

A case report of Tapia's syndrome after mastectomy and breast reconstruction under general anesthesia

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Abstract

Rationale: Tapia's syndrome is a rare and potentially anesthesia-related complication that may cause considerable distress to the patient. Here we describe a case of unilateral Tapia's syndrome in a patient undergoing a skin sparing mastectomy and immediate breast reconstruction which, to the best of our knowledge, has not been reported in the literature.

Patient concerns: A 41-years old female underwent right skin sparing total mastectomy and breast reconstruction with latissimus dorsi flap under general anesthesia. On the first postoperative day, she complained left sided tongue deviation, subtle hoarseness and swallowing difficulty.

Diagnosis: Tapia's syndrome, a combined paralysis of ipsilateral vocal cord and tongue due to injury to the hypoglossal and recurrent laryngeal nerves, in this case, resulting potentially from head and neck position changes

Interventions: The patient was closely observed with the administration of empirical prednisolone 5 mg/day for 3 weeks.

Outcomes: One month after the surgery, functions of the tongue and vocal cord were completely resolved.

Lessons: Particular attention should be paid to the maintenance of adequate cuff pressure, proper position of endotracheal tube and correct neck positioning, especially when procedures taking a long operation time under endotracheal anesthesia and requiring frequent position changes of the patient's head and neck.

Abbreviations: LD = latissimus dorsi, MRI = magnetic resonance imaging.

Keywords: breast reconstruction, position, Tapia's syndrome, unilateral

1. Introduction

Tapia's syndrome is a rare and potentially anesthesia-related complication that may cause considerable distress to the patient.^[1] It was firstly reported by the Spanish otorhinolaryngologist Tapia AG in 1904, characterized by combined paralysis of the recurrent laryngeal branch of the vagus nerve and the hypoglossal nerve.^[2] The causes of this syndrome are either central (due to an intramedullary lesion of the nucleus ambiguus,

the nucleus of the hypoglossal nerve, and the pyramidal tract) or peripheral (extracranial involvement of the hypoglossal and recurrent laryngeal nerves).^[3] Tapia's syndrome after general anesthesia with endotracheal intubation has been related with extracranial compression or stretching neuropathy of both nerves at the base of tongue.^[1,4,5] Patients complain mainly tongue deviation, hoarseness, aspiration, dysarthria, dysphonia, and swallowing difficulty, and ipsilateral paralysis of the vocal cord and tongue is observed.^[6]

Here, we describe one case of unilateral Tapia's syndrome in a patient undergoing a skin sparing mastectomy and immediate breast reconstruction, which, to the best of our knowledge, has not been reported in the literature. This report aims to draw plastic surgeons' and anesthesiologists' attention to this unanticipated complication. We discussed the possible association of this rare complication with mastectomy and immediate breast reconstruction under general anesthesia, highlighted long operation time, head positioning, and several changes of position during the general anesthesia as risk factors, and summarized the potential preventive measures.^[1]

2. Case report

A 41-year-old woman whose height and weight were 160 cm and 48 kg underwent right skin sparing total mastectomy and breast reconstruction with latissimus dorsi (LD) flap due to breast cancer. Preoperative physical examination showed good neck extension and mouth opening with Cormack-Lehane Grade 1. Anesthesia was induced using a combination with propofol 2 mg/kg and remifentanyl 0.5 µg/kg. Muscle paralysis was obtained

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with rocuronium 50 mg. Administration of general anesthesia and orotracheal intubation preceded without difficulty. Laryngoscopy was carried out using a Macintosh blade 3 and a 7.0-mm reinforced endotracheal tube (Mallinckrodt) was inserted successfully on the first trial. The endotracheal tube cuff was inflated with air, until no audible leak was heard. Anesthesia was maintained with desflurane and 50% oxygen in the air. Remifentanyl was infused continuously 0.25 µg/kg/min. The patient was maintained hemodynamically stable and well oxygenated throughout the surgery.

The skin sparing total mastectomy was performed with the patient in the supine position. Her head was kept in a neutral position using a donut-type pillow throughout the total mastectomy procedure. After the procedure, she was turned from the supine to the left lateral decubitus. During the process of harvesting the LD flap, the waist on the contralateral side was stabilized with rolls to either side. After harvesting the LD flap, the patient was turned back to the supine position. The flap was rotated on the thoracodorsal neurovascular pedicle, and then breast molding and reconstruction with the LD flap were carried out. During breast molding and reconstruction with the LD flap, upright seated position was required for size and contour adjustment. The patient was placed in a seated position with the head of the bed 80° upright from the horizontal plane. The head was secured with the tape to prevent undue torsion on neck. Total anesthetic time was 395 minutes and total operation time was 430 minutes. At the end of the surgery, the patient was extubated after fully recovering of respiration and consciousness.

On the first postoperative day, she complained left sided tongue deviation, subtle hoarseness, and swallowing difficulty (Fig. 1). Immobility of true left vocal cord without edema or bruising was detected during flexible laryngoscopy. Otherwise, a complete neurological evaluation showed normal results includ-

ing the pharynx and soft palate. Brain magnetic resonance imaging (MRI) showed no other evidence of central involvement. The patient empirically received 5 mg prednisolone daily for 3 weeks. One month after the surgery, functions of the tongue and vocal cord were completely resolved.

3. Discussion

General anesthesia-related Tapia's syndrome can be understood as a localized compression lesion at the crossing of the recurrent laryngeal and hypoglossal nerves.^[1,4,5] Therefore, possible mechanisms of injury include direct trauma from laryngoscopy or from continuous pressure of inflated cuff of the endotracheal tube within pharynx or indirect trauma from prolonged stretch of these nerves after hyperflexion of the head.^[1]

The hypoglossal nerve, cranial nerve XII, leaves the skull through the hypoglossal canal and passes just above the hyoid bone.^[7] Then, it crosses the vagal nerve in order to continue its route medially and passes anteriorly, deep to the posterior belly of the digastric muscle at the mandibular angle and finally reaches the tongue innervating all the extrinsic muscle of the tongue.^[8] Concomitant injuries of the hypoglossal and vagal nerves can be caused by their close proximity in the pharynx.^[8]

Neck hyperextension during intubation stretches the hypoglossal nerve on the anterior aspect of the C1 transverse process. Furthermore, aggressive laryngoscopy for anterior displacement of tongue by blade at the base of the tongue can cause damage the hypoglossal nerve via soft tissue compression against the hyoid bone. It is likely that even trivial position changes after airway securement, including during surgical preparation and draping, could affect hypoglossal nerve injury.^[7,8] It was found that the tip of endotracheal tube cuff can move from 3.8 to 6.4 cm with neck flexion or extension, potentially injuring the subglottis. This type of trauma would likely bring about recurrent laryngeal nerve injury.^[9]

Excessive neck flexion for surgical exposure can cause the endotracheal tube to compress the lateral wall of oro- and hypopharynx where 2 neighboring nerves are. That is, as the neck is more flexed, the endotracheal tube in the pharynx may more compress the pharyngeal wall. The endotracheal tube is fixed with tape at the patient's mouth with ballooning, but inner part of endotracheal tube may slide freely in the pharynx. Moreover, aggressive neck flexion makes the ramus of mandible move toward to cervical spine and these 2 nerves. In this circumstance, the hypoglossal and recurrent laryngeal nerves are entrapped between endotracheal tube, ramus of mandible, and cervical spine. As a flexed neck posture sustains, 2 nerves may facilitate damage each other.^[8,10]

In our case, the underlying mechanism of the hypoglossal and recurrent laryngeal nerve injury is not evident. Intubation was uneventful, the cuff pressure was low, and no nitrous oxide was used. However, we speculate that the possible explanations include compression at the crossing point of the vagal and hypoglossal nerves or stretching of these nerves by inadvertently excessive extension and lateral flexion of the neck at some point during the lateral decubitus or anterior flexion during the upright position, in conjunction with endotracheal tube malposition.

There have been several reports on Tapia syndrome associated with oro-tracheal intubation resulted from compression of the recurrent laryngeal and hypoglossal nerves by the cuff pressure of the endotracheal tube and prolonged stretching of these nerves by improper positioning of the head^[8,11] in a variety of clinical conditions such as tonsillectomy, septoplasty,^[12] shoulder



Figure 1. Left side deviation of the tongue.

surgery,^[13] mediastinal tumor, coronary bypass,^[6] carotid artery aneurysm,^[14] neurofibroma of vagal, and hypoglossal nerve.^[15] From our search of the literature, we believe this is the first report of Tapia's syndrome following breast cancer surgery. Mastectomy and immediate breast reconstruction surgery generally takes a long time and requires various position changes such as supine, lateral decubitus, and upright position which potentially might cause nerve damage. Prolonged anesthesia may also aggravate the influence of the aforementioned manipulations. To avoid unnecessary injury of the recurrent laryngeal and hypoglossal nerves, the following recommendations are proposed. First, vigilant monitoring is needed to maintain adequate cuff pressure and proper position of endotracheal tube. Second, attention should be paid to correct neck positioning in avoidance of excessive prolonged hyperflexion. Third, 2 recommendations above are especially important when procedures, taking a long operation time under endotracheal anesthesia with frequent changes of the position of the patient's neck, are performed.

Author contributions

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