Acquired Paediatric Vocal Cord Paralysis Secondary to Cardiothoracic Surgery

Kwong-Yew Leong, Bee-See Goh

ABSTRACT

Objective: A retrospective study conducted on paediatric cases diagnosed with iatrogenic vocal cord paralysis in University Kebangsaan Malaysia Medical Centre ('UKMMC')

Design: A case series of five patients diagnosed in UKMMC

Material and Method: Retrospective case series of patients diagnosed with vocal cord paralysis secondary to cardiothoracic surgery between the years 2011 to 2016. Cases were retrieved from the database of the UKMMC paediatric otorhinolaryngology clinic. Five cases were selected, reviewed and discussed.

Results: Out of the five cases of vocal cord palsy, four patients underwent patent ductus arteriosus ('PDA') clipping and one patient underwent a procedure for transposition of great arteries ('TGA'). Of the patients who underwent PDA clipping, the recurrent laryngeal nerve was identified intraoperatively in all but one case. The mean duration of Ryle's tube feeding was 3.4 months (range: 1 to 12 months). Four patients had left sided vocal cord palsy and one patient had right vocal cord palsy. Three patients had full compensation and two had partial compensation with phonatory gap.

Conclusion: Vocal cord paralysis is a known complication following cardiothoracic surgery. Routine flexible nasopharyngolaryngoscopy ('FNPLS') to ascertain vocal cord function should be conducted for all paediatric patients post-cardiothoracic surgery as many cases may be underdiagnosed. The early identification of vocal cord paralysis may help to reduce the risk of the onset of aspiration pneumonia or hyperactive airway [disease].

KEY WORDS

vocal cord, paralysis, paediatric, cardiothoracic, recurrent laryngeal nerve

INTRODUCTION

Vocal cord paralysis ('VCP') is the absence of movement of one or both vocal folds due to the disruption of motor nerve supply to the larynx1,3,6). Holinger et al states that bilateral VCP in infants is primarily caused by congenital disorder or secondary congenital anomalies of the cardiovascular system2).

On the other hand, unilateral VCP in infants is commonly iatrogenic followed by traumatic birth, intubation or neurological problems. Twenty percent of unilateral VCP cases are diagnosed as idiopathic1). Stridor is the most common presenting symptom and is the main reason neonates are unable to be weaned off positive airway pressure3). Patients with unilateral VCP will have poorer vocal characteristic such as hoarseness, breathiness, loudness and straining4).

METHOD

A retrospective review was conducted on all cases referred to Department of Otorhinolaryngology UKMMC between the years 2011-2016. Cases were collected from the database of the paediatrics otorhinolaryngology. Case notes were reviewed and the following facts were extracted: the onset, clinical presentation, intraoperative findings of recurrent laryngeal nerve, duration of Ryle's tube feeding and swallowing assessment.

In the given time span, 13 cases of VCP were diagnosed. One case was due to neurological problem, 5 of the cases involved children with VCP secondary to thoracic surgery, 3 cases were due to vincristine toxicity and 4 were idiopathic.

In the following case studies, we have only included cases concerning iatrogenic VCP secondary to congenital anomalies of the cardiovascular system. In all cases, VCP had been confirmed by FNPLS and patients subsequently underwent follow-up treatment for swallowing assessment and recovery.

Case 1

A 3-month old girl with underlying Down syndrome was referred for stridor post-extubation. She was diagnosed with PDA and underwent ligation at 3 months old due to heart failure. Recurrent laryngeal nerve was identified intraoperatively. On examination, the child was pink under continuous positive airway pressure (CPAP) support, and presented with symptoms of inspiratory stridor, subcostal recession ('SCR') and suprasternal recession ('SSR'). A bedside FNPLS was performed which showed left VCP. At 5 months of age, the child was examined under anaesthesia as she could not be weaned off CPAP support. Findings showed left VCP with a short aryepiglottis fold and redundant mucosa.

The child was fed via Ryle's tube for 1 year. She subsequently underwent swallowing assessment by VideoFluoroscopic Swallow Studies (VFSS) which showed that she could safely be given oral feeding. She was then allowed oral feeding supplemented with Ryle's tube feeding.
At 2 years old, the child was in a good condition of health with no aspiration symptoms albeit with stridor noted on exertion. An outpatient FNPLS was conducted which still showed left vocal fold paralysis in paramedian position. Her right vocal cord was mobile. However, a small phonatory gap had been observed. A repeated swallowing assessment with Fiberoptic Endoscopic Evaluation of Swallowing (FEES) was carried out which showed no residual or penetration with fluid or thickened feed. She was then allowed full feeding orally.

Case 2
A 6-month old boy was referred for stridor post-PDA clipping. He was born pre-term at 36 weeks and underwent surgery at the age of 2 months. Intraoperative findings did not mention if the recurrent laryngeal nerve was identified. He was discharged post-operative day 3 of surgery. His mother noticed that the child had noisy breathing while sleeping and he was brought to hospital. On examination, the child was pink under room air but had inspiratory stridor, SCR and intercostal retraction (‘ICR’). A bedside FNPLS was performed which showed left VCP and a mobile right vocal cord. He was able to tolerate oral feeding well and had no aspiration symptoms and was confirmed via VFSS at 7 months even though he had stridor on exertion. The child subsequently underwent regular follow up treatment and his symptoms gradually improved over the years. During his last follow up at 5 years of age, FNPLS findings showed left VCP with full compensation from the right vocal fold.

Case 3
A 2-month old girl was referred for weak cry post-PDA ligation. She was born pre-term at 35 weeks and underwent surgery at the age of 3 months and underwent surgery at the age of 4 months. Intraoperative findings did not mention if the recurrent laryngeal nerve was identified. She was discharged post-operative day 3 of surgery. Her mother noticed that the child had noisy breathing while sleeping and he was brought to hospital. On examination, the child was pink under room air but had inspiratory stridor, SCR and intercostal retraction (‘ICR’). A bedside FNPLS was performed which showed left VCP and a mobile right vocal cord. Clinically, the child did not have hoarseness upon crying. On examination, the child was pink with minimal SCR but with no audible stridor. A FNPLS showed left VCP with mobile right vocal cord. Clinically, the child did not have aspiration symptoms. An assessment with FEES showed no pooling or penetration. The child was kept on Ryle's tube feeding for 2 months from birth but was discharged after tolerating oral feeding well. At the age of 9 months, a FNPLS was conducted which showed full compensation by the right vocal cord. The child is currently 2 years old and is undergoing follow up treatment, with the latest FNPLS still showing left VCP with full compensation by the right vocal cord.

Case 4
A 1-month old boy was referred for aphonia post-repair of TGA. The child was born term via SVD but noted as having cyanosis on day 1 of life. He was subsequently referred to a cardiothoracic surgeon who performed a balloon atrial septostomy and an arterial switch operation on days 2 and 23 of life respectively. There was no documentation in the operative notes regarding the identification of recurrent laryngeal nerve. Post extubation, the child was noted to be aphonie when crying, with symptoms of aspiration. On examination, the child had a weak cry but no stridor. Although there was SCR, he was not in respiratory distress. A FNPLS and swallowing assessment were conducted which showed left vocal cord paresis with the right vocal cord compensating. The child was subsequently kept on Ryle's tube feeding for 1 month. At 3 months post-operative, he was able to feed well orally, and had a loud cry with no aspiration symptoms. A repeat FNPLS showed bilateral vocal cords were mobile with no gap seen.

Case 5
A 4-month old child was referred to us for aphonie post-right arch right-sided PDA ligation. The child was born pre-term at the age of 29 weeks due to maternal indication of severe pre-eclampsia. On examination, the child was comfortable and not in respiratory distress. The child was on nasal prong oxygen at a flow rate of 0.1 litre per minute. The right recurrent laryngeal nerve was identified and preserved according to intraoperative finding. A bedside FNPLS was conducted which showed right vocal cord paresis. The left vocal cord was mobile but not fully compensating. A repeated scope after 2 months did not reveal any improvement.

The child was fed via Ryle's tube for 2 months prior to a Percutaneous Endoscopic Gastrostomy (PEG) tube insertion due to poor recovery of vocal cord mobility in addition to swallowing in-coordination. The child was then transferred to another hospital for follow-up due to logistical reasons.

DISCUSSION
VCP is one of the most common causes of stridor in the paediatric age group. It is second only to the most common cause – laryngomalacia which is due to the prolapse of the supraglottic structure. Iatrogenic factors are currently the leading cause of unilateral VCP. This is due to the increased number of cardiothoracic surgeries, especially for PDA closure. This is a direct result of advancement in the paediatric field in managing premature babies with low birth weights.

In a prospective study, Pereire et al states that 11.5% of patients who underwent surgery for PDA closure suffered from complications. Further, Daya et al reports that 88% of these iatrogenic cases suffered from left VCP. In our study, four patients had left VCP. This can be explained by the anatomy of recurrent laryngeal nerve looping at the level of aortic arch. One patient had right VCP which is due to a variant of the right sided arch of PDA. Symptoms of VCP include hoarseness, stridor, a weak cry and also aspiration. In our case study, two infants had stridor, one had dysphonia and two other infants had aphony. These symptoms were the reason for referral for assessment of vocal cord mobility. However, it must be noted that many patients will be asymptomatic in unilateral VCP cases, making diagnosis more difficult. Periera et al reports that their studies indicate that a higher number of patients suffer from VCP postoperation as it was mandatory for the patient to undergo endoscopy evaluation postoperatively. In their studies, some infants did not show any signs or symptoms of VCP.

Currently, assessment has been made easier with the availability of FNPLS. This can be done at the bedside and alleviates the need for general anaesthesia. If further evaluation is needed, a direct laryngoscopy can be conducted under general anaesthesia. All of the patients in our study underwent bedside diagnostic FNPLS examination.

Table 1. Demographic data with clinical features

<table>
<thead>
<tr>
<th>Term/preterm</th>
<th>Age of cardiac surgery</th>
<th>Cardiovascular Pathology</th>
<th>Reason for referral</th>
<th>Long term Ryle tube feeding</th>
<th>Vocal cord palsy</th>
<th>Compensated</th>
<th>Recovery</th>
</tr>
</thead>
<tbody>
<tr>
<td>Term</td>
<td>3 months</td>
<td>PDA</td>
<td>Stridor</td>
<td>Yes</td>
<td>Left</td>
<td>No</td>
<td>No</td>
</tr>
<tr>
<td>Preterm</td>
<td>6 months</td>
<td>PDA</td>
<td>Stridor</td>
<td>No</td>
<td>Left</td>
<td>Yes</td>
<td>No</td>
</tr>
<tr>
<td>Preterm</td>
<td>2 months</td>
<td>PDA</td>
<td>Weak cry</td>
<td>No</td>
<td>Left</td>
<td>Yes</td>
<td>No</td>
</tr>
<tr>
<td>Term</td>
<td>1 month</td>
<td>TGA</td>
<td>Aphonia</td>
<td>No</td>
<td>Left</td>
<td>Yes</td>
<td>Yes</td>
</tr>
<tr>
<td>Preterm</td>
<td>4 months</td>
<td>PDA</td>
<td>Aphonia</td>
<td>Yes</td>
<td>Right</td>
<td>No</td>
<td>No</td>
</tr>
</tbody>
</table>

Table 2. Duration of Ryle’s tube feeding

<table>
<thead>
<tr>
<th>Case</th>
<th>Months</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>12</td>
</tr>
<tr>
<td>2</td>
<td>-</td>
</tr>
<tr>
<td>3</td>
<td>2</td>
</tr>
<tr>
<td>4</td>
<td>1</td>
</tr>
<tr>
<td>5</td>
<td>2</td>
</tr>
<tr>
<td>Total</td>
<td>17</td>
</tr>
</tbody>
</table>

Mean: 3.4
Out of the 5 cases studied, only 1 patient had full recovery of their vocal cord function. In most cases, VCP patients recover spontaneously as the cause of the paralysis is neuropaenia, a complication of traction during surgery intraoperatively. The median time taken for recovery was noted to be 6.6 months, but it can take up to 5 years. Daya et al mentioned that iatrogenic VCP has the lowest recovery rates with only 46% achieving recovery. In most cases, compensation from the contralateral vocal cord helps to prevent aspiration symptoms. This is evidenced in our case study where 3 out of 5 patients experienced compensation.

Of all the 5 patients, only 1 patient required long-term Ryle's tube feeding. One patient had PEG tube insertion while the other 3 were able to tolerate oral feeding with no symptoms of aspiration after compensation occurred. The mean duration of Ryle's tube feeding was 3.4 months. Aspiration symptoms occurred in children due to lateral position of the vocal cord with poor compensation by the contralateral cord. Ross et al concluded that if aspiration symptoms are present, a barium swallow test is recommended and that feed may need to be thickened. Malcom et al note that feeding was a problem in patients with VCP. There is a risk of developing reactive airway disease due to microaspiration. It is reported that 64% of the infants concerned required gastrostomy tube placement. Malcolm further suggests that the thickening of formula or breast milk with crushed baby cereal may reduce the risk of aspiration.

The treatment of VCP is mainly focused on establishing airways for the infant, managing aspiration symptoms and rehabilitation of the voice. Cohen et al state that unilateral VCP requires no surgical intervention as compensation by the normal vocal cord will also give adequate phonation. Whilst tracheostomy is the mainstay of treatment for patients with bilateral VCP, it is rarely done in unilateral VCP cases. Truong et al reports that 4% of their patients required tracheostomy. However, it must be noted that Truong did not state whether the patients were suffering from bilateral or unilateral VCP. Tiago et al mentions that the preferred management is vocal therapy. Sethur et al states that 80% of cases of unilateral VCP will be effectively compensated by the contralateral side. However, if there are persistent glottal aperture defects, speech therapy can be used to strengthen compensatory methods of glottic closure. This will also help in preventing aspiration symptoms.

If conservative methods fail, surgical treatment is an option that requires consideration. Intervention is aimed at controlling aspiration and also improving the quality of voice for patients while maintaining an adequate airway. Surgical treatment includes injection laryngoplasty, thyroplasty and laryngeal re-innervation. This should be considered properly since recovery from VCP in children may occur only after several years. Bhattacharyya et al reports that patients who underwent medialisation had clinical improvement in their overall swallowing function and reduction in aspiration.

As part of their rehabilitation, all patients who have VCP should be seen by a speech and swallowing therapist. These therapists will help to assess not only vocal quality and also the safety of oral feeding. The patients in UKMMC either underwent swallowing assessment with VFSS or FEES. VFSS is able to give an objective measure of swallowing that can be observed. King et al quotes a study done by Schindler et al 2008 in which it was found that speech and swallowing therapy alone resulted in improved glottic closure on endoscopy as well as subjective and objective improvement in quality of the voice.

CONCLUSION

In conclusion, vocal cord paralysis is a known result of complication for cardiothoracic surgery for infants. From our review, intraoperative findings of recurrent laryngeal nerve do not ensure the recovery or mobility of the vocal cord function. Routine flexible laryngoscope should be conducted for all patients after cardiothoracic surgery, especially post-PDA ligation, to ascertain that there is no vocal cord palsy as many cases are asymptomatic. The early identification of vocal cord paralysis may help reduce the risk of aspiration pneumonia or reactive airway disease.

REFERENCES