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Quadraplegia: A Rare Complication Associated with Cricotracheal and Tracheal Resection

Research Article

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Abstract

Introduction: Cricotracheal resections and tracheal resections are highly successful procedures for the treatment of severe airway pathology including high-grade stenoses. The major complication of resection is anastomotic dehiscence and various methods have been developed to decrease this complication including cervical flexion, the guardian suture, braces, cervical and intrathoracic releasing procedures. We have recently become aware of a potential association between quadriplegia and cricotracheal resections and tracheal resections.

Objectives: To identify cases of quadriplegia associated with cricotracheal resections and/or tracheal resections and to identify commonalities.

Methods: After a case of quadriplegia was identified, a thorough review of the medical literature was performed including search for quadriplegia, tetraplegia, paralysis and cricotracheal resection, tracheal resection, Grillo stitch, cervical flexion, subglottic stenosis, tracheal stenosis. All references within these papers were similarly examined.

Results: Six case reports of quadriplegia associated with airway surgery were in obscure medical literature. We present a summary of patient demographics, etiology, and underlying pathology, type of surgery, risk factors, management and outcomes. Additional cases that have not been reported were identified in discussions with airway surgeons. Spinal cord dysfunction can be reversible but is often unpredictable.

Conclusions: Quadriplegia following CTR or TR is an extremely rare but devastating complication of unknown pathogenesis. Cervical flexion is a common but not universal finding. Potential interventions are discussed.

Keywords: Cricotracheal resection; Tracheal resection; Subglottic stenosis; Tracheal stenosis; Airway reconstruction; Complication; Tetraplegia; Quadriplegia; Paralysis; Paraplegia; Neck brace; Chin-to-chest suture; Cervical flexion; Cervical flexion myelopathy

Introduction

Cricotracheal (CTR) or tracheal (TR) resection is a successful procedure for treatment of high grade subglottic (SGS) or tracheal stenosis (TS) in carefully selected patients. The success rate has been reported to be greater than 90% [1]. The overall surgical complication is reported to be around 16.7-18.3% [2,3]. This is broadly classified

into anastomotic complications (anastomotic separation, fistulation into surrounding structures, granulation tissue formation, re-stenosis); and non-anastomotic complications (haemorrhage, wound infection, laryngeal edema, vocal cord dysfunction, aspiration, hypoxemia, subcutaneous emphysema and dysphagia) [1,2,4,5]. The overall morbidity is 17-45%, and mortality is 0-2.4% [1].

The largest series to date at Massachusetts General Hospital reported anastomotic complications in 9% of their 901 patients, with 4% of these being anastomotic separation [6]. Bibas BJ et al., reported 21% anastomotic complications in their 94 patients, with dehiscence accounting for 1% [1].

Anastomotic failure whilst rare, may adversely impact on patients and surgical outcomes. Several methods have been used to minimise suture line tension to avoid this. This starts in the pre-operative setting by identification of patient, disease and surgical factors that may influence the type of surgery offered and outcome, with rigorous pre-operative patient work-up and selection Juvenile trachea does not tolerate tracheal tension well [6,7]. As such, only 30% of trachea can be resected, in contrast to 50% in adult [6,7]. The safe limit of resection is reported to be 4.5cm but the risk of anastomotic separation doubles when greater than 4cm of trachea is resected in adult patients [6]. Intra-operatively, cervical flexion, peri-tracheal dissection, supra- and infra-hyoid release, pericardial release, mobilisation of the lung hilus, and left main bronchus division have been used to reduce anastomotic tension. Post-operatively, sedation, chin-chest-sutures and or neck braces are used to maintain cervical flexion.

We report a rare, but devastating neurologic complication in an 8-year-old following CTR, attributed to cervical flexion. We also performed a literature search to identify the risk factors, management and outcome in similar cases.

Case Report

A former 25-weeker with a history of pulmonary insufficiency secondary to Broncho pulmonary dysplasia and a Cotton-Myer grade three SGS, required tracheotomy at two months old. He underwent laryngotracheal reconstruction (LTR) with anterior and posterior costal cartilage grafts at 20 months old. He subsequently underwent multiple endoscopic excision of scar tissue (three cold with mitomycin C applications, and two laser excisions with mitomycin C applications) to manage his tracheal granulation tissue. As he was unable to be decannulated and had recurrent pneumonia, he underwent a revision LTR (anterior costal cartilage graft and posterior cricoid split) at two and a half years old. He was successfully decannulated six months later as the SGS improved to a grade two with baseline stridor. He had his third LTR revision at four years old to improve his airway as he had a baseline stridor.

He continued to be symptomatic with recurrent stridor and ongoing respiratory distress secondary to a recurrent SGS and supraglottic obstruction with epiglottis prolapse. His parents refused tracheostomy due to concerns regarding the psychosocial impact of a tracheostomy tube and opted for CTR. At eight years old, he underwent CTR, following a right hilar release. There was no apparent complication or hypotension during surgery. He was kept intubated with a size 7.0 endotracheal tube (ETT), and mechanically ventilated in the pediatric intensive care unit (PICU). His cervical flexion was maintained with a chin-to-chest stitch, and a modified Minerva brace for support and to prevent lateral displacement of his head.

He had a diagnostic direct laryngoscopy and bronchoscopy on post-operative day (POD) 7. The Grillo stitch was removed and his ETT downsized to 6.5mm. The patient returned to PICU and his recovery was complicated by ventilator acquired pneumonia which was treated with gentamicin and levofloxacin. His cervical flexion was maintained by the Minerva brace while he was sedated. Extubation was postponed due to ventilator associated pneumonia.

On POD 14, his second diagnostic direct laryngoscopy and bronchoscopy findings were favorable for extubation and his ETT was further downsized to 5.5mm. Sedation was weaned in preparation for PICU bedside extubation. The Minerva brace was removed from his chest and head on POD 15. The patient was able to open his eyes and wiggle his toes, but was found to have profound weakness and flaccid paralysis of all four extremities. The ETT was upsized to 6.5mm tube to optimize ventilation as extubation was delayed.

Neurology and neurosurgery were consulted and a magnetic resonance imaging (MRI) of the brain and spine was performed. The result was concerning for ischemia given the abnormal T2 signal extending from the medulla oblongata to majority of the thoracic cord. The entire cord

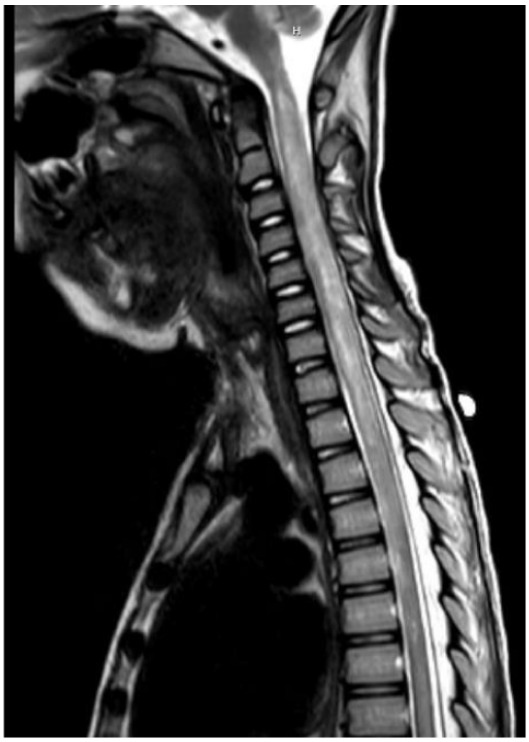

Figure 1: Sagittal T2 weighted MRI showing hypo intensity spanning from the medulla oblongata to majority of the thoracic cord, with associated expansion and edema of the cord.

appeared edematous and expanded, presumed secondary to infarction or trauma (Figure 1). The MR angiogram did not demonstrate any abnormality of the vasculature. A 5-day course of 15mg methylprednisolone was given for the inflammation and edema. Unfortunately, there was no significant improvement. His upper extremities were areflexic, and lower extremities were hyper reflexic with extensor plantar reflex. His sensory exam was inconsistent and therefore suspected to have no sensation in his extremities.

On POD 21, he underwent tracheotomy (5.0 pediatric Bivona tracheotomy tube) as he remained ventilator dependent, and gastro-jejunal tube placement for feeding. At his discharge on POD 89, he remained tetraplegic and wheelchair-bound. His facial movements were symmetrical and had full lingual mobility without issues feeding or swallowing.

Discussion

This devastatingly rare complication of CTR was first reported in 1981 [8]. In the largest series of CTR and TR to date, tetraplegia has not been identified as a non-anastomotic complication [6]. We have identified six case reports in the literature, summarised in (Table 1). Our patient is the youngest with this complication to date.

The aetiology is unknown but postulated to be multifactorial: Mechanical compression from positioning during cervical flexion; cervical spine abnormalities like neuroforaminal narrowing compressing segmental vasculatures; local or systemic haemodynamic abnormalities resulting in changes of arterial blood flow and or venous congestion. The mechanical and or vascular injury results in endothelial damage of the blood vessels [9]. The pathological changes begin in the central gray matter with thrombi formation on the exposed sub endothelial tissue, emboli into small central vessels of spinal cord, increased vascular permeability, venular distention and diapedesis of red cells, followed by areas of ischemia and haemorrhage [9]. This leads to progressively evolving necrosis of the central gray matter, followed by the white matter [9].

Hyperflexion, rather than hyperextension is believed to be more likely to cause spinal cord damage [5]. The primary fulcrum of flexion and extension occurs in the C4-6 levels [12]. Mechanism preventing pure flexion forces includes the posterior ligamentous complex, posterior nuchal muscles and impact of chin on the chest [9]. When these mechanism fails, it may lead to cervical vertebral subluxation or dislocation, bilateral interfacial dislocation, vertebral fractures, and or backward herniation of the intervertebral disc [9]. Extreme cervical flexion may produce stretch injury to the spinal cord, altering its auto-regulation by mechanically affecting the arterial supplies [10]. However, short moments of flexion have been reported to compromise the spinal cord's vascular supply: Paraplegia following sit-ups and push-ups [12], from the prone position [13]. Levy L et al., reported a temporary tetra paresis in a patient bound in extreme flexion by bandits for 12 hours, which resolved in three months [9].

Hypermobility in children may predispose them to mechanical and ischemic mechanisms resulting in juvenile flexion myelopathy by over-stretching the spinal cord, dorsal compression by dura, and or arterial ischemia secondary to venous congestion during flexion [11,12]. Insufficient dural growth relative to the spine during adolescence causes forward displacement of the dural with cervical flexion [11]. This mechanical compression of the cord against the vertebral bodies compromises the spinal microcirculation [11]. Over longer periods, larger calibre

Table 1: We have identified six case reports in the literature.

Authors	Patient Demographic	Symptoms	Pathology	Surgery	Risk Factors	Sequelae
Borrelly, 1981 [8]	48y♀	Progressive dyspnea Not reported	Tracheal adenoid cystic carcinoma (4/5) 12m later, recurrent stenosis	Hyoid & TR via sternotomy TR via thoracic approach	1) Chin-to-chest steel wire 1) Plastic neck brace to maintain cervical flexion 2) CT neck revealed cervical cord compression by large osteophyte at C5/6	Immediate hypoesthesia of lower limbs but complete recovery in 48 hours. Wires removed at day 20 post-op Lower limbs sensory deficits & left leg paralysis. Stiff bandage in place of sutures with recovery except left leg hypesthesia 2.5-year follow up
Pitz C, 1994 [4]	27y♂ Smoker	Progressive dyspnea & inspiratory stridor	Distal TS (75%) from adenoid cystic carcinoma	TR (5cm) via sternotomy with end-to-end anastomosis, laryngeal release	1) Two chin-to-chest sutures 2) "Several hours" of hypotension (70mmHg) day 1 post-op 3) MRI evidence of medullary channel stenosis secondary to spondylosis with maximal cervical flexion without signs of spinal cord compression	Day 2 post-op post extubation: Tetraplegia (below C7) but complete recovery in "a few weeks" with suture removal 2-year follow up
Dominguez J, 1996 [10, 14]	21y♀	Dyspnea & inspiratory stridor	Prolonged intubation (18d) resulting in 2cm TS (2mm thick)	TR (2^{nd}-5^{th} rings) with end-to-end anastomosis	1) Chin-to-chest suture to maintain guardian-like cervical flexion 2) Sitting position 3) Hypotension (80-90mmHg systolic)	Extubated 1h post-op, 8h post extubation developed tetraplegia (C4/5) despite suture removal and high dose methylprednisolone for 24h 4-year follow up
Silver J, 2007 [15]	17y♂	Dyspnea on exertion Inability to play clarinet	Congenital TS	TR	1) Chin-to-chest sutures 2) Small annular C6/7 prolapse on MRI	Extubated day 2 post-op & noted to be paraplegic & incontinent. Referred to national centre but persistent paraplegia 6-year follow up
Windfuhr J, 2010 [5]	18y♀	Recurrent upper airway infections with dyspnea & hospital admissions	TS	CTR	1) Chin-to-chest suture in neutral position	Extubated on day 3 post-op but re-intubated due to severe dyspnea Lower limb motor deficits found on day 5 & diagnosed with C7/8 paraplegia Treated with 5d of 500mg methylprednisolone with no improvement Wound infection required tracheostomy on day 8 & multiple surgical revisions of distorted cricoid Trach dependent with gross movement of upper limbs & paraplegia 8-year follow up
Fehre K, 2016 [11]	20y♀	Increasing dyspnea	TS	TR Post-operative surgical re-anastomosis	1) Chin-to-chest suture during surgical re-anastomosis for air leak 2) MRI showed spinal edema only	Sedated & ventilated 10 days post re-anastomosis & diagnosed with tetraplegia when extubated. Rehabilitation improved deficits to paraplegia at 3.5 months 3.5-month follow up

CTR: Cricotracheal Resection, CT: Computed Tomography, MRI: Magnetic Resonance Imaging, SGS: Subglottic Stenosis, TR: Tracheal Resection, TS: Tracheal Stenosis

vessels like posterior spinal artery and epidural veins may cause more extensive circulatory problems affecting the white matter in the dorsal half of the spinal cord [11]. Venous congestion may cause changes in the paraspinal muscles reflecting myonecrosis in the context of a flexion induced compartment syndrome [11].

However, this can also occur without any obvious risk factors: Windfuhr J reported a case of quadriplegia in an adult without any of the above risk factors [5]. The prognosis depends on the duration of the occlusion and presence of collateral circulation [4,9]. Spinal cord dysfunction may be reversible but is often unpredictable (Table 1).

Some steps to potentially decrease the risk of this complication include identification of patients with abnormal spinal anatomy and circulation in the pre-operative work up with imaging. Intra-operative utilization of extended release manoeuvres to avoid cervical flexion while achieving a tension free anastomosis may be helpful. There should be robust communication between the surgeon and anaesthetist to maintain normotension, which should be continued in the post-operative setting, along with regular neurological examination. Cervical flexion should be used judiciously and assessed regularly.

If neurological abnormalities are identified, MRI is the investigation of choice. The patient should be managed in a multi-disciplinary setting with the neurologist, rehabilitation physicians, physiotherapist and occupational therapists. It is unclear if early institution of high dose steroids improves the neurological outcome.

Conclusion

Quadriplegia following CTR or TR is an extremely rare but devastating complication of unknown pathogenesis. Identifying patient factors such as those with abnormal spinal anatomy; surgical factors to achieve a tension free anastomosis with judicious use of cervical flexion and maintaining normotension intra- and post-operatively is important to prevent it.

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