CASE REPORT

# Tapia's syndrome after surgery for recurrent pleomorphic adenoma of the parotid gland

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## ABSTRACT

Tapia's syndrome consists of concurrent injury to the recurrent laryngeal and hypoglossal nerves. Trauma as a result of direct pressure from inflated cuff of the tracheal anesthetic tube and/or overextension of the neck during surgery have been reported to be possible causes of this syndrome. Here, we report a case of Tapia's syndrome following surgical excision of a very large recurrent parotid tumor. The aim of this report is to draw the attention of head and neck surgeons and anesthetists to this often unexpected condition. A 30-year-old female presented to the surgical outpatient clinic of the Lagos University Teaching Hospital with a massive, multinodular, right facial swelling. There was no sensory or motor nerve paresis on presentation. The patient underwent surgical excision of the swelling under general anesthesia. Two hours after extubation, the patient had difficulty moving the entire tongue and had difficulty with phonation. A working diagnosis of Tapia's syndrome was made based on clinical presentation and assessment. The patient was reassured and placed on tablets neurobion three times daily and tablets prednisolone 20 mg daily. Fourteen days after surgery, hoarseness of voice had resolved completely and full tongue control returned after 2 months. Tapia's syndrome must be considered, especially by all head and neck surgeons and anesthetists even though it is usually a rare complication of surgery.

**Key words:** Hypoglossal nerve, parotid gland, pleomorphic adenoma, recurrent laryngeal nerve, Tapia's syndrome

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## INTRODUCTION

Tapia in 1904 first described what is now known as Tapia's syndrome.[1] This relatively rare syndrome[2] consists of concurrent injury to the recurrent laryngeal and hypoglossal nerves either unilaterally in most cases[3-6] or bilaterally as reported in a few other cases.[4-6] It usually presents as difficulty in moving the tongue (or deviation to one side if unilateral), hoarseness of the voice, and disturbed swallowing and speech, all of which present immediately after surgery. Trauma as a result of direct pressure from inflated cuff of the tracheal anesthetic tube and/or overextension of the neck during surgery have been reported to be possible causes of Tapia's syndrome.[4,7,8]

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Here, we report a case of Tapia's syndrome following surgical excision of a very large recurrent parotid tumor which had extended into the submandibular regions. The aim of this report is to draw the attention of head and neck surgeons and anesthetists to this often unexpected condition, and to discuss its management in a resource-limited institution such as ours.

## CASE REPORT AND RESULTS

A 30-year-old female presented to the surgical outpatient clinic of the Lagos University Teaching Hospital with a massive recurrent right facial swelling extending



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into the submandibular region bilaterally [Figure 1]. A swelling from the samesite was observed 20 years before current presentation. During the 20-year period, the patient has had two major surgeries and several biopsies performed to treat the condition. The presenting swelling started about a year before presentation. On examination, a multinodular painless lesion measuring about 50 cm × 36 cm was observed. There was no sensory or motor nerve paresis on presentation. Radiographic investigation with computed tomography (CT-scan) revealed multiple nodular aggregates of soft tissue in the right cheek involving the submandibular and submental regions bilaterally. Fine-needle aspiration cytology of parotid and submandibular lesions was requested and the result revealed the lesion as benign. A working impression of recurrent pleomorphic adenoma was made.

The patient underwent surgical excision of the swelling under general anesthesia. Orotracheal intubation was used; the high-volume, low-pressure cuff of the tube was inflated using 10 ml of air and a throat pack was placed. Intraoperatively, it was found that the tumor was not attached to the underlying structures though tethered to the left submandibular gland and skin of the cheek in some areas. Thus, the surgical excision of the lesion involved a right total parotidectomy and excision of the left submandibular gland and affected skin areas [Figure 2]. The operation was carried out with the patient in a supine position with the head extended and turned from side to side on several occasions to gain access to the base of the tumor. Surgery lasted about 6 h, patient was transferred to the recovery room unextubated after the removal of the throat pack and was finally extubated uneventfully 15 h later.

Review of the patient 2 h postextubation revealed that the patient had difficulty with phonation and moving the entire tongue (demonstrated by patient's inability to raise the tongue to touch the palate and the corners of the mouth). There was also difficulty in swallowing though food placed at the back of the tongue could be swallowed and a gag reflex could effectively be stimulated. The neurology unit of the hospital on review gave a provisional diagnosis of a postoperatory neuropraxia of hypoglossal nerve and recurrent laryngeal branch of the vagus nerve postparotidectomy. This diagnosis was arrived at based on the facts that there was no sensory or motor nerve paresis before surgery, very low likelihood of direct trauma to any of these nerves during surgery, and the required head and neck positioning for this surgery which could have resulted in hyperextension of these nerves. In the absence of adequate finances for further investigations such as magnetic resonance imaging (MRI), a clinical diagnosis of Tapia's syndrome was made based on clinical presentation and assessment. The patient was reassured and placed on tablets neurobion three times daily and tablets prednisolone 20 mg daily. Six days after surgery, there was some improvement in the phonation although patient's voice was still hoarse. The tongue, however, remained flaccid. The prednisolone therapy was discontinued on the 6$^{th}$ postoperative day because patient suffered an extraoral wound breakdown. Ten days after surgery, patient claimed that there was mild fasciculation in the tongue though this could not be ascertained by the doctors. Fourteen days after surgery, the hoarseness had resolved completely and patient was discharged home with reassurance on the recovery of the tongue. At a 2-month postoperative visit [Figure 3], it was discovered that full tongue control had returned as patient could now raise the tongue to touch the palate and the corners of the mouth. Postoperative biopsy report confirmed tumor as recurrent pleomorphic adenoma.

## DISCUSSION

Tapia's syndrome refers to concurrent injury to the recurrent laryngeal and hypoglossal nerves. It could be central or peripheral. It is central when there is an intramedullary lesion of the nucleus ambiguus (of the hypoglossal nerve) and the pyramidal tract. Peripheral involvement occurs when there is concomitant injury to

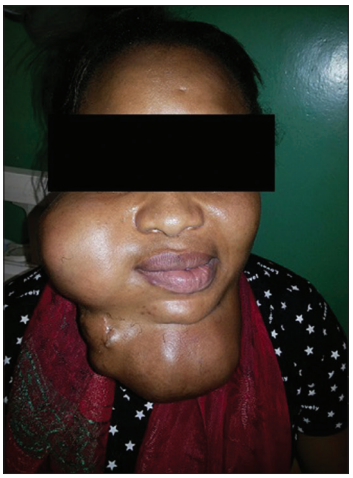

**Figure 1:** Clinical photograph of patient before surgery

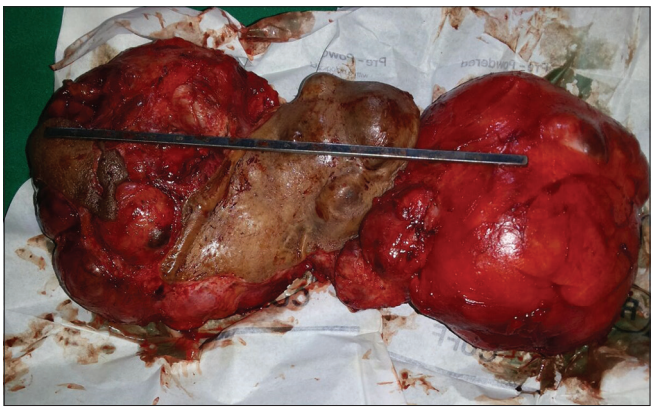

**Figure 2:** Photograph of tumor after excision

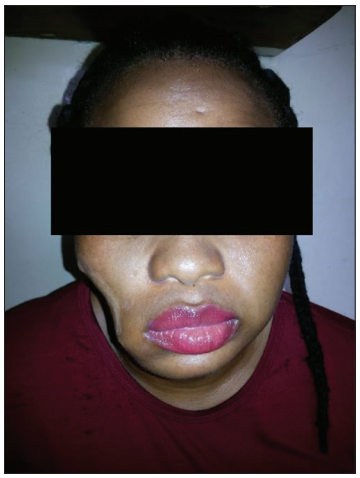

**Figure 3:** Clinical photograph of patient 2 months after surgery

these nerves at the base of the tongue where they lie in proximity.[9]

Lesions of these nerves may be a rare complication of anesthetic airway management or displacement of endotracheal tube during general anesthesia. Other reported causes are direct trauma, neurofibromatosis of X and XII nerves, carotid artery dissection involving the ascending pharyngeal artery.[2] Lo Casto *et al*. reported a case of nontraumatic Tapia's syndrome caused by a nasopharyngeal inflammatory pseudotumor.[10]

In the case we presented here, injury to the nerves could be as a result of one or a combination of both of the following mechanisms: (1) Excessive compression of the lower oropharynx by the endotracheal tube and the throat pack which can result in injury to these nerves as they travel along the lateral wall of the oropharynx in proximity; (2) excessive anterior and lateral extension of the head which may have resulted in stretching of the nerves. Structurally, the hypoglossal nerve crosses the vagal nerve where it rests on the most lateral prominence of the anterior surface of the transverse process of C1.[11] On hyperextension of this joint, these nerves would be stretched and pressed against this prominence. In this case, hyperextension of the neck was inevitable due to the size and position of the tumor and this appears to be the primary mechanism of injury. Furthermore, the use of endotracheal tubes with inflated cuffs and use of throat packs are routine for oral and maxillofacial surgery and ENT surgeries in this center and yet this is the first known reported case of Tapia's syndrome. This, therefore, agrees with several authors[2,7,12] who have postulated that though the actual compression is done by the tracheal tube and/or the throat pack, head position, especially neck hyperextension has a significant impact on the incidence of this syndrome.

In the head and neck region, reports of Tapia's syndrome appear to be most commonly related to rhinoplasty. Lykoudis and Seretis[9] while reporting a case of Tapia's syndrome following a rhinoplasty in 2012 found that of the other nine reported cases of Tapia's syndrome, half of the patients had undergone rhinoplasty or septorhinoplasty. Tapia's syndrome has also been reported following other head and neck surgeries such as repair of fractured mandible[13] and orthognathic surgery[12] which consisted of bilateral sagittal split ramus osteotomy, Le Fort I osteotomy, and genioplasty. Boisseau *et al*.[7] in 2002 reported Tapia's syndrome following a shoulder surgery in which patient was positioned upright throughout the surgery. In that report, however, when the surgical drapes were removed, the patient's head was found lying with very pronounced right lateral flexion because the body had moved. Our case report, according to our knowledge, is the first reported case of Tapia's syndrome following a parotid tumor excision. In line with other previously reported cases, the most likely mechanism of trauma to the hypoglossal and recurrent laryngeal nerves appears to be prolonged hyperextension of the neck during surgery.

A review of literature of reported cases from developed nations revealed that on initial presentation, several other investigations (such as cerebral CT, nuclear MRI, vertebral and carotid ultrasonography, and magnetic resonance angiography) were carried out to exclude other causes of the paresis of the nerves. Given limited resources in our environment coupled with unavailability of finances on the part of the patient, our diagnosis of Tapia's syndrome in this case was based strictly on clinical examination of the patient before and after surgery. The absence of any form of nerve paresis before surgery, timing of onset signs, and the specific nerves affected were key factors used to rule out other possible differentials in this case.

The type of nerve injury in Tapia's syndrome has been found to be neuropraxia.[2,6,14] This is evident by the fact that majority of cases resolved spontaneously with conservative management. Gevorgyan and Nedzelski[15] in 2013 reported that recovery is excellent in 30% of patients, incomplete in 39% of patients, and none in over 26% of patients. Period of recovery usually varies from about 2 weeks to several months after surgery.[2-16] In our reported case, the recurrent laryngeal nerve recovered first within 2 weeks after surgery while the hypoglossal nerve recovered fully about 8 weeks after surgery. This is similar to the report by Lykoudis and Seretis[9] where the recurrent laryngeal and hypoglossal nerves recovered at 3 weeks and 4 months, respectively, following a rhinoplasty.

The type of conservative management used in the management of Tapia's syndrome in literature varied from use of corticosteroids, vitamins, logopedic (speech), and swallowing therapy to use of nasogastric tubes for nutrition and to prevent aspiration. While the need for supportive treatment is apparent, there is no strong evidence in literature to support the need for corticosteroid therapy in Tapia's syndrome given that the type of injury is neuropraxia, hence expected to resolve

within a few weeks to months. In our case report, though prednisolone therapy was discontinued for other reasons, recovery period still fell within the range reported in literature which happens to be same for those who used corticosteroids[2,7,14] and those who did not.[12,16] Functional recovery in our patient was also progressive, similar to other cases in literature, and thus also suggests a neuropraxic type of nerve damage.

## CONCLUSION

Tapia's syndrome must be considered by all surgeons (especially head and neck surgeons) and anesthetists even though it is usually a rare complication of surgery. Recovery is often spontaneous and recovery period varies, however, supportive therapies such as speech and swallowing therapy have been reported to be helpful. Most importantly, Tapia's syndrome can potentially be prevented by paying attention to neck positioning during surgery and avoiding neck hyperextension when possible.

### Declaration of patient consent

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

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Nil.

### Conflicts of interest

There are no conflicts of interest.

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