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Case Report

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Speech therapy rehabilitation in Tapia´s syndrome: case report

Abstract

Tapia's syndrome is charaterized by damage to the recurrent laryngeal nerve (branch of the Xth) and the hypoglossal nerve (XIIth), and in some cases, the accessory nerve (XIth) may also be affected. The cause is usually trauma to the point where the hypoglossal and vagus intersect. This results in lingual hemiparesis, vocal fold paralysis and dysphagia. Objective: to present the results of speech therapy in a case that evolved with Tapia´s Syndrome after the excision of a tumour in the parapharyngeal space. Case report: male patient, 71 years old, submitted to excision of a Schwannoma in the vagus nerve, with parotid invasion. Evolved with: dysarthria; severe dysphagia; lack of flow control; dysphonia; facial paralysis and dyspnoea. Six months after surgery, thyroplasty and botulinum toxin injections were performed in the cricopharyngeal muscle and salivary glands, which ensured a temporary improvement in voice and control of salivary flow. Results: The rehabilitation process consisted of stimulating the mobility of the muscles responsible for facial mimicry, reintegration of stomatognathic functions, swallowing and voice. After two years of speech therapy, the patient showed improvement in the mobility of the facial mimic muscles, a slight improvement in the vocal pattern, in addition to an improvement in the breathing pattern. Conclusion: despite the limitations and functional complexities encountered in the postoperative period, there was evidence of improvement in the patient's quality of life after performing clinical procedures combined with the therapeutic process.

Keywords: syndrome, recurrent laryngeal nerve, hypoglossal nerve, swallowing disorders, dysphonia

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Introduction

Tapia's syndrome was described in 1904 by the Spanish otorhinolaryngologist Antonio García Tapia.[1] It is characterized by unilateral paralysis of the tongue and ipsilateral vocal cord. It is uncommon, and may have peripheral extracranial origin or centrally, even more rarely, due to a bulbar lesion of the nucleus ambiguous and pyramidal tract.[2] In most cases, peripheral injury is more frequently attributed to manipulation of the airways during surgery.[2,3]

According to[4,5,6] nerve injuries occur due to the anatomical proximity of hypoglossal and recurrent laryngeal nerves or, among other factors, injury during general anaesthesia; laryngoscopy; tube insertion and balloon pressure; mask ventilation; triple aerial manoeuvre; tube insertion mode during endotracheal intubation; head and neck position during aspiration.

Treatment includes speech therapy for swallowing, speech and voice rehabilitation. In most cases, the prognosis tends to be good with almost complete recovery after the injury in most cases because it is neuropraxia.[4-6] One should consider the etiological study of patients with injuries to cranial nerves IX, X, XI, XII, investigating skull, brain and cervical images (skull tomography and magnetic resonance imaging) to assess the presence of other alterations such as bleeding, fractures of the base of the skull and vascular study (neurosonology, angiotomography and/or magnetic resonance of the supra-aortic trunks) that exclude vascular lesion of the carotid or vertebral artery, which are alternative etiologies to the lesion of instrumentation of the airways.[7-10]

The parapharyngeal space is considered an area of complex anatomical relationships and is involved in a variety of benign and malignant neoplasms.[11,12] Among the primary neurogenic tumors in this region are schwannomas, which are extremely rare, mostly benign (70-80%) and represent less than 1% of head and neck tumors.[12,13]

The differential diagnosis of masses found in the parapharyngeal space is wide and may include deep parotid lobe tumours, tumours originating in the minor salivary glands, metastatic lymph nodes, branchial cysts, paragangliomas, lymphomas and internal carotid artery aneurysms.[13]

Schwannomas can originate in any nerve that has Schwan cells, except the optic nerve.[13,14] These tumours are usually encapsulated except those in the nasopharynx and sinonasal tract. The head and neck region are the most commonly affected (25-45%), with the lateral part of the neck being the most frequently involved area.[14,15] They tend to occur between the ages of 30 and 60[13,14] They present as asymptomatic, slow-growing masses and the symptoms depend on the anatomical location of the tumour and the nerve of origin. Among the symptoms reported in most cases, dysphagia, dyspnea and dysphonia are the most mentioned.[13-15]

In cases where the tumors manifest together in regions such as the parotid, the functional impairment may involve alterations in the stomatognathic functions, such as speech, mastication, suction, in addition to alterations in facial expressiveness.[16] Such changes can compromise the individual's emotional and psychosocial aspects, which directly interfere with their quality of life.[16] Nervous injuries in this region can affect a single isolated nerve or cause paralysis of two nerves together, such as the recurrent laryngeal and hypoglossal nerves.[17]

The vagus nerve (X pair) is mainly responsible for the sensory-motor actions of the larynx, pharynx and palate, including the cricopharyngeal muscle. Injury to the X pair and/or its branches can cause swallowing disorders of varying degrees, ranging from mild to severe. High lesions of the vagus nerve can generate not only paralysis of the ipsilateral vocal fold, but also loss of sensitivity in the hemilarynx. According to[12,16,17] recurrent laryngeal nerve injury may be responsible for pharyngoesophageal dysphagia in 11 out of 15 patients. Anatomically, we can observe that three to five branches

of the recurrent laryngeal nerve innervate the pharyngoesophageal transition, in addition to the motor innervation of the cricopharyngeal muscle involved in the swallowing process[17,18]

With regard to dysphonia, patients with paralysis caused by damage to the vagus nerve in its recurrent inferior laryngeal branch are characterized by having a hoarse, breathy, low-intensity voice, great vocal fatigue and the emission of short and choppy sentences. Furthermore, vocal fold paralysis determines an incomplete glottic closure and, therefore, a weak and ineffective cough, causing accumulation of secretions in the larynx with the need to constantly clear one's throat[18–20]

Speech therapy in cases of tumors of the parapharyngeal space has as its main focus to provide the rehabilitation of functionality, ensuring an improvement in the patient's quality of life.[18,19] The present study aims the results of speech therapy in a case that evolved with Tapia´s Syndrome after excision of a tumor in the parapharyngeal space.

Case Report

Male patient, 71years old, sought the hospital service after noticing bulging in the right parotid region. On 03/19/2020, he underwent a total parotidectomy; section of the digastric muscle, dissection and preservation of the hypoglossal nerve; dissection of the facial nerve and its branches; section of the vagus nerve (X pair) below the tumor lesion and dissection to the base of the skull and section with a transfixing stitch; and removal of the part (Schwannoma). He performed speech therapy during hospitalization and maintained follow-up after hospital discharge five times a week in an outpatient clinic. In the immediate postoperative period, he presented severe dysphagia, facial paralysis, hypoacusis, dysarthria and dysphonia. Due to the severity of the sequel, it was necessary to indicate an enteral diet through a nasogastric tube, evolving to the indication of gastrostomy even during hospitalization.

The therapeutic process consisted of stimulating the mobility of the muscles responsible for facial mimicry and the reintegration of stomatognathic functions. Oromyofunctional exercises were performed with cold thermal stimulation, toning and inducing massages, followed by isometric and isotonic exercises and proved to be efficient with gain in movement, isotonically, after four months. In addition to the proposed exercises, electrotherapy and photobiomodulation were performed twice a week.

In a new otorhinolaryngological evaluation, six months after the surgery, right vocal fold paralysis was identified with absence of constriction of the palatoglossal and palatopharyngeal pillars, absence of velopharyngeal closure, formation of cleft in abduction in all extension and accumulation of secretion in the supraglottic area and in piriform sinuses.

The vocal sequel was characterized by a breathy voice, medium pitch and reduced loudness, absence of modulation for high and low, strong and weak emissions, as well as intense pneumophonoarticulatory incoordination. In a new medical consultation, one year after the surgery, the patient underwent application of Botox in the cricopharyngeal muscle and medicalization thyroplasty (Ishiiki type I) and application of botulinum toxin in the salivary glands. After the procedure, the patient presented temporary but satisfactory results throughout the speech therapy, which provided an improvement in the vocal pattern, with improvement in glottic coaptation, minimizing breathiness and improvement in vocalization.

There was a limitation for speech therapy focused on dysphagia due to the extent of the lesion. In a videofluoroscopic exam carried out nine months after discharge, severe neurogenic dysphagia was evidenced, characterized by alteration in tongue propulsion and ejection, posterior escape, delay in the onset of the pharyngeal phase, laryngeal penetration, residues in valleculae and pyriform sinuses. Exercises aimed at improving elevation, anteriorization and stabilization of the hyoid-larynx set and efficiency of airway protection mechanisms were performed, but without success over one year.

Discussion

Tapia´s syndrome is characterized by unilateral paralysis of the tongue and vocal cords. It is caused by concomitant damage to the recurrent laryngeal and hypoglossal nerves. As it is a rare condition and with numerous sequelae involved, the recognition of the presence of concomitant paralyzes is an important step in the diagnosis and treatmen[t5,6,20,21]

Tumors of the parapharyngeal space are rare, with the four major primary tumors being: deep parotid lobe masses, minor salivary gland lesions, neuromas and glottic tumors.[23,24]

The vagus (Xth) and hypoglossal (XIIth) nerves are of great relevance for fundamental processes in human life, such as communication and feeding, since they are responsible, respectively, for the innervation of most of the laryngeal muscles, pharyngeal constrictor muscles, airway smooth muscle, intestine[25-27] and tongue mobility (28-31). Corroborating the findings in the literature[21,32] the isolated lesion of the hypoglossal nerve is usually unilateral and considered a rare postoperative complication with multifactorial causes.

According to [17,18,32,33] the vagus nerve injury can cause cricopharyngeal dysfunction, clarifying the severe dysphagia in the case studied.

Based in literature, we observed in the patient the deviation of the tongue to the right side together with the loss of strength in the ipsilateral half-tongue and absence of mechanical restraint, which suggest a sequel to the XII cranial nerve; while dysphagia, hoarseness and paralysis of the right vocal fold suggest sequelae of the X cranial nerve.[32,33]

Dysphagia and dysphonia presented by the patient are deficits commonly found after excision of parapharyngeal space tumors with postoperative cranial nerve dysfunction.[34,35] Confirm that early assessment and speech therapy offer better prognosis in functionality rehabilitation.[18,19,34,35]

It is known that the clinical manifestations presented in facial paralysis range from facial asymmetry to form and function disorders such as incomplete eye closure, nasolabial sulcus deviation, reduced orbicularis oris strength, inability to and disfigurement of the muscles of facial mimicry. In this study, it was observed, through the performance of exercises for the stomatognathic functions and mainly aimed at the rehabilitation of facial mime, which had as objectives the gain of tonus and strength of the oral musculature.[36,37]

According to the literature and observed in the reported case, photobiomodulation[38–42] and electrostimulation[43–45] associated with orofacial motor exercises[38,41,42,44] promoted positive results in control of saliva flow and ensure greater oromiofacial mobility, accelerating strength gain, maintaining fatigue levels and improving muscle performance. There are still limitations in the literature that do not allow a deeper understanding of the issue, as few articles related speech therapy rehabilitation in patients diagnosed with Tapia´s Syndrome. More studies are needed. This case report shows that

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the speech therapy intervention had limitations due to the sequelae found in the postoperative period but obtained satisfactory results as symmetry and movement in the orofacial muscles can already be observed, facilitation of oral communication by voice improvement and dysarthria and consequent better quality of life as a result of the therapeutic process.

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Conflicts of Interest

There was no conflict of interest in this research.

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