Case report

Bronchial selective ventilation in a wide tracheocutaneous fistula

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ABSTRACT

We present the treatment and management of a wide tracheocutaneous fistula after tracheotomy correlated with excessive cuff pressure in a 36-year-old woman with cerebral palsy since infancy in which persistent type II respiratory failure required continuous ventilatory support. We discuss the surgical treatment adopted for the management of this particularly wide lesion. At the end of surgery, mechanical ventilation through a tracheal cannula was hindered by the reduced length of the residual trachea below the tracheotomy. The need to guarantee mechanical ventilation to the patient led to the implementation of a cuff securing system in the two main bronchi. We describe the approach that may be attempted under extreme conditions, when traditional ventilation methods cannot be applied for anatomical reasons.

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1. Introduction

Tracheocutaneous fistula is a complication of tracheotomy that adds a difficult and troublesome aspect to the patient’s care and may exacerbate respiratory disease. Closure of the fistula is recommended, but complications associated with fistula closure include pneumothorax and respiratory compromise. Several surgical approaches have been advocated in the literature, but in some cases, direct or flap surgical closure were not possible due to the wide dimensions of the lesions. Moreover, management of large tracheocutaneous fistulas is not well described in the otolaryngology literature. In our case, in addition to the difficulty in surgical management of the lesion, the patient had required continuous ventilatory support with mechanical ventilation and the extreme anatomical conditions and reduced length of the residual trachea led to the implementation of a particular approach to bypass this kind of problem.

2. Case report

A 36-year-old woman with cerebral palsy and severe kyphoscoliosis was admitted to our respiratory intensive care unit with severe respiratory failure secondary to pneumonia.

Twenty-four hours following admission, her respiratory condition deteriorated and orotracheal intubation was performed for invasive mechanical ventilation. XX days later, a tracheotomy was performed due to persistent type II respiratory failure requiring continuous ventilatory support.

Four weeks after tracheotomy, the patient presented a peristomal skin diastase that developed into a wide tracheocutaneous fistula, as a result of excessive cuff pressure (Fig. 1), due to difficult ventilatory support management.

The size of the fistula was approximately 4 cm in diameter, making surgical repair of the diastase impossible. We performed a tracheotomy with suturing of the distal stump of the trachea diastase to the skin and suturing of the previous tracheotomy breach. During surgery, a careful dissection of the trachea was conducted in its distal portion and the anonymous artery was protected by a muscle flap.

At the end of surgical treatment, mechanical ventilation through a tracheal cannula was hindered by the reduced length of the residual trachea below the tracheotomy (about 2 cm from the tracheal carina). Any cannula or endotracheal tube could not be secured to the trachea using the cuff.

The need to guarantee mechanical ventilation to the patient led to the implementation of a cuff securing system in the two main bronchi. Therefore, we selectively intubated the main bronchi under bronchoscopy guidance using two tubes (Portex Tracheal Tube, ID 5.5 mm; OD 7.4 mm), then inflating the cuffs at both main stem bronchi inlets (Fig. 2). A Y-shaped bridge was then added for ventilator connection to allow the same ventilation mode for both bronchial systems. Thus, we achieved an adequate minute volume to the patient, allowing us to correct the blood gas levels.
During mechanical ventilation, no significant leaks were reported and ventilatory parameters remained stable. Four days later, a bronchoscopic examination showed no evidence of alterations on both main bronchi.

The patient died forty days after surgery of sepsis.

3. Discussion

Currently, tracheotomy is largely performed on patients presenting with acute respiratory failure requiring prolonged mechanical ventilation so as to facilitate weaning, reduce the effort of breathing and curb complications due to prolonged intubation. Although the optimum timing of tracheotomy in critically ill patients with acute respiratory failure still remains controversial, the current trend is to anticipate the procedure within the first week of mechanical ventilation. The results of a Cochrane meta-analysis performed on five clinical trials showed a significant reduction of days on mechanical ventilation and of hospitalization in ICU wards with early rather than late tracheotomy, while no significant difference was reported in mortality or risk of occurrence of hospital-acquired pneumonia.

Complications of tracheotomy can be acute, related to or subsequent to the surgical procedure (haemorrhage, pneumothorax, infections, incidental decannulation), or late stage. The most frequent late complication is the formation of granulation tissue, with subsequent tracheal stenosis, which can remain asymptomatic for a long time, but which, in some cases, can lead to severe respiratory failure.

Other types of late complication are rare but life-threatening events, such as tracheoesophageal fistula and tracheo-innominate artery fistula. Therefore, the presence of a cuff with a higher critical pressure may cause ischemia and tracheal mucosal necrosis. In the case of shock or hypotension, an ischemic event may be triggered even when the tracheal cuff is inflated to lower pressures.

Several case reports have described late complications secondary to tracheotomy and prolonged mechanical ventilation, but the detection of a broad tracheocutaneous fistula located on the cuff of the tracheotomy cannula is a rare event that may lead to severe difficulties in airway management for ventilator-dependent patients.

In the literature, it has been reported that the frequency of occurrence of tracheocutaneous fistula ranges from 3.3% to 29%. The incidence of fistula formation is closely related to the time of cannulation. Kulber and Passy reported that a fistula does not develop when the duration of cannulation is less than 16 weeks, but its incidence increases to 70% when the retention period is 16 weeks or more.

The presence of a fistula increases the possibility of respiratory tract infections, including repeated aspiration and pneumonia. It also causes difficulties in phonation, coughing, cosmetic problems, and limitations to daily activities, including swimming and bathing. Therefore, surgical closure is necessary when a fistula occurs, but the management of large tracheocutaneous fistulas is not well described in the otolaryngology literature. Some authors have focused on the excision of the fistula tract with or without the use of a strap muscle or sternocleidomastoid flap. Others have proposed staged closures over a period of months to allow secondary healing to occur in order to avoid complications of dehiscence, pneumomediastinum, and infection. In the literature, it is difficult to find a report on the surgical closure of a fistula.
whose diameter is 1 cm or greater. Only Berenholz et al.\(^9\) have reported that a fistula of greater than 1 cm diameter was successfully closed using a muscular flap after a fistulectomy, but there has not been a report on a simple and safe surgical closure of a large tra-cheocutaneous fistula greater than 1 cm diameter.\(^9\)

In our case report, the diameter of the fistula was approximately 4 cm and its size prevented any possibility of surgical repair or resection of the lesion. In fact a small tracheocutaneous fistula may generally be sutured after fistulectomy or the fistula can be closed using a hinged flap or a bipedicle flap, but these techniques were impossible in our case.

Another key feature of this clinical case involved the difficult management of the airways to allow adequate mechanical ventilation. The opening of a tracheocutaneous fistula in the above described location required a tracheotomy very proximal to the tracheal carina.

Since the insertion of a cuffed cannula in the trachea was hindered by the reduced size of the residual trachea, a variation on the lung isolation technique was attempted, usually performed on patients undergoing thoracic surgery.\(^11\) In tracheotomised patients, the use of a double-lumen endotracheal tube to achieve lung isolation is at risk of malposition due to the reduced tracheal length and to the excessive length of the tube itself.

The Univent Tracheoport (W.Ruesch AG, Kernen, Germany) is a double-lumen tracheal tube specifically designed for lung isolation in tracheotomised patients, having limited diffusion.\(^12\) An alternative approach involving the use of a single-lumen tracheal tube with an enclosed bronchial blocking device positioned using a bronchoscope, provides the advantage of a smaller sized tube compared to a double-lumen tracheal tube.

The Univent tube is a single-lumen tube associated with a bronchial blocker presenting, when positioned, a small internal lumen through which continuous positive airway pressure (CPAP) can be applied during ventilation of the contralateral lung.\(^13\) In tracheotomised patients requiring one-lung ventilation, an independent bronchial blocker (Arndt blocker or Cohen blocker) can also be introduced through a tracheotomy cannula.

In our case, the possibility of anchoring the tube in at least one of the main bronchi had to be considered. The approach, based on the application of a cuffed left double-lumen tracheal tube in the homolateral main bronchus, was not viable due to the dimensions of the residual trachea. Therefore, both bronchi were intubated with two single-lumen tubes of adequate size.

Since separate lung ventilation was not necessary in our case, a Y-shaped connector was positioned at the distal end of the tubes, and then connected to the mechanical ventilator. This allowed us to apply the same ventilation mode for both lungs with a homogeneous distribution of tidal volume.

Ventilation was continued for several days without any leaks, maintaining stable ventilatory settings and without complications. After extubation, bronchoscopy was performed showing no lesions in either bronchial hemisystem.

The above described selective intubation of the bronchial hemisystems can certainly be considered to be a procedure involving risk of complications such as possible malposition of endotracheal tubes and bronchial rupture, although such complications are also described with double-lumen tracheal tubes.\(^14\) However, under extreme conditions, when traditional ventilation methods cannot be applied for anatomical reasons, the above described approach may be attempted, especially when a short period of mechanical ventilation is required.

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