Case Report

Tapia syndrome following TMJ gap arthroplasty: A case report and review of literature

ABSTRACT

Tapia syndrome is an extremely rare condition involving simultaneous paralysis of cranial nerves X (recurrent laryngeal branch) and XII. It is mostly believed to occur as a neuropraxic complication of intraoperative airway management. We present a unique case of a 17-year-old female with dysphonia, dysphagia, and deviation of tongue to the right side following temporomandibular joint gap arthroplasty for release of left TMJ ankylosis. A clinical diagnosis of Tapia's syndrome was made on exclusion of surgical or intracranial etiology and conservative management was performed. The aim of this study is to discuss the possible etiology, symptoms, and treatment of this disease along with a review of seven cases of Tapia syndrome associated with maxillofacial surgeries.

Keywords: Hypoglossal nerve, nerve paralysis, recurrent laryngeal nerve, Tapia syndrome, TMJ ankylosis, TMJ gap arthroplasty

INTRODUCTION

Tapia's syndrome, also known as "matador's disease",[1] involves concurrent paralysis of recurrent laryngeal branch of vagus and hypoglossal nerves. This rare condition was first described almost 117 years ago by a Spanish surgeon, Dr A Garcia Tapia, and continues to intrigue the medical fraternity till date.

While some case reports suggest an intracranial etiology,[2] most are believed to be due to a neuropraxic injury as a complication of perioperative airway management. Overinflation of the cuff during endotracheal intubation, excessive lateral flexion of patient's head intraoperatively, and extubation with inflated cuff or neurovascular trauma following laryngoscopy could be some of the probable causes.[3-7]

Diagnosis is based on clinical findings that include varying degrees of dysphonia, dyspnea, dysphagia, and deviation of tongue to the affected side.

Most studies also suggest a complete recovery of nerve function within six months.

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We present a case report of a 17-year-old patient with postoperative unilateral hypoglossal and recurrent laryngeal nerves palsy following TMJ gap arthroplasty for ankylotic release of left temporomandibular joint.

To our knowledge, any incidence of Tapia's syndrome after such a procedure has not been previously reported.

CASE REPORT

A 17-year-old female reported to our hospital with a complaint of difficulty in breathing, swallowing, and hoarseness of voice since one month.

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The patient first noticed these symptoms following her surgery of gap arthroplasty for ankylotic release of left TMJ, under general anesthesia, a month ago [Figure 1]. Patient had intratracheal intubation, and Ryle's nasogastric tube was positioned intraoperatively. Immediate post operatively, a Heister's appliance was also placed in the patient's oral cavity in order to maintain the gap between the osteotomized segments. Tracheostomy decannulation was performed 15 days after the surgery and hoarseness of voice with difficulty in swallowing was noted. The patient was then discharged from the previous hospital after counselling.

A few days after discharge, patient's symptoms worsened. She began to have difficulty in breathing again, in addition to the previously mentioned symptoms of hoarseness and dysphagia and thus reported to our center.

On intraoral examination, a mouth opening of 1.5 cm was observed, with tongue deviation to the right side, on protrusion [Figure 2]. No deviation of uvula was noted.

Cough on swallowing was observed. Laryngoscopy revealed unilateral vocal cord paralysis on the right side [Figure 3].

Neurological examination showed no evidence of sensory-motor disorder or any evidence of central cranial nerve involvement. No other pathological findings were observed on otolaryngological or maxillofacial examination. Results of all laboratory blood tests were within the normal range.

Since there was no sensory or motor nerve dysfunction prior to surgery and there was a very low likelihood of direct trauma to the involved nerves intraoperatively, a provisional diagnosis of right hypoglossal and right recurrent laryngeal nerve palsy was made, consistent with Tapia's syndrome.

Conservative management including oral corticosteroid therapy for 15 days, with multivitamins, speech, and swallowing therapy were initiated.

Patient was followed up after six weeks and had made a partial recovery with restoration of speech and swallowing reflexes. Slight deviation of tongue to the right side persisted.

DISCUSSION

Over the years, Tapia's syndrome, also known as matador's disease, has been reportedly associated with a range surgeries such as rhinoplasty,[3] septoplasty,[4] malar augmentation,[5] thoracotomy,[6] shoulder surgery,[7] cervical spine surgery,[8] zygomatic complex fracture fixation,[9] etc.

In our review of literature, we found only seven reported cases of Tapia's syndrome following maxillofacial surgeries, as summarized in Table 1. However, none of these included

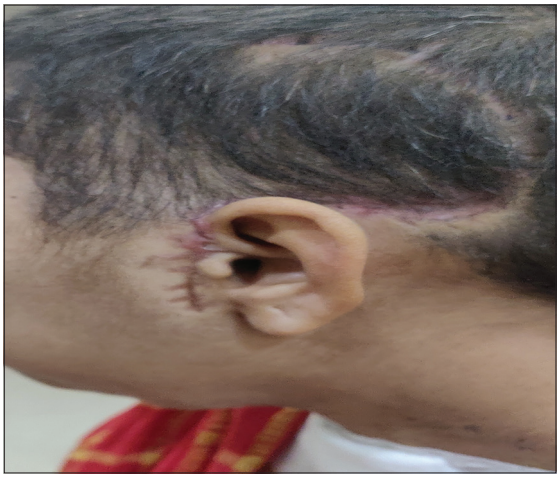

Figure 1: Left temporal and preauricular surgical scar post gap arthroplasty

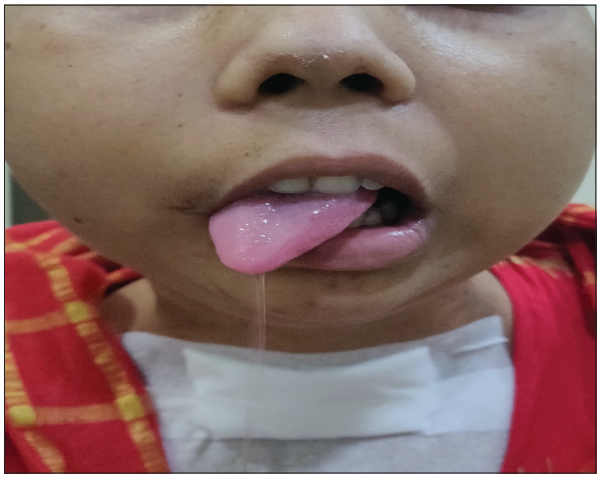

Figure 2: Tongue deviation to the right side on protrusion

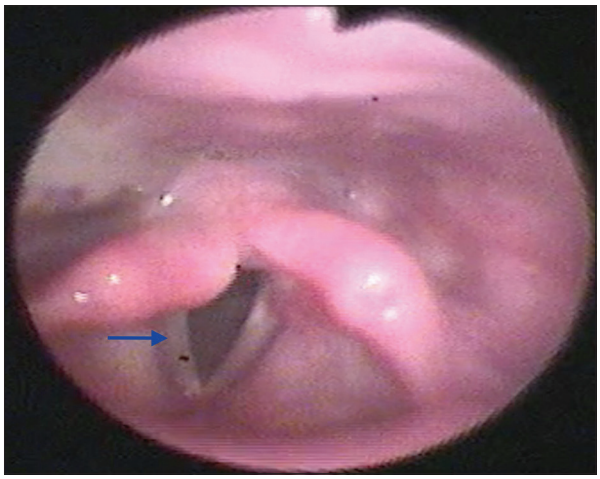

Figure 3: Laryngoscopy revealed right vocal cord paralysis

Table 1: Summary of seven cases previously reported after maxillofacial surgeries

Author	Year of publication	Gender	Age	Type of Surgery	Possible etiology	Treatment	Recovery period
Varedi *et al*.[9]	2013	M	31	ORIF after ZMC # fixation	Not mentioned	Oral prednisolone 60 mg/d, Vitamin B complex for two weeks; speech and swallowing therapy (two months)	Speech and swallowing after three months, tongue deviation after ninemonths
Kashyap *et al*.[13]	2010	M	41	ORIF following right parasymphysis and left condylar fracture	Not mentioned	Not mentioned	Incomplete recovery after one year and four months
Crespo *et al*.[1]	2021	M	25	ORIF after Left mandibular ramus and condylar fracture	Injury during intubation	Cytidine disodium with uridine phosphate (5 mg/3 mg for one month)	Complete recovery after six months
Ghorbani *et al*.[5]	2019	M	30	Malar augmentation	Pressure from the laryngoscopy to the tongue, placement of throat pack, or change in the position of the neck during intubation	Oral dexamethasone 0.5 mg, BD for two weeks along with intramuscular injection of neurobion every week	Complete recovery in six months
Mumtaz *et al*.[11]	2018	M	23	Surgical management of oroantral communication	Not mentioned	Supportive therapy along with speech and swallowing therapy	three months
Ota *et al*.[14]	2018	M	34	BSSO, lefort 1 osteotomy, genioplasty	Inappropriate use of a Macintosh blade to the lateral tongue base due to difficult intubation	Prednine for 20 days, ATP for five months and satellite ganglion block done 20 times	Complete recovery in three months
Fashina *et al*.[12]	2017	F	30	Excision of recurrent pleomorphic adenoma of parotid	Excessive compression of the lower oropharynx by the endotracheal tube and the throat pack or excessive anterior and lateral extension of the head	Neurobion tablets thrice a day with oral prednisolone 20 mg/day	Phonation after two weeks and tongue movements after two months

an association with TMJ gap arthroplasty, as is the case in the present scenario.

Depending on the degree of injury to the associated nerves, the symptoms may vary. Patients typically present with hoarseness, dysphonia, dysphagia, dyspnoea, caused as a result of unilateral paralysis of glottic muscles and palsy of the vocal cords. Clinical signs observed are tongue deviation on protrusion to the affected side along with ipsilateral paralysis of vocal cords, confirmed by laryngoscopy. While uncommon, bilateral involvement of these nerves, especially hypoglossal, may also occur.[3]

Some studies suggest a possible intracranial role caused by lesion in the brainstem at the nucleus ambiguous of XII and pyramidal tract and should be ruled out as a possible cause by computed tomography and magnetic resonance imaging.[2] However, most documented cases of Tapia syndrome have been reported to be extracranial in nature.[10-12]

Anatomically, hypoglossal nerve is believed to be in close proximity to the vagus nerve at the base of the tongue and pyriform fossa, around the junction of oropharynx and hypopharynx. A concomitant paralysis indicates a possible injury to the nerves at this site.[13]

A neuropraxic injury at the aforementioned site may be caused by:

1. Cuff of endotracheal tube: Overinflation (>20 cm H2O), inflation at improper position of tube in larynx instead of trachea, extubation with cuff inflated.[5,11,12]
2. Displacement of tube caused by: Excessive extension of the neck during laryngoscopy or surgery, intraoperative rotation or repositioning of head.[7,8]
3. Overpacking the throat pack in the region of hypopharynx and lower oropharynx: leading to pressure over tracheal tube or directly to pharyngeal soft tissues.[9,13]
4. Application of excessive force through Macintosh blade of laryngoscope.[14]

Alternatively, Boisseau *et al*.[7] suggested that dissection of ascending pharyngeal branch of carotid artery, which exclusively supplies both X and XII, could lead to their ischemic neuropathy and thus explain some of the symptoms reported.

In the present case, there is no direct correlation between the surgical site and the nerves affected. In maxillofacial region, the surgical site is adjacent to the airway, leading to a risk of possible misplacement of tracheal tube during surgical manipulation or due to rotation of patient's head on surgical table. Use and over packing of throat pack or a direct injury to pharynx due to repeated attempts at intubation, especially in cases with mandibular micrognathia, could also be the probable mode of injury in these procedures. In our patient, the above factors might have occurred individually or in combination.

In most literature, Tapia's syndrome has been found to be transient in nature.[3-7,9-14] Patients make a full recovery with conservative management, by an average of six months.[5,8]

Corticosteroid therapy is generally recommended to reduce inflammation of the pharyngeal soft tissues and nerves. Various authors have also used different dosages of oral corticosteroids, ranging from 0.5 mg dexamethasone twice a day,[5] to 60 mg/day prednisolone,[9] over an average of two weeks duration. Based on the degree of symptoms presented, Boğa *et al*.[4] formulated a treatment protocol for their use [Table 2]. However, due to the rare nature of this condition and its varying degrees of presentation, this suggested protocol may warrant further validation.

Use of multivitamins, especially B complex has also often been advised, given its role in aiding neural recovery.[7]

Some authors have also reported success with other adjunct therapies such as using a combination of oral corticosteroid therapy with ATP for five months and satellite ganglion block.[14] Crespo *et al*.[1] also suggested using cytidine disodium with uridine phosphate (5 mg/3 mg for one month), in order to stimulate the production of phospholipids and sphingolipids which form the basic structure of neural membrane and myelin sheath.

Furthermore, a combination of physiotherapy in the form of speech and swallowing therapy along with medical treatment was found to shorten the recovery period in a few studies.[7,9,12]

In some cases with severe neurological damage, an incomplete recovery may also be expected as in case reported by Kashyap *et al*.,[13] where symptoms continued to persist even after one year and four months post-operatively.

CONCLUSION

While Tapia's syndrome may be a temporary condition, it causes significant distress to the patient. Knowledge of the causes, symptoms, and management of such a rare syndrome is therefore extremely important for the surgeons and anesthesiologists. It becomes imperative for clinicians to follow proper protocols in anesthetic management and quick identification of the condition in order to initiate treatment as soon as possible, thereby reducing the recovery period.

Table 2: Atkins and Boga classification

Classification	Signs and symptoms	Treatment
Grade I/Mild	Unilateral vocal cord and tongue paralysis with no involvement of uvula, no swelling of tongue, or difficulty in swallowing	Conservative management without corticosteroids
Grade II/ Moderate	Vocal cord and tongue paralysis with mild difficulty with speech, swelling in tongue, difficulty swallowing, cracked speech	Oral Corticosteroid treatment for 15 days
Grade II/ Severe	Vocal cord and tongue paralysis, significant uvula distortion, significant difficulty with speech, swallowing, swelling in tongue, challenges in feeding and drinking	Intravenous Corticosteroid therapy for one week

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the legal guardian has given his consent for images and other clinical information to be reported in the journal. The guardian understands that names and initials will not be published and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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