



Hoarseness following heart valve replacement under general anesthesia: Bilateral tapia's syndrome as a rare cause

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Keywords

Tapia's Syndrome; Hoarseness; Hypoglossal Nerve; Recurrent Laryngeal Nerve; Intubation

Tapia's syndrome (TS) is a rare condition that develops as a result of concomitant ipsilateral extracranial lesion of hypoglossal nerve and recurrent laryngeal branch of vagus nerve.^{1,2} Dysphonia and dysphagia are common clinical findings in patients.^{1,2} It can be seen after any surgical operation performed under general anesthesia with orotracheal intubation.² It is frequently seen as a unilateral case and bilateral TS cases have been rarely observed. In this report, it is aimed to present a case of bilateral TS that developed in a patient who underwent cardiac surgery under general anesthesia.

A 51-year-old man was admitted to emergency department with high fever and confusion. He had diabetes mellitus (DM) and hypertension (HTN) and was taking oral antidiabetic and antihypertensive treatment. In his neurological

examination, he was alert, oriented, had no motor or sensory deficits, and cranial nerve and cerebellar examinations were evaluated as normal. He had high body temperature which was 38 °C. Leucocyte count was $18.22 \times 10^3/\mu\text{l}$, C-reactive protein (CRP) was 153.9 mg/l, and other blood parameters were normal. In diffusion magnetic resonance imaging (MRI), multiple milimetric acute infarcts were shown in the left frontoparietal subcortical and periventricular white matter. Transthoracic and transesophageal echocardiography was performed. Vegetation attached to aortic valve and mitral anterior leaflet root and periaortic abscess were detected. After evaluation of cardiology, cardiovascular surgery, neurology, and infectious diseases departments, the patient underwent surgery under general anesthesia.

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Vegetation resection and valve annuloplasty were done. Uneventful orotracheal intubation was performed for the surgery. After surgery, he was transferred to the intensive care unit (ICU) and underwent mechanical ventilation. On the second day postoperatively, he was extubated. After a few hours from the extubation, he was consulted to neurology department due to dysphagia and hoarseness. In neurological examination, he was alert, oriented, had no motor or sensory deficits, and cerebellar examination were evaluated as normal. He had dysphonia and an absent gag reflex. No voluntary movements of the tongue were observed and no atrophy or deviation was found. Other cranial nerve examinations were normal. Brain computed tomography (CT) was evaluated as normal. Because of the presence of the cardiac pacemaker placed, brain MRI could not be performed. He was consulted to otolaryngology-head and neck surgery department. In the flexible laryngoscopy, vocal cords were fixed in the paramedian line and diffuse mucus accumulation was observed in the bilateral rosenmular fossa.

According to the findings of the isolated cranial nerve involvements, he was evaluated as bilateral TS and respiratory support with nasal oxygen and intravenous (IV) methylprednisolone 1 mg/kg/day was started. The patient received nutrition through a nasogastric tube. No improvement was observed in the control neurological or laryngeal examination one week later. The patient developed hemodynamic instability and desaturation in the follow-up and died on the 20th day of hospitalization.

It is important to distinguish TS from many other diseases that include multiple cranial nerve involvements, vocal cord paralysis, dysarthria, and paralysis in the tongue muscles.¹ It can be developed as a result of tumours, radiation therapy, meningitis, infectious mononucleosis, and carotid artery dissection.³ In addition, different surgical operations under general anesthesia and orotracheal intubation can cause TS.²

During surgery, the suggested mechanisms of the TS are the neuropraxia due to the stretching and pressure following the intubation or airway managements and the exaggerated hyperextension or lateral flexion position of the neck during the procedure or operation.² Therefore, careful attention should be paid for positioning and airway managements in order to prevent the occurrence of this syndrome. The diagnosis of the syndrome is based on clinical signs and is confirmed by demonstrating the vocal cord paralysis in flexible laryngoscopy in the patient.^{1,2}

Unilateral TS is generally reported and bilateral involvements are rare in literature.¹ TS has been presented only in a few rare reports after cardiac surgeries.³ Sotiriou et al.³ and Nalladaru et al.⁴ reported it after coronary artery bypass grafting (CABG) surgery and Rotondo et al.⁵ reported after aortic valve replacement surgery. These reported cases were unilateral and only Kim et al. presented a case similar to ours with bilateral TS after a cardiac operation.⁶

Treatment for TS is mainly supportive. Steroid treatment, speech/language therapies, and swallowing rehabilitation can be applied. However, TS cannot be always reversible and it can cause morbidity in patients. Especially, the bilateral cases can be more severe and can cause permanent paresis.¹

Bilateral TS is a rare underdiagnosed disease that can be seen in perioperative period. We believe that it is important especially for the otolaryngologists, neurologists, and anesthesiologists to distinguish TS from other diseases and implement a multidisciplinary approach for an early accurate diagnosis and appropriate treatment.

Conflict of Interests

The authors declare no conflict of interest in this study.

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References

1. Brotis AG, Hajjiannou J, Tzerefos C, Korais C, Dardiotis E, Fountas KN, et al. Bilateral Tapia's syndrome secondary to cervical spine injury: A case report and literature review. *Br J Neurosurg* 2023; 37(4): 745-9.
2. Cariati P, Cabello A, Galvez PP, Sanchez LD, Garcia MB. Tapia's syndrome: pathogenetic mechanisms, diagnostic management, and proper treatment: A case series. *J Med Case Rep* 2016; 10: 23.
3. Rotondo F, De PS, Modoni A, Schiavello R. Peripheral Tapia's syndrome after cardiac surgery. *Eur J Anaesthesiol* 2010; 27(6): 575-6.
4. Nalladaru Z, Wessels A, DuPreez L. Tapia's syndrome-a rare complication following cardiac surgery. *Interact Cardiovasc Thorac Surg* 2012; 14(1): 131-2.
5. Sotiriou K, Balanika M, Anagnostopoulou S, Gomatos C, Karakitsos D, Saranteas T. Postoperative airway obstruction due to Tapia's syndrome after coronary bypass grafting surgery. *Eur J Anaesthesiol* 2007; 24(4): 378-9.
6. Kim SW, Kim MS, Choi JH, Kim CD. A case of bilateral peripheral Tapia's syndrome subsequent to coronary artery bypass graft. *Korean Journal of Otorhinolaryngology-Head and Neck Surgery* 2013; 56(8): 535-7.