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Case Report

Tapia's Syndrome after Cosmetic Malar Augmentation: a Case Report

Farhad Ghorbani [1], Saeid Tavanafar [1], Hamidreza Eftekharian [2]

[1] Postgraduate Student, Dept. of Oral and Maxillofacial Surgery, School of Dentistry, Shiraz University of Medical Sciences, Shiraz, Iran.
[2] Dept. of Oral and Maxillofacial Surgery, School of Dentistry, Shiraz University of Medical Sciences, Shiraz, Iran.

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ABSTRACT

Tapia's syndrome is an infrequent complication of airway manipulation. It is usually due to an extra-cranial ipsilateral injury to the hypoglossal nerve and the recurrent laryngeal branch of the vagal nerve, which can happen after any surgery. It is usually characterized by unilateral paralysis of the muscle of the tongue and vocal cords although it can also occur bilaterally. We present a patient with postoperative unilateral hypoglossal and recurrent laryngeal nerves palsy that occurred after cosmetic malar augmentation for esthetic correction of the left cheek flatness with an uncomplicated transnasal intubation. We report the first case of Tapia's syndrome after porous polyethylene implantation for cosmetic cheek reconstruction. The patient was treated immediately after the diagnosis with 0.5mg dexamethasone for two weeks. After three months, the movements of the vocal cord and tongue movement started to improve and the patient's hoarseness fully recovered after six months.

Corresponding Author: Tavanafar S., Dept. of Oral and Maxillofacial Surgery, Shiraz University of Medical Sciences, Shiraz, Iran. P.O.Box:71479-83889 Tel: +98-7136263193-4 Fax: +987132334481 Email: s.tavanafar@gmail.com , tavanafar@sums.ac.ir

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Introduction

Tapia's syndrome, also known as "Matador's disease", is named after Antonio Garcia Tapia who first described the unilateral paralysis of the tongue muscles and the vocal cord in a bullfighter that was struck by a bull behind his right angle of the mandible. [1] It is due to the concurrent unilateral paralysis of the hypoglossal nerve (XII) and recurrent laryngeal branch of the vagal nerve (X), although bilateral paralysis of these nerves is also reported in the literature. Based on the location of the nerve injury, this syndrome could be caused by the peripheral nerve damage (extra-cranial nerve), or central nerve damages (any damage to the nucleus ambiguous, hypoglossal nerve nucleus, and the pyramidal tract in the central nervous system). [1-2] Tapia's syndrome usually refers to the peripheral type of nerve damage due to direct trauma to the hypoglossal and recurrent laryngeal nerve at the base of the tongue and the pyriform fossae during surgeries requiring intubation or neck hyperextension. [3-4] The central type of this syndrome is less frequent and could be due to malignancies. Typical clinical findings of the peripheral unilateral type of this syndrome are hoarseness, difficulties in speech and swallowing, uncontrolled tongue movement, ipsilateral deviation of the tongue on the protrusion, and contralateral deviation on rest. [2-3, 5-7]

We report a case of Tapia's syndrome after cosmetic malar augmentation of the left cheek flatness by using porous polyethylene implant (Medpor, Stryker Craniomaxillofacial [CMF], Portage, MI). Tapia's syndrome is a rare complication of the airway manipulation and is possibly underestimated if practitioners are not familiar with its presentations. Acknowledging this syndrome through the publication of reporting cases will alert the scientific community to create a standard protocol for prevention, diagnosis, and treatment strategies.

Case Report

Before reporting this case, the patient signed an in form-

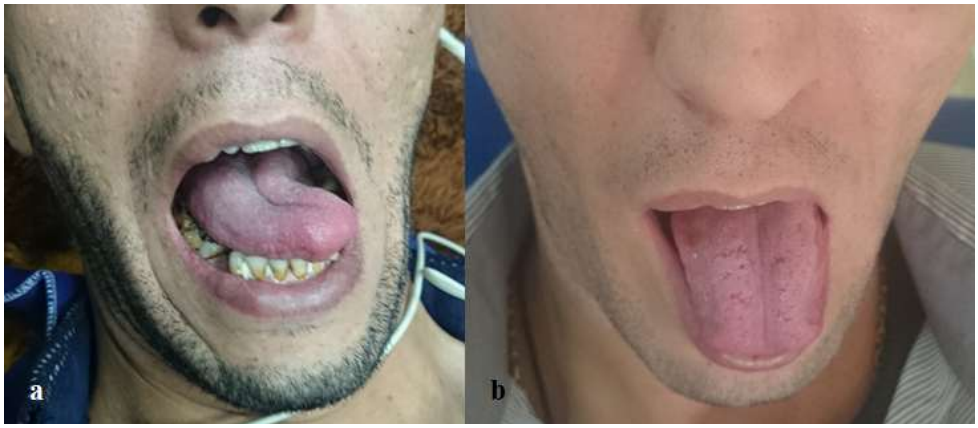

Figure 1a: Tongue deviation to left upon protrusion, **b:** Resolution of tongue deviation after three months.

ed consent. A 30-year-old male with a history of seven-month fracture of the left zygomaticomaxillary complex fracture due to the motor-vehicle accident was a candidate to receive a Medpor implant to correct the left cheek flatness. The patient was recommended to eat nothing by mouth for 6 hours before surgery, and no premedication was given. The anesthetic method for operation consisting of morphine 0.1 mg/kg was administered intravenously at induction of anesthesia which was achieved using 2-2.5 mg/kg propofol, 2-3 mcg/kg fentanyl; also, 0.5 mg/kg Atracorium was used to facilitate the transnasal intubation. Transnasal intubation was performed by passing the tube from the right nostril through the nose, and using a Macintosh blade the tube passed through the vocal apparatus into the trachea using a wire-reinforced, armored endotracheal tube with an internal diameter of 7.5 mm.

Anesthesia was maintained using 100-200 mcg/kg/min propofol and remifentanil 0.25 mcg/kg/min by continuous infusion. Non-invasive arterial pressures, electrocardiography, pulse oximetry, and capnography were monitored continuously. The patient was mechanically ventilated to keep EtCO2 between 35 and 40 mm Hg and normothermia was maintained in the operation theater.

The left subcilliary incision was made, and after flap reflection, the implant prosthesis was shaved, reshaped and inserted in the left cheek. The left zygomaticofrontal incision was also made to access the implant and reshape and fix the implant. The surgery lasted for one hour without any complication during the surgery. Extubation was performed by the removal of the throat packs, deflation of endotracheal cuff, and gentle withdrawal of the naso-tracheal tube.

Postoperatively, the patient complained of mild hoarseness, disturbed speech, and tongue deviation upon protrusion to the left (Figure 1a). Maintaining spontaneous ventilation under conscious sedation during awake fiber optic laryngoscope examination, the unilateral paralysis of the left vocal cord was evident. A spiral computerized tomography (CT) of the head and neck and magnetic resonance imaging (MRI) of the brain and neck ruled out any pathologic lesion of the central nervous system. All laboratory and blood tests were within the normal ranges. The diagnosis of Tapia's syndrome was made, and a management protocol for the patient was designed. The patient was prescribed intramuscular injected of Neurobion once a week and oral administration of 0.5 mg dexamethasone every 12 hours (twice daily) for two weeks. The patient also consulted with speech and language therapist, which monitored the symptoms regularly. The patient was followed by maxillofacial team weekly for one month and then followed monthly. Tongue deviation resolved after three months (Figure 1b) and the patient's hoarseness recovered after six months.

Discussion

We report the first case of unilateral Tapia's syndrome after cosmetic malar augmentation for the cheek flatness and third case of Tapia's syndrome with transnasal intubation. It is a highly rare condition with only a few reported cases to date in relation tp maxillofacial surgeries. [6-7] Tapia's syndrome can have a unilateral and bilateral presentation and could be due to peripheral or central nervous system damages. Literature reveals several causes of Tapia's syndrome such as rough endotracheal intubation, fungal infection, and neoplasm, vascu-

lar and traumatic problems that can lead to the unilateral or bilateral presentation of the symptoms. In the peripheral type of Tapia's syndrome, the pharyngeal wall and its underlying neurovascular structures (cranial nerves X and XII) can be damaged during laryngoscopy with excessive flexion of the head, excessive pressure during cuff inflation, positioning the cuff in the larynx rather than the trachea, or extubation while the cuff still is inflated. When the tracheal tube and its cuff compress the anterior branch of the inferior laryngeal nerves of the vagal nerves against the posterior-medial part of the thyroid cartilage, it may paralyze the recurrent laryngeal nerve. In our case, the left recurrent laryngeal nerve was damaged, which happens more usual than its right counterpart because it travels through the thoracic cavity, passes around the aortic arch, and then it returns to the neck to supply the vocal cords. [6-8]

In the same way, if the hypoglossal nerve is pressured by the endotracheal tube or the posterior part of the laryngoscope against the greater horn of the hyoid bone, it can damage the nerve. [4] Hypoglossal nerve which also passes through the posterior-medial of the angle of the ramus of the mandible and posterior to the lateral wall of the pharynx can be compressed in excessive flexion of the neck between the endotracheal tube and pharyngeal wall, ramus of the mandible and the cervical vertebra. [9-10] In our case, no apparent injury to the hypoglossal and recurrent laryngeal nerve was clinically visible. Central nervous system pathology was ruled out by negative CT scan and MRI. The possible cause of Tapia's syndrome in our case could be pressure from the laryngoscopy to the tongue, placement of throat pack, or change in the position of the neck during intubation. [4, 11]

Another mechanism of injury could be overstretching and compression of these two nerves at their crossing point of the first cervical transverse process due to hyperextension of the neck and positioning of the body on the surgical table, in combination with poor endotracheal intubation or tight packing of oropharyngeal. [4]

Tapia's syndrome is a diagnosis of exclusion and can be suspected rather quickly with a complete history and neurological examination of head and neck. CT and MRI of the brain and neck are essential to rule out the central part from the peripheral nature of the nerve damage with possible causes such as a tumor, hemorrhage, ischemia, and abscess formation. There is a classification and treatment protocol proposed by Aktas and Boga (Table 1). [5]

Although most of the reported cases are unilateral, bilateral cases of Tapia's syndrome have been reported. The most common feature in bilateral casesis the prolonged intubation under mechanical ventilators.

The period of recovery for Tapia's syndrome is between four to six months, which is suggestive of the neuropraxic type of injury due to either compression or stretching of the nerves. Complete recovery occurs in 30% of patients while 2/3 of the patients have incomplete (39%) or no improvement (over 26%). [12] The main treatment for this syndrome is corticosteroids, but other supportive treatment such as speech and swallowing therapy and warm moist air inhalation can help to reduce the patient's recovery period.

This syndrome can happen after any surgery with endotracheal intubation. Most of the previous cases had orotracheal intubation, but our case is the third case of Tapia's syndrome after nasotracheal intubation. Because of its morbidity, clinicians and anesthesiologist should be familiar with its preventive measures, diagnosis, and treatment modalities. Proper head positioning, smooth and gentle intubation and extubation, careful placement of the throat pack are necessary preventive methods.

Table 1: Aktas and Boga classification and treatment protocol for Tapia's syndrome

Classification	Signs and symptoms	Treatment
Grade I Mild type	Unilateral cord and tongue paralysis, no uvula distortion, minimal slowdown in speaking, no swelling in tongue, no swallowing problems	Corticosteroid treatment is not recommended.
Grade II Moderate type	Unilateral cord and tongue paralysis, no uvula distortion, mild slowdown in speaking, swelling in tongue, dryness in pharynx, trouble in swallowing, cracked in speech, and regular feeding and drinking	15 days of corticosteroid treatment is recommended.
Grade III Severe type	Unilateral cord and tongue paralysis, significant uvula distortion, great difficulty in speaking, swelling in tongue, dryness in pharynx, trouble in swallowing, and challenges in feeding and drinking	Intra-venous corticosteroid treatment is recommended for one week.

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Conflict of Interest

None declared.

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