

Case Report: Tapia's Syndrome after Prolonged and Repeated Intubations

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Abstract:

Tapia's syndrome is the extra cranial involvement of cranial nerves X and XII, causing often reversible ipsilateral paralysis of the vocal cords and tongue. In the majority of reported cases, it is a rare complication of anaesthetic airway management and head positioning during the surgery. We describe a patient with post-operative unilateral Tapia's syndrome after multiple general anaesthetics with uncomplicated prolonged endotracheal intubation.

Introduction

Tapia's syndrome is a well-defined condition first described in 1905 following an injury to a bullfighter gored in the neck, injuring the extracranial parts of the hypoglossal nerve and the recurrent laryngeal branch of the vagal nerve^[1]. The nerves involved can be affected in varying degrees, producing a range of symptoms including dysphagia, dysarthria, hoarseness and focal symptoms mimicking an intracranial pathology^[2].

Patient information

MR is a 52-year-old Caucasian male who underwent an elective open abdominal aortic aneurysm (AAA) repair, complicated by rhabdomyolysis, acute kidney injury and Tapia's syndrome, requiring a 13-week hospital admission and further rehabilitation before returning home.

Clinical findings

The abdominal aortic aneurysm repair was performed under general anaesthesia which was induced using Fentanyl and Propofol. Muscle paralysis was obtained with Rocuronium. Direct laryngoscope visualised an airway of IIb. An easy and uncomplicated trans oral intubation was performed with the aid of routine video laryngoscope, Macintosh blade D and a size 8mm diameter tracheal tube inserted on the first attempt. The cuff of the tube was inflated with a pressure of < 20cm H₂O with no adjustments to cuff volume made intra-operatively. General anaesthesia was maintained with Sevoflurane and total operative time was 3 hours and 15minutes.

Post operatively, the patient was kept intubated and transferred to ICU due to periods of persistent hypoxia intraoperatively. A chest X-ray found the patient to have atelectasis in both lung bases, which we postulate to be the cause of intraoperative hypoxia. In the post-operative period, the patient developed

an ischemic right lower limb due to a thrombosed right femoral aneurysm. This was repaired and fasciotomies were performed in the calf. This resulted in an acute kidney injury requiring continuous venovenous haemofiltration and remained intubated for a further 7 days. His recovery period was further complicated by hospital acquired pneumonia requiring IV antibiotics, devitalisation of a left leg wound muscles requiring surgical debridement on three separate occasions under general anaesthetics.

Day 15 post operatively, the ICU team observed MR's slurred speech, poor swallow and inability to protrude his tongue. Formal assessment was performed by a neurologist with the following neurological deficits: dysarthria with a hoarse voice, inability to protrude his tongue, absent gag reflex on the right and reduced gag reflex on the left and absent soft palate and lingular actions. All other cranial nerves were examined and deemed intact. A non-contrast CT head was performed to exclude intracranial pathology and brainstem involvement. As he remained a high aspiration risk with copious amounts of excretion, he required a tracheostomy for airway protection and a PEG tube for feeding.

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Therapeutic intervention

Conservative management was established, encompassing rigorous speech and swallowing rehabilitation, chest physiotherapy and PEG feeding. Day 25 into his admission, symptoms of dysphonia and dysarthria improved, the amount of secretions were noted to have reduced significantly and the patient was able to start tolerating periods of having the tracheostomy cuff down.

Follow-up and outcomes

After 1.5 months in hospital, marked improvement was noticed in the patient's speech, swallow and movement of the tongue. A week later saw further improvement in his symptoms, PEG feeding was ceased, the tracheostomy removed and the patient commenced on an oral diet. Daily inpatient speech and swallowing therapy was performed at a rehabilitation facility for a further two weeks before the patient was discharged home.

Follow up at 3 months showed marked improvement in the patient's symptoms and at 6 months, complete resolution of speech and swallow symptoms with residual subjective weakness in tongue protrusion.

Patient perspective

Through the large majority of his admission, MR experienced significant frustration. The multi-disciplinary approach to his management included physiotherapy, dietetics, social work, occupational therapy and pastoral care, all of which aided in his wellbeing and transition back into the community.

Discussion

Tapia's syndrome is a relatively rare condition, with just over 70 cases being reported since the first reported case in 1905^[1]. The signs include ipsilateral tongue atrophy, paralysis of the vocal cord, tongue, soft palate and pharynx, although the latter two being supplied by the cranial aspect of the XII nerve through the vagus is often spared^[3]. The reported cases are attributed to a few identifiable causes: direct trauma to the X and XII nerves, neoplasms, carotid artery dissection involving the ascending pharyngeal artery, nasopharyngeal fungal infection, neurofibromatosis affecting the X and XI nerves and as a result of injury from endotracheal intubation. The majority of reported cases have been unilateral, with only 6 reported cases of bilateral Tapia's Syndrome, all of which have been attributed to trans oral intubation in general anaesthesia.

Anatomically, cranial nerves X and XII course in close proximity at the base of the tongue and in the piriform fossa and are supplied exclusively by the ascending pharyngeal branch of the external carotid artery^[4]. The hypoglossal nerve crosses the vagus nerve whilst lying on the lateral most prominence of the anterior surface of the transverse process of the first cervical vertebrae. The mechanism of injury to these cranial nerves as a result of anaesthesia is postulated to be due to (i) the intubation tube and associated cuff and motion of the patients head intraoperatively leading to likely compression of the pharyngeal wall and its underlying neurovascular structures, (ii) prolonged stretch of these nerves following excessive head anterior posterior and lateral flexion and (iii) direct trauma to the carotid artery in particular involving its ascending pharyngeal artery branch^[5-7].

Simultaneous injury of the vagus and hypoglossal nerves may be explained by their close vicinity. A recurrent laryngeal nerve paralysis may be the result of localised pressure from the tracheal tube and its cuff on the anterior branch of the inferior laryngeal nerve against the posteromedial part of the thyroid cartilage. Concurrently, the hypoglossal nerve may be damaged due to excessive stretch against the greater horn of the hyoid bone by an oro-tracheal tube or compression of the posterior part of the laryngoscope^[8,9].

Diagnosis of Tapia's Syndrome is made based on thorough history and examination and exclusion of intracranial pathology with computed tomography or magnetic resonance imaging of the brain. Vocal cord paralysis can be further evaluated via laryngoscope or telescope can be used to differentiate paralysis from paresis^[10]. Damage to these nerves are most probably neuropraxic and is reversible in the majority of cases^[11].

The treatment is usually supportive and involves a multidisciplinary approach. This encompasses extensive speech and swallow rehabilitation, chest physiotherapy and warm air inhalation. In some case series, a short course of systemic corticosteroid has been used depending on the extent and acuity of the nerve damage^[12].

The cause in our case is most likely due to direct compression of the X and XII nerves as a result of prolonged and repeated intubations. CT and MRI were performed to rule out intracranial pathology. The need for multiple prolonged oropharyngeal intubations as well as positioning of the patient's head may have been the source of unilateral nerve compression. The progressive recovery of the patient's symptoms with conservative treatment is also high suggestive of a compression injury. It is unlikely that these symptoms were due to or worsened by insertion of the tracheostomy which is typically carried out in the midline, in the 3rd or 4th tracheal ring. Anatomically, the recurrent laryngeal nerve is the only likely nerve to be damaged as a complication of a tracheostomy.

In carrying out routine oro-tracheal intubation, special attention needs to be paid to preventive measures including:

- (i) Careful positioning of the head and neck during intubation, intra-operatively and during extubation
- (ii) Gentle intubation, avoiding excessive pressure with laryngoscope
- (iii) Proper cuff inflation avoiding excessive cuff pressure
- (iv) Avoiding excessive movements of inflated cuff and extubation with inflated cuff^[13-15].

Conclusion

In conclusion, despite Tapia's syndrome being a rare occurrence, both surgeons and anaesthesiologist should be made aware of it as a possible complication of surgery, especially ones with a prolonged intraoperative time and when the patient's head is in the flexed position.

Informed consent: Written consent was obtained from the patient for discussion and publication of this case report.

Author Contribution: Dr Rebekah Tan: Literature review, col-

lation of information and composition of case report. Dr Kishore Sieunarine: Primary surgeon, proof reading of case report.

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