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SYSTEMATIC REVIEWS

Isolated bilateral Tapia's syndrome after liver transplantation: A case report and review of the literature

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Abstract

AIM

To describe one case of bilateral Tapia's syndrome in a liver transplanted patient and to review the literature.

METHODS

We report a case of bilateral Tapia's syndrome in a 50-year-old man with a history of human immunodeficiency virus and hepatitis C virus child. A liver cirrhosis and a bi-nodular hepatocellular carcinoma, who underwent liver transplantation after general anesthesia under orotracheal intubation. Uneventful extubation was performed in the intensive care unit during the following hours. On postoperative day (POD) 3, he required urgent re-laparotomy due to perihepatic hematoma complicated with respiratory gram negative bacilli infection. On POD 13, patient was extubated, but required immediate re-intubation due to severe respiratory failure. At the following day a third weaning failure occurred, requiring the performance of a percutaneous tracheostomy. Five days later, the patient was taken off mechanical ventilation and severe dysphagia, sialorrea and aphonia revealed. A computerized tomography and a magnetic resonance imaging of the head and neck excluded central nervous injury. A stroboscopy showed bilateral paralysis of vocal cords and tongue and a diagnosis of bilateral Tapia's syndrome was performed. With conservative management, including a prompt establishment of a speech and swallowing rehabilitation program, the patient achieved full recovery within four months after liver transplantation. We carried out MEDLINE search for the term Tapia's syndrome. The inclusion criteria had no restriction by language or year but must provide sufficient

available data to exclude duplicity. We described the clinical evolution of the patients, focusing on author, year of publication, age, sex, preceding problem, history of endotracheal intubation, unilateral or bilateral presentation, diagnostic procedures, type of treatment, follow-up, and outcome.

RESULTS

Several authors mentioned the existence of around 70 cases, however only 54 fulfilled our inclusion criteria. We found only five published studies of bilateral Tapia's syndrome. However this is the first case reported in the literature in a liver transplanted patient. Most patients were male and young and the majority of cases appeared as a complication of airway manipulation after any type of surgery, closely related to the positioning of the head during the procedure. The diagnosis was founded on a rapid suspicion, a complete head and neck neurological examination and a computed tomography and or a magnetic resonance imaging of the brain and neck to establish the origin of central or peripheral type of Tapia's syndrome and also the nature of the lesion, ischemia, abscess formation, tumor or hemorrhage. Apart from corticosteroids and anti- inflammatory therapy, the key of the treatment was an intensive and multidisciplinary speech and swallowing rehabilitation. Most studies have emphasized that the recovery is usually completed within four to six months.

CONCLUSION

Tapia's syndrome is almost always a transient complication after airway manipulation. Although bilateral Tapia's syndrome after general anesthesia is exceptionally rare, this complication should be recognized in patients reporting respiratory obstruction with complete dysphagia and dysarthria after prolonged intubation. Both anesthesiologists and surgeons should be aware of the importance of its preventing measurements, prompt diagnosis and intensive speech and swallowing rehabilitation program.

Key words: Liver transplantation; Follow-up; Outcome; Postoperative complications; Bilateral Tapia's syndrome

Core tip: Tapia's syndrome is a rare entity characterized by the concomitant extracranial injury of the hypoglossal nerve (Ⅻ) and the recurrent laryngeal branch of the vagus nerve (Ⅹ) at the base of the tongue and the pyriform fossa. Anesthesiologists, surgeons and otorhinolaryngologist should be aware of its presentation at any type of surgery as in the present case, after liver transplantation. The purpose of this study is to present our even rarer presentation of bilateral Tapia's syndrome to the liver transplant community and to review the literature to update the current management and treatment. The most relevant common feature in most cases of bilateral syndrome was orotracheal intubation prolonged for more than 14 d.

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INTRODUCTION

Tapia's syndrome was described for the first time by the Spanish otorhinolaryngologist Antonio García Tapia in 1904[1]. It is characterized by the unilateral paralysis of the tongue and the vocal cord caused by extracranial injury to the hypoglossal nerve (Ⅻ) and the recurrent laryngeal branch of the vagal nerve (Ⅹ) at the base of the tongue and the pyriform fossa)[1-6]. Although the Tapia's syndrome refers to the extracranial lesion of the hypoglossal and recurrent laryngeal nerves, some authors also describe a central type of Tapia's syndrome, referring to those patients with the same symptoms, but whose damage has occurred in the nucleus ambiguous, the nucleus of the hypoglossal nerve, and the pyramidal tract in the central nervous system. We describe one case of bilateral Tapia's syndrome in a liver transplant patient, which is not previously reported in the literature.

MATERIALS AND METHODS

We report herein a case of bilateral Tapia's syndrome together with a review of the literature. We carried a literature research in the MEDLINE database through the PubMed search service for the term Tapia's syndrome. The inclusion criteria had no restriction by language or year but must provide sufficient available data to exclude duplicity. We described the clinical evolution of the patients, focusing on author, year of publication, age, sex, preceding problem, history of endotracheal intubation, unilateral or bilateral presentation, diagnostic procedures, type of treatment, follow-up, and outcome.

Case report

A 50-year-old man with a history of human immunodeficiency virus (HIV) and hepatitis C virus positive serology, with class A of Child-Pugh classification liver cirrhosis and a bi-nodular hepatocellular carcinoma underwent liver transplantation after general anesthesia under orotracheal intubation. Body mass index at time of transplantation was 21 kg/m^2. An 8.0 endotracheal tube was placed. The cuff was inflated with 3 mL of air and verified with a manual manometry to reach a filling pneumotamponade of 20 cm water. Surgery lasted 375 min. The procedure was well tolerated and required a low dose of inotrops (noradrenalin 0.5 mL/h) during surgery. Immunosuppression therapy during induction was based on mycophenolate mophetil and tacrolimus. Patient was transferred to the intensive care unit (ICU) under mechanical ventilation, sedated with remifentanyl. Uneventful weaning was performed during the following

hours. On postoperative day (POD) 3, he required urgent re-laparotomy due to a perihepatic hematoma and was transferred to the ICU under mechanical ventilation, sedated with propofol and remifentanyl. Extubation was postponed due to a respiratory gram negative bacilli infection and agitation after several attempts of decreasing sedation. On POD 13, patient was extubated and required immediate re-intubation after severe respiratory failure. A third weaning failure occurred the following day requiring re-intubation for the third time. Then percutaneous tracheostomy was performed with no events. Five days later, patient was taken off mechanical ventilation progressively and oral diet was started the day after, appearing severe dysphagia and important sialorrhea, being hardly able to swallow a pureed diet. Aphonia was another significant symptom presented at that time. At POD 28 patient was decanulated and persisted with swallowing difficulty, requiring parenteral nutrition. A computerized tomography (CT) of the head and neck and a magnetic resonance imaging (MRI) of the brain and neck were then performed to exclude central nervous injury. Both explorations did not show pathological findings.

At POD 34, patient was transferred to the ward and enteral nutrition was initiated *via* nasogastric tube. He was evaluated by speech and swallow theraphysts and diagnosis of a bilateral tongue paralysis and aphonia was made. Evaluation by otorhinolaryngologist excluded a recurrent laryngeal nerve injury. Detailed neurological examination revealed bilateral tongue paralysis, severe dysarthria and dysphagia for liquids and solids. A stroboscopy was performed showing bilateral paralysis of vocal cords in addition to the bilateral tongue paralysis. Cervical electromyography was also performed. Bilateral Tapia's syndrome was then diagnosed; a bilateral hypoglossal and laryngeal recurrent nerve neuroapraxia. At three months post-transplant, subjective improvement in aphonia and dysphagia were observed and the patient was discharged with enteral nutrition.

Outpatient neurological follow-up regarding speech and swallow training was performed twice weekly. Satisfactory recovery of his aphonia and dysphagia were observed. At four months post-transplant, videofluoroscopy was performed with no significant findings; however, laryngeal stroboscopy showed severe hypomotility of cricoaritenoideal articulations, cordal atrophy and minimal adduction movements with severe longitudinal hiatus. Despite that, the patient presented no problems during intake, being able to take out the nasogastric feeding tube. At that time, the nasogastric tube was preferred to the percutaneous gastrostomy to avoid invasive procedures in a patient with a complex postoperative.

RESULTS

In total around 70 cases were initially described in the literature, but only 53 fulfilled the inclusion criteria: To have patients with sufficient available data in the description of cases in order to rule out duplicity. Table 1[1-2,7-51] summarizes the 54 cases (including ours) of Tapia's syndrome, focusing on author, year of publication, age, sex, preceding problem, history of endotracheal intubation, unilateral or bilateral presentation, diagnostic procedures, type of treatment, follow-up and outcome.

The majority were young. Only 13 cases were older than 50 years (range 16-95). All cases except 10 were males. Two cases were attributed to a central cause (metastatic hemangiosarcoma in the medulla oblongata[2] and infiltration of a large B-cell lymphoma[14]), but the remaining 53 patients were peripheral type. Six patients[8,22,24,36,42,43], apart from ours, had a bilateral presentation of the syndrome; four with complete deficit of hypoglossal and recurrent laryngeal nerves and three[22,24,43] incomplete with bilateral paralysis of the hypoglossus nerves and unilateral recurrent laryngeal nerve palsy. All the cases, except one[36], followed to a prolonged oro-tracheal intubation for more than 14 d. In the systematic review, we have found two other cases of isolated bilateral hypoglossal paralysis without other nerve involvement after oro-tracheal intubation[52,53].

All, except seven of peripheral cases[9,15,29,39,40,47,51], have been attributed to orotracheal intubation for surgery or respiratory failure. The most frequently involved operations were: Osteoarticular surgery of the shoulder, mandible and cervical spine in 14 cases, otorhinolaryngology surgical procedures in 11 cases, cardiac surgery in 4 cases, thoracic surgery in 2 cases, abdominal surgery in 2 cases, and direct traumatic nerve injury in 2 cases. However, several causes have been described in the literature such as: Vascular (vertebral artery dissection, carotid artery aneurism); metastasic or primary neoplasia (lymphoma, hemangiosarcoma, prostate, pseudotumor of the neck, nasopharyngeal carcinoma, neurilemoma, neurofibrome, *etc.*); infectious of the neck (bacterial, viral, fungal), *etc*.

The diagnosis and management of Tapia's syndrome in the majority of cases was based on a complete neurological examination, including laryngeal endoscopy and a head and neck CT or MRI. Some authors have advocated for the use of video-fluoroscopic swallowing and electromyography to confirm the diagnosis and to predict prognosis.

The treatment was supportive in all cases with a prompt establishment of a swallowing rehabilitation program. The administration of intravenous or oral steroids in combinations with B1, B6, B12 vitamins or hialuronic acid injection has been proposed by many authors in the acute setting. At least 4 patients[8,17,23] required percutaneous endoscopic gastrostomy and 2 a naso-gastric tube insertion[20,42] to ensure nutritional requirements while the oro-esofageal route was unable to be used. In two cases (Takimoto[43] and ours), where bilateral paralyses were discovered, reintubation with subsequent tracheotomy was necessary to prevent respiratory failure.

Recovery was excellent for the majority of non-tumour peripheral cases after a duration of 3 to 6 mo, ranging from 15 d to 3 years. In 9 cases the patients reported only

Table 1 Cases of Tapia's syndrome reported in the literatura to date (including our case): 54 peripheral type and 2 central type

Ref.	Age	Sex	Clinical procedure	OTI	Bil	Diagnosis	Treatment	Follow-up	Recovery
Bilbao 2016	50	M	Liver transplantation due to HCV cirrhosis coinfected with HIV and hepatocellular carcinoma	Yes	Yes	N eurological examination Electromiography Laryngeal endoscopy Head and neck CT and MRI Video fluoroscopic examination	Temporary tracheotomy for airway management Nasogastric tube feeding Speech and swallowing therapy	4 mo	Yes
Cariati *et al*[7] 2016	36	M	Neck abscess drainage	Yes	No	Neurological exam Barium swallow X-ray Swallowing endoscopy	Rehabilitation program	3 mo	Yes
	61	M	Neck abscess drainage	Yes	No	Neurologic exam Airway endoscopy	Rehabilitation program	3 mo	Yes
	42	M	Shoulder fracture reduction	Yes	No	Neurologic exam Airway endoscopy	Rehabilitation program	3 mo	Yes
Coninckx *et al*[8] 2015	64	M	Liver cirrhosis. Pneumonia and respiratory failure	Yes	No	Neurological examination Lumbar puncture Laryngeal endoscopy Head and neck CT and MRI Chest CT	Speech and swallowing therapy Percutaneous endoscopic gastrostomy	22 mo	Yes
	49	M	Myocardial infarction. Percutaneous coronary intervention. Penumonia	Yes	Yes	Neurologic examination Brain CT	Corticosteroid therapy 8 wk Speech and swallowing therapy Percutaneous endoscopic gastrostomy	4 mo	Yes
Yilmaz *et al*[9] 2015	61	M	Bone metastatic prostate cancer	No	No	Neck CT and MRI	-	-	-
Paramalingam *et al*[10] 2015	38	M	Eagle syndrome. Pneumonia	Yes	No	Head and neck CT			
Brandt *et al*[11] 2015	23	M	Otorhinolaryngology surgical procedure	Yes	No	-	-	-	-
	67	-	Arthroscopic intervention of left shoulder	Yes	No	-	-	-	-
Ghorbani *et al*[12] 2014	27	M	Septorhinoplasty	Yes	No	Neurological examination Head and neck MRI	Systemic corticosteroids	6 mo	Yes
Ulusoy *et al*[13] 2014	19	F	Nasoseptal deformity	Yes	No	Neurological examination Head and neck MRI Airway endoscopy	Systemic corticosteroids	6 mo	Yes
Cantalupo *et al*[14] 2014	16	M	Large B-cell Lymphoma	No	No	-	-	-	-
Lo Casto *et al*[15] 2013	42	F	Inflammatory pseudotumor of the neck	No	No	Neurological examination Electromiography Laryngeal endoscopy Head and neck MRI Chest and abdomen CT	Corticosteroid therapy	-	-
Kang *et al*[16] 2013	47	M	Cervical spine surgery	Yes	No	Head and neck CT and MRI	Corticosteroid therapy Speech therapy rehabilitation	8 mo	Partially
Emohare *et al*[17] 2013	17	M	Artrodesis T1-L1	Yes	No	Barium swallow X-ray Head and neck MRI Airway endoscopy	Percutaneous endoscopic gastrostomy Hialuronic acid inyection Rehabilitation program	1 mo	Yes
Varedi *et al*[18] 2013	27	M	Zygomatic complex fracture	Yes	No	Neurological examination Head and neck CT and MRI Laringoscopic examination	Systemic corticosteroids Vitamin B complex Rehabilitation program	9 mo	Yes
Gevorgyan *et al*[19] 2013	48	F	Liposuction 3 yr previously rhinoplasty 25 yr previously	Yes	No	Neurological examination Head and neck CT and MRI Laringoscopic examination	Vocal cord injection Rehabilitation program	3 yr	Partially
Lim *et al*[20] 2013	64	M	Cervical spine surgery	Yes	No	Neurological examination Head and neck CT and MRI Laringoscopic examination Video fluoroscopic examination	Systemic corticosteroids Electrical stimulation therapy Nasogastric tube feeding	3 mo	Yes
Park *et al*[21] 2013	53	M	Posterior cervical spine surgery Posterior cervical spine surgery	Yes	No	Head and neck CT and MRI Laryngeal electromyography	-	6 mo	Yes
	56	M		Yes	No	-	-	2 mo	Yes

Sønnichsen *et al*[22] 2013	-	-	Legionella infection	Yes	Yes	-	-	2 mo	Partially
Nalladuru *et al*[23] 2012	49	M	Cardiac surgery	Yes	No	Neurological examination Head and neck CT and MRI	Systemic corticosteroids Percutaneous endoscopic gastrostomy	2.5 mo	Yes
Turan *et al*[24] 2012	15	M	Acute lymphoblastic leukemia pneumonia	Yes	Yes	Neurological examination Laringoscopic examination	Systemic corticosteroids	0.5 mo	Partially
Wadelek *et al*[25] 2012	57	M	Arthroscopic shoulder	Yes	No	Neurological examination Head and neck MRI Laryngeal endoscopy	Rehabilitation program	+ 2 mo	Yes
Lykoudis *et al*[26] 2012	32	M	Rhinoplasty	Yes	No	Head and neck CT Laryngeal endoscopy	Oral corticosteroid therapy Speech and swallowing therapy	4 mo	Yes
Park *et al*[27] 2011	42	M	Anterior cervical spine surgery	Yes	No	Neurological examination Electromyography Video fluoroscopic swallowing Laryngeal endoscopy Head and neck MRI	Rehabilitation program	7 mo	Yes
Torres-Morientes *et al*[28] 2011	32	M	Tracheostomy and right thoracostomy	[1]	No	Neurological examination	Speech and swallowing therapy	4 mo	Yes
Al-Sihan *et al*[29] 2011	63	M	Vertebral artery dissection	No	No	-	Clopidogrel for 6 wk Speech and swallowing therapy	-	Partially
Kashyap *et al*[30] 2010	41	M	Mandibular fracture	Yes	No	-	None	16 mo	Partially
Rotondo *et al*[31] 2010	-	-	Cardiac surgery	-	-	-	-	-	-
Boğa *et al*[32] 2010	35	M	Septorhinoplasty	Yes	No	-	Systemic corticosteroids	0.5 mo	Yes
Dursun *et al*[33] 2007	-	-	Hunting rifle-shot	-	-	-	-	-	-
Sotiriou *et al*[34] 2007	-	-	Coronary bypass grafting surgery	Yes	-	-	-	-	-
Tesei *et al*[35] 2006	30	F	Rhinoplasty	Yes	No	Neurological examination Head and neck MRI	Systemic corticosteroids Speech and swallowing therapy	4 mo	Yes
Cinar *et al*[36] 2005	20	M	Open rhinoplasty	Yes	Yes	-	Systemic corticosteroids	1 mo	Yes
Yavuzer *et al*[37] 2004	42	F	Septorhinoplasty	Yes	No	-	Oral corticosteroid therapy	6 mo	Yes
Krasnianski *et al*[2] 2003	77	M	Metastasic hemangiomasarcoma in the medulla oblongata	-	No	-	None	-	-
Boisseau *et al*[38] 2002	42	M	Shoulder surgery	Yes	No	Vertebral and carotid ultrasonography Head and neck CT and MRI	Systemic corticosteroids Speech and swallowing therapy	6 mo	Yes
Johnson *et al*[39] 1999	44	M	Surgical repair of a shoulder injury	No[1]	No	Head and neck CT and MRI	None	2 mo	Partially
Shimohata *et al*[40] 1994	61	F	Aneurism of extracranial internal carotid artery	No	No	Carotid angiography Head and neck CT and MRI	-	-	-
Millán Guevara *et al*[41] 1993	-	-	Viral etiology?	-	-	-	-	-	-
McCleary *et al*[42] 1993	95	F	Fracture of the odontoid process	-	Yes	-	Naso-gastric tube	12 mo	Partially
Takimoto *et al*[43] 1991	18	F	Nasopharyngeal carcinoma radiation	-	Yes	-	Temporary tracheotomy for airway management during pregnancy	4 yr	No
de Freitas *et al*[44] 1991	37	F	Paracoccidiodomycosis fungus in the nasal mucosa	-	-	-	Oral Ketoconazol	2 yr	No
Quattrocolo *et al*[45] 1986	24	M	Neurilemoma of vagus and hypoglossal nerves	-	-	-	-	-	-
Gelmers *et al*[46] 1983	41	M	Thoracotomy	Yes	No	-	-	12 mo	No
	36	M	Thoracotomy	Yes	No	-	-	12 mo	No
Andrioli *et al*[47] 1980	25	M	Neurofibrome of X and XII nerves below the nodose ganglion	No	No	-	Surgery: Resection of the two nerves	-	No
Mayer *et al*[48] 1974	51	M	Hiatus hernia repair. Pneumonia	Yes	No	-	None	0.5 mo	Partially
Ruhrmann *et al*[49] 1963	-	-	Congenital	-	-	-	-	-	-
Babini *et al*[50] 1961	-	-	Obstetrical trauma	-	-	-	-	-	-

Symonds *et al*[51] 1923	35	F	Chronic otitis media	No	No	-	-	2 yr	Partially
Tapia *et al*[1] 1905	-	M	Bullfighter injury behind the angle of the jaw		No				

Interscalene brachial plexus block [1]Tracheostomy. OTI: Orotracheal intubation; BIL: Bilateral; F: Female; M: Male; CT: Computed tomography; MRI: Magnetic resonance imaging; HCV: Hepatitis C virus.

partial recovery.

DISCUSSION

The case described above, is the first reported case of complete bilateral Tapia's syndrome (paralysis of the tongue' muscles and vocal cords because of an extracraneal injury of the Ⅹ and Ⅻ cranial nerves) occurring after liver transplantation and oro-tracheal general anaesthesia requiring re-intubation for three times. There are many causes of Tapia's syndrome, including general anaesthesia, fungal infections[44], neoplasms[2,9,14,15,24,43,45,47], vascular[29,40] and traumatic problems[1,33,50], being general anaesthesia the main cause. Intubation tube or its cuff and motion of the head during surgery can lead to injury to the pharyngeal wall and its underlying neurovascular structures (Ⅹ and Ⅻ cranial nerves)[32]. Excessive dorsiflexion of the head during laryngoscopy, excessive cuff pressure, malposition of the cuff in the larynx rather than the trachea, or extubation while the cuff is still inflated is the most likely cause[18]. The tracheal tube and its cuff may press on a localized area just at the crossing of the vagal and hypoglossal nerves, compressing the anterior branch of the inferior laryngeal nerve against the posteromedial part of the thyroid cartilage and this can lead to a recurrent laryngeal paralysis[6]. Hypoglossal nerve damage can be caused by a stretching of the nerve against the greater horn of the hyoid bone by an orotracheal tube or compression of the posterior part of the laryngoscope or orotracheal tube[35]. There was no clear mechanism for injury to the hypoglossal and recurrent laryngeal nerves in our patient. Intracraneal pathology was unlike because of negative CT scan and MRI. We postulate that low blood pressure during surgery and post-operatively due to intrabdominal hemorrhage requiring reintervention and the need of several orotracheal reintubations (3 times), 2 of them in emergency conditions, in addition to prolonged intubation with probable unnoticed overinflation and malposition of the endotracheal cuff, might have been the source of the bilateral nerve compression. A change in the position of the neck at some point, compression by the endotracheal tube and pressure to the lateral roots of the tongue with the McIntosh blade during intubation could be additional mechanisms. The caquexia of the patient and some degree of lypodistrophya due the HIV coinfection at time of transplant could also play a role. Liver transplantation is usually a long lasting surgical procedure, which could contribute, along with other factors to the development of Tapia's syndrome. This fact should be taken into account by all clinicians involved in the liver transplantation care: Liver surgeons, anesthetists, intensivists, hepatologists, gastroenterologists, *etc*.

Although most patients were male and young, there is no an explanation to relate the syndrome to sex or age. We believe that this syndrome is more related to anatomical, positional and lasting-time issues than to other characteristics.

The diagnosis is founded on a rapid suspicion, a complete history around the paralysis and a complete head and neck neurological examination. A computed tomography and or a magnetic resonance imaging of the brain and neck is essential to establish the diagnosis of central or peripheral type of Tapia's syndrome and also the nature of the lesion, ischemia, abscess formation, tumor or haemorrhage.

Tapia's syndrome classification and a treatment protocol have been proposed by Aktas and Boğa[32]: Grade Ⅰ/mild type, unilateral cord and tongue paralysis, no uvula distortion, minimal slowdown in speaking, no swelling in tongue and no trouble in swallowing, Corticosteroid treatment is not recommended; GradeⅡ/moderate type, unilateral cord and tongue paralysis, no uvula distortion, mild slowdown in speaking, swelling in tongue, dryness in pharynx, trouble in swallowing, cracked speech and normal feeding and drinking, 15 d of corticosteroid treatment is recommended; Grade Ⅲ/severe type, unilateral cord and tongue paralysis, significant uvula distortion, significant difficulty in speaking, swelling in tongue, dryness in pharynx, trouble in swallowing and difficulties in feeding and drinking, endovenous corticosteroid is recommended for 1 wk.

To our knowledge, only six cases[8,22,24,36,42,43] of isolated bilateral Tapia's syndrome have been reported in the literature and all of them were related to transoral intubation during general anaesthesia. The most relevant common feature was the prolonged oro-tracheal intubation for more than 14 d in all the cases except one[36]. Our patient was reintubated three times, two of them as an urgent procedure, and remained ventilated for more than 18 d.

The majority of all reported cases, even unilateral or bilateral, recovered in 4-6 mo and this progressive recovery of function suggests nerve damage of a neuropraxic type, which is typical of compression injury. But there are some reports in the literature regarding its irreversible form[43,44,46,47] or partially reversible form[16,19,22,24,29,30,39,42,48,51].

Apart from corticosteroids and anti-inflammatory therapy described above as key of the therapy, other support treatments recommended are speech and swallow therapy and warm air inhalation. Most studies

have emphasized that the recovery is usually completed within 6 mo, but with an intensive and multidisciplinary approach the patients' recovery time could be reduced. In our case, despite no corticosteroids were administered, the recovery was complete four months post-transplant after intensive speech and swallow training.

In conclusion, Tapia's syndrome is mainly a rare complication of airway manipulation. It can occur after any type of surgery under endotracheal general anesthesia. Clinicians should be aware of its preventive strategies, diagnosis, treatment and almost always transient outcomes. Although bilateral Tapia's syndrome after general anaesthesia is exceptionally rare, this complication should be recognized in patients reporting respiratory obstruction with complete dysphagia and dysarthria after extubation. Special attention should be paid to correct positioning of the head during surgery to avoid such problems.

COMMENTS

Background

Tapia's syndrome is an extracraneal ipsilateral palsy of the recurrent laryngeal and the hypoglossal nerves. It is a very rare complication with few cases reported in the literature. The predisposing factors are most commonly orotracheal intubation for general anesthesia but also other etiologies.

Research frontiers

This study tries to collect all articles published to date, emphasizing the common aspects of all reported cases.

Innovations and breakthroughs

The rarity in the presentation of Tapia's syndrome makes its incidence probably underestimated if clinicians are not aware of its symptoms. The publication of this review will help the scientific community to keep in mind Tapia's syndrome and to establish common guidelines for diagnosis, management and treatment.

Peer-review

This is a very interesting case report and a good literature review about the topic.

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