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Case Report

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The challenge of chronic ventilatory support in a laryngectomized patient with Guillian-Barré Syndrome

Abstract

Laryngectomized patients become obligate stoma-breathers, able to breathe with or without a tube, which may be either of silicone or plastic (rigid tube), particularly when ventilatory support is required. Silicone tubes are more comfortable, but only rigid tubes are suitable for mechanic ventilation. We present the case of a 57-year-old male who underwent a total laryngectomy with tracheoesophageal prosthesis placement in September 2022 as a treatment for laryngeal carcinoma. On December 31, 2022, the patient developed a demyelinating form of Guillain-Barre Syndrome, presenting with severe tetra paresis. A few days later, the patient experienced severe type 2 respiratory failure, requiring invasive ventilation. His condition improved, and during his stay in the Physical Medicine and Rehabilitation ward, he required only nocturnal ventilation through a rigid tube. During the day, a fenestrated silicone tube was used to allow phonation training. His neuromotor status improved, and after returning home, by June 2023, the patient discontinued ventilator use due to the discomfort caused by frequent tube changes. A Pulmonology assessment in July revealed normal respiratory parameters without invasive ventilation. For laryngectomized patients who require chronic ventilation, the use of a rigid tube for ventilatory support poses significant challenges. The continuous presence of the rigid tube can be uncomfortable and impair communication, while the constant insertion and removal of the tube may lead to reduced adherence to ventilatory support. The absence of a rigid tube during the day is crucial for patient comfort and speech/phonation training. Our patient's decision to discontinue ventilation at home was influenced by the challenges posed by frequent tube changes. Fortunately, he recovered from his chronic respiratory insufficiency, allowing him to discontinue nocturnal ventilation without major complications. Further research is needed to address the ventilation challenges in laryngectomized patients, aiming to improve comfort, communication skills, and adherence to necessary ventilatory support.

Keywords: total laryngectomy; chronic ventilatory support; rigid tube changes; speech training; tracheoesophageal prosthesis; rehabilitation.

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Abbreviations

TL, Total laryngectomy; PMR, Physical Medicine and Rehabilitation; PE, Pharyngoesophageal; TE, Tracheoesophageal, ICU, Intensive care unit

Introduction

Total laryngectomy (TL) remains the primary procedure for advanced-stage and recurrent laryngeal carcinoma, despite major strides in conservation laryngeal surgery and the increasing use of concurrent chemo radiation modalities.[1] Although this procedure can be curative, it is associated with significant morbidity, including a permanent alteration of the physiology of breathing, phonation and swallowing.[2] Post laryngectomy rehabilitation is crucial in these patients, and should focus on optimizing breath, speech and swallow functions, since the goals of treatment are not only to eradicate cancer but also to return patients to a life fulfilled with the greatest function and social integration possible.[1]

Laryngectomized patients are obligate stoma-breathers. They can breathe directly through the stoma, as the trachea is sewn to the skin at the base of the neck, or they can breathe through a silicone tube, which is commonly used in the first weeks after surgery for protection. When there is a need for ventilation, it can only be performed via the stoma, since there is no further communication between the lungs and upper airways.[3] The best way to effectively ventilate a patient with a laryngectomy stoma is by using a rigid tube, as it not only provides the necessary rigidity to support high pressures (unlike silicone tubes) but also features a universal connector for attachment to the ventilator and may have an inflatable cuff to prevent air leaks.[3,4] Manual ventilation, if needed, should be done using a Bag-Valve Mask with a pediatric mask over the stoma site.[5] We present a case report of a laryngectomized patient who required mechanical ventilation due to a neuromuscular disease, highlighting the challenge of tube changing in a patient who needs post-laryngectomy speech and swallow training, while also requiring ventilation to optimize respiratory function.

Case report

We report the case of a 57-year-old male, a former smoker with emphysema, who underwent a total laryngectomy in September 2022 as a treatment for an invasive squamous cell carcinoma of the larynx. During the procedure a tracheoesophageal (TE) fistula was created and a Provox 2 phonation prosthesis was placed, along with a rigid tracheostomy tube for greater protection post-surgery, with plans to later switch to a silicone tube. However, three months later, on December 31, 2022, the patient developed a demyelinating form of Guillain-Barre Syndrome, characterized by significant hyporeflexic tetra paresis following an upper respiratory infection the week before.

The patient had their vaccination plan up to date, including the COVID-19 vaccine. Upon admission, the patient's motor function test (according to Medical Research Council) revealed a wrist flexion at grade 1 bilaterally, finger flexion and extension at grade 2 bilaterally, with no other active movements in the upper limbs; hip flexion and knee extension at grade 4 bilaterally, and dorsiflexion and plantar flexion at grade 3 bilaterally. Initially admitted to the Intermediate Care Unit, the patient was transferred to the Intensive Care Unit (ICU) after 10 days due to severe dysautonomia, respiratory arrest, and severe type 2 respiratory failure requiring invasive ventilation through the tracheostomy (he was still using a rigid tube at that moment). As his condition improved, requiring only nocturnal ventilation, multiple weaning attempts of invasive ventilation were made, and by February 5, 2023, he no longer required invasive ventilation. The patient was then transferred to the Neurology ward and subsequently to the Physical Medicine and Rehabilitation (PMR) ward on February 20, 2023.

Upon admission to the PMR ward the patient was evaluated by stoma care team, who recommended switching from the rigid tube to a fenestrated silicone tube to begin phonation training via the tracheoesophageal prosthesis, and to improve patient comfort. At the same time, he was diagnosed with chronic respiratory insufficiency by the Pulmonology team, with hypercapnia and elevated bicarbonate levels observed in gasometry, which were attributed to the neuromuscular disease in a patient with a history of emphysema. Given the necessity of nocturnal invasive ventilation to mitigate chronic respiratory insufficiency, which was only possible with a rigid tube, along with the need for phonation training and patient comfort during the day using a silicone tube, it was decided that the patient would use the silicone tube during the day, and a rigid tube with a cuff at night for ventilation. After a week, gasometry showed resolved respiratory insufficiency, and the patient continued nocturnal invasive ventilation through a rigid tube. The cuff was removed after a few days, making tube changes more comfortable for the patient. Despite this, the patient remained resistant and uncomfortable with constant tube changes.

The patient was transferred to the Northern Rehabilitation Center on March 22, 2023, to continue neuromotor rehabilitation, phonation training, and to optimize nocturnal invasive ventilation. By discharge on June 10, 2023, he was able to walk with a cane and required minimal assistance with daily activities. After returning home, the patient discontinued ventilator use on his own due to the difficulties caused by frequent tube changes. In July 2023, a Pulmonology assessment showed normal gasometry and nocturnal oximetry without invasive ventilation, with normal levels of carbon dioxide and bicarbonate, and a sleep study revealed an Apnea-Hypopnea Index of 11. Respiratory function tests were attempted, but the patient wasn't able to cooperate fully. He continued phonation training with a Speech Therapist at our Hospital for another 2 months, with great success. He continued to recover his neuromotor and functional status, and by the time we are writing this article, the patient is fully autonomous in all daily activities.

Discussion

In a TL procedure, the three laryngeal portions (supraglottic, glottic and infraglottic) of the cartilaginous skeleton, the pre-epiglotic space and the laryngeal musculature are removed.[6] With the removal of the larynx, the end of the patient's proximal trachea is mobilized and brought to the skin to create a stoma (laryngectomy stoma) in the lower anterior neck. Therefore, patients who have undergone laryngectomy are obligate stoma-breathers and can only be ventilated via the stoma, since there is no further communication between the lungs and upper airways, which is crucial for breathing and also for phonation.[3] Following total laryngectomy, a neoglottis or pharyngoesophageal (PE) segment is formed, connecting the pharynx to the esophagus.[2]

Unlike a tracheostomy stoma, which serves as a bypass through the skin and soft tissues to the trachea, a laryngectomy stoma is created by bringing the trachea directly to the skin at the base of the neck. This type of stoma carries no risk of closure, allowing patients to breathe directly through it without needing a tracheostomy tube. If needed for any reason– such as for protection after the surgery until complete healing or for adaptation of a heat and moisture exchanger – silicone cannulas are usually preferred as they are smaller, flexible and more comfortable than the rigid plastic cannulas.[7,8]

Recovering of the swallow function is usually quick and straightforward because the oral cavity is directly connected to the esophagus via the PE segment, eliminating any risk of oral aspiration. The challenge in laryngectomyzed patients is that tongue base interaction with pharyngeal wall contraction must overcome the resting pressure of the closed PE segment for boluses to enter the esophagus. This change in physiology is a set up for dysphagia, causing slow bolus transit or accumulation of neopharyngeal residue and is more prevalent in patients who have already received chemo radiation therapy.[9]

Voice restoration is challenging because the vocal cords are removed and there is no connection between the lungs and the upper airways. Tracheoesophageal speech through a TE prosthesis is considered the most successful method of voice restoration currently available.[10] The prosthesis is implanted in a surgically created fistula between the trachea and the esophagus. It consists of a unidirectional valve that allows expiratory air to flow freely while preventing food and liquid from entering the tracheal segment. The passage of pulmonary air into the PE segment causes the mucosa and muscles in this area (constricting pharynx and cricopharyngeal muscles) to vibrate for voice production.[11,12] For speech to occur, the stoma must be covered to prevent air from escaping and to direct it into the PE segment. This can be accomplished manually or with a tracheostoma valve, which occludes when sufficient air pressure has accumulated.[13] Compared to healthy individuals, TE speakers have rougher voices with reduced loudness and limited range.[14] The implantation of a tracheoesphageal prosthesis for vocal rehabilitation can be performed either immediately after total laryngectomy (during the same surgical procedure) or at a later time. Delaying the procedure may be necessary for patients at higher risk of postoperative complications, such as those with severe radiation sequelae, to minimize the risk of developing a fistula.[2,13] For effective voice production, the patient must be able to overcome the pressure gradient between the trachea and the oral cavity, which can be tiring and requires training.[12]

In a patient with a total laryngectomy who requires ventilation due to chronic conditions such as obstructive pulmonary disease or neuromuscular disease, the need for a rigid tube to provide ventilatory support can present significant challenges. The patient may either keep the rigid tube in place all day, which can be very uncomfortable and significantly hinder communication, or he must insert and remove it daily to achieve more comfort and enable speech production during the day. In our patient's case, we opted to alternate between silicone and rigid tubes, allowing the patient to be more comfortable during the day and continue with speech training, which is a fundamental part

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of the rehabilitation program. At night, the patient had to switch to a rigid tube to enable necessary ventilatory support. Vocal restoration in post-laryngectomy patients is a complex, lifelong process, so initiating vocal and speech rehabilitation was crucial for this patient. However, regularly switching between silicone and rigid tubes can be very uncomfortable and may reduce adherence to ventilation. This was the case with our patient, who eventually discontinued ventilation after returning home due to the difficulties associated with changing the tubes. Fortunately, he recovered from his chronic respiratory insufficiency as he regained his neuromotor and functional status. This allowed him to discontinue nocturnal ventilation without major complications, even though it was against medical recommendations. One month after discharge, the patient showed no signs of respiratory insufficiency in all Pulmonology exams, including gasometry and nocturnal oxymetry. However, this situation could have been dramatic if the patient had not recovered as well as he did, potentially delaying the entire rehabilitation process due to a lack of adherence to ventilation. In our opinion, effective strategies and more research are needed to address the unique ventilation challenges in laryngectomized patients, aiming to improve their comfort and communication skills while ensuring they maintain adherence to necessary respiratory support.

Conclusion

In this case underscores the complexities of ventilating a laryngectomized patient, as comfortable, phonation-permitting tubes are unsuitable for invasive ventilation. This significantly impacts the quality of life for laryngectomyzed patients requiring chronic ventilation, such as those with neuromuscular diseases or obstructive pulmonary diseases, for example. The literature on invasive ventilation in laryngectomized patients is scarce, mostly limited to ICU cases and short-term ventilation, where sedation mitigates the challenge of tube exchanges, and the patients often remain with the rigid tube all day.

Acknowledgments

None.

Conflicts of interest

The authors have no conflicts of interest to declare.

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