankovic, 1988; Jankovic & Ford, 1983). Platysma may also be involved resulting in contractions of neck. Involvement of laryngeal muscles has also been reported in the form of breathing problems, dysphagia, dysphonia, and dysarthria. Gradually, these uncontrolled spasms lead to accumulation of products of anaerobic metabolism in muscles and thus myalgia precipitates. The pain further splints the antagonist muscles. On the other hand, spasms continue leading to further damage to soft tissue envelope. The condition worsens if the treatment is not rendered and the facial pain becomes intolerable. Triggers or stimulating factors like stress, depression or any kind of activity involving the oro-facial musculature can start the spasmodic episodes (Tolosa, 1981; Jankovic, 1988). Trauma has also been cited as a triggering factor (Sankhla, Lai, Jankovic, 1988). In most cases, the patients report to have learned to cope with these movements by engaging in more pleasant activities. The

contractions gradually increase in intensity and duration resulting in bruxism/clenching, loosened teeth and dislocation of condyles.

As the condition has no diagnostic test or biomarkers to date, so the different types of focal dystonias have not been assessed (Logroscino, Livrea, & Anaclerio, 2003). Differential diagnosis of orofacial dystonia includes temporomandibular joint disorders, spontaneous condylar dislocation, hemimasticatory or hemifacial spasms, orofacial dyskinesia, tardive dyskinesia; drug induced extra-pyramidal reactions (Jankovic, 1988). The clinical parameters serve as diagnostic and management guidelines for oro-facial dystonia (Logroscino et al., 2003). Currently there is no established cure for dystonia. Treatment options for oro-facial dystonia can be broadly divided into:

- 1) Medical management : this includes administration of pharmacological agents like anticholinergics, anticonvulsants, antiparkinsonians, benzodiazepines, carbamezipine, lithium and gabapentin, which in particular has been quite effective (Fahn & Jankovic, 1984; Goldman & Comella, 2003).
- 2) Chemodenervation: Injections of botulinum neurotoxin in the form of Botulinum toxin A and B in focal dystonia (Tan & Jankovic, 1999).
- 3) Surgical management including peripheral/central nervous system procedures: they are used as a last resort for patients not responding to medicinal and injection therapy. Peripheral procedures include TMJ arthroscopy and surgery, myotomies, rhizectomies and ramisectomies. CNS procedures involve deep brain stimulation or identification and ablation of a desired nucleus in the brain. Thalamotomy and pallidotomy have also been reported to

provide a delayed but lasting improvement in about 70% of the cases (Cooper, 1976).

The angle is the most common site of isolated fracture in mandibular bone.

This is due to following reasons:

- 1) Inherent weakness due to presence of impacted teeth;
- 2) Change in vector of osseous grains;
- 3) Splinting of posterior angular region by pterygomassetric sling
- 4) Prominence of angle region on lateral impact
- 5) Contrecoup forces from opposite parasymphysis

The mandible is attached to both elevators and depressors. These muscles have a coordinated but antagonistic action. If one group is activated, the other group relaxes. If both the groups are activated simultaneously, they act against each other. This causes the products of anaerobic metabolism to accumulate in the muscles which causes pain. This vicious cycle of dystonia and pain progresses, ultimately leading to debilitation of patient's physical and mental state.

The elevators include the pterygomassetric sling and the temporalis. The depressors include lateral pterygoid, mylohyoid, anterior belly of digastric and geniohyoid. The depressors, except for lateral pterygoid, are primarily attached to the anterior mandible and the elevators at the angle. When both the groups are simultaneously activated, there is a fulcrum created at the region just anterior to the attachment of the pterygomassetric sling. Repetitive and sustained spasms may lead to negative remodeling and demineralization. This makes the already weak angle region weaker. In this case, the patient had chronic dystonia which started with intermittent episodes. The muscles of the right side of face were involved to a greater extent than the other side. This could have caused spontaneous fracture of the weakened angle region upon

precipitation of a strong spasmodic episode. This was confirmed by the operative finding of a comparatively narrow and thin mandible in the region around fracture. Another reason for believing in spontaneous fracture is simultaneous fracture of contra lateral coronoid and a strong history given by the patient against any sort of trauma. Orofacial trauma (fracture of angle and coronoid) further would have acted as triggering factors causing exacerbation of dystonic movements. It would have led to further resorption of fractured ends, loosening of teeth, pain and inflammation at angle region.

A locking reconstruction plate of 2.4 mm, which was used for fixation, is a load bearing implant. The advantages of the locking plate over the earlier non-locking plates are well known. The chances of screws getting loosened due to muscular forces is close to nil owing to double fixation of screw to the plate and bone.

The herpes zoster in postoperative period was an additional complication which was managed effectively with acyclovir and carbamezipine. The onset of the attack could be attributed to extreme amount of psychological and physical stress. Additionally, there would have been an immune-compromised condition afflicted due to nutritionally deprived state and steroids for control of edema and inflammation.

After the first stage operation, there were multiple fracture segments and sequestrum at the operated site suggesting osteomyelitis.

Reconstruction plate has the potential to bear the entire non-functional as well as functional muscular load. These forces are transferred to the plate by the locking screws which are bicortically engaged in the bone. The plate didn't fracture despite of repetitive insult by dystonic forces. Probably the bone-screw interface didn't have enough

strength to hold against the forces being transmitted. This led to inflammation of the marrow ultimately leading to fracture of the segments. The fractured segments were held together by the fixation device but the inflammation present already was compounded. This precipitated the lytic process and rapid spread of the same, resulting in osteomyelitis. The effected ramus condyle unit was resected under general anesthesia but the reconstruction plate was left in situ to maintain the continuity. In the second stage, the patient became almost symptom free within 2 months of surgery. On 6 months follow-up, the patient had mild dystonic movements but her occlusion still remained collapsed. She had no pain and had significantly improved her quality of life. No intervention is being planned as of now until the dystonic movements completely subside in order to avoid further complications.

CONCLUSION

Presently, there is no specific treatment or cure for orofacial dystonia. Until a reliable treatment guideline has been established or any new treatments developed, complications will continue to occur which have to be treated symptomatically. A chronic intermittent spasm of orofacial muscles has the potential to negatively remodel the bone making it prone to fracture. A locking reconstruction plate is a minimum to bear the load of muscle spasms. The patient should be kept in close follow up as any recurrent episodes of dystonia may lead to implant failure. A retrospective view of the whole case still produces a question mark in the mind of the authors; "what better could have been done for the patient?" Any suggestions from peers are welcome.

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