Paediatric Vocal Fold Paralysis

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Abstract

Objective: Vocal fold paralysis (VFP) is a relatively common cause of stridor and dysphonia in the paediatric population. This report summarises our experience with VFP in the paediatric age group.

Methods and materials: This was a retrospective review of 45 consecutive patients presenting with VFP over a 12-month period. The diagnosis was performed by flexible endoscopic examination. The cases were evaluated with respect to aetiology of the paralysis, presenting symptoms, delay in diagnosis, affected side, vocal fold position, need for surgical treatment and outcome.

Results: The presenting symptoms were stridor and dysphonia. Iatrogenic causes formed the largest group, followed by idiopathic, neurological and obstetric VFP. Unilateral paralysis was found in most cases. The median value for delay in diagnosis was 1 month and it was significantly higher in the iatrogenic group. Only eight patients (18%) underwent surgical treatment.

Conclusions: The diagnosis of VFP may be suspected based on the patient’s symptoms and confirmed by flexible endoscopy. Infants who develop stridor or dysphonia following a surgical procedure have to be examined without delay. The surgeon has to keep in mind that there is a possibility of late spontaneous recovery or compensation.

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Parálisis laringea en la población infantil

Resumen

Introducción y objetivos: La parálisis laringea es una causa relativamente frecuente de estridor y disfonía en la edad pediátrica. Este artículo describe nuestra experiencia en parálisis laringea en población pediátrica.

Métodos: Se incluyeron en el estudio todos los pacientes que acudieron a consulta con el diagnóstico de parálisis laringea durante un año completo. Se examinaron las historias clínicas de forma retrospectiva. El diagnóstico de parálisis laringea se llevó a cabo mediante exploración clínica con nasofibroscopio flexible. Se registraron los siguientes datos: etiología de la parálisis,


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Introduction

Laryngeal paralysis frequently causes stridor and dysphonia in the paediatric population. In fact, it is the second leading cause of neonatal stridor and represents 10% to 22% of all congenital laryngeal anomalies after laryngomalacia.\(^1\) Occasionally, the symptoms can be mild and cause the diagnosis to be overlooked. Moreover, exploration of the larynx is particularly difficult in paediatric patients compared to adults, and requires otolaryngologists with experience in the management of this population. The situation is complicated further when the suspected diagnosis is laryngeal paralysis, because paediatric patients, in most cases due to their young age, do not cooperate with instructions regarding phonatory and respiratory manoeuvres required for the assessment of laryngeal paralysis.

The incidence of this condition has increased in recent years, probably due to both an increase in flexible laryngoscopies performed on children by otolaryngologists and an improvement in survival of newborns with laryngeal paralysis.\(^2\)

Laryngeal paralysis can be unilateral or bilateral. Most unilateral cases affect the left vocal cord (or fold). The causative factors are numerous: iatrogenic, idiopathic, neurological diseases and obstetric factors.

Diagnosis is usually performed by endoscopic assessment using a flexible nasofibroscope. Laryngeal electromyography is the reference test to confirm the diagnosis, but is rarely employed in children. Treatment of laryngeal paralysis depends on the symptoms, mainly involvement of the airway.

Our service covers a very large population of children, therefore including a large number of patients with laryngeal paralysis. This is the reason why we decided to carry out a descriptive study of these patients.

The aim of our study was to assess the characteristics of the infant population with laryngeal paralysis and to describe the clinical characteristics of this condition in early ages, both in relation to the causes for its appearance and to exploratory findings, clinical evolution and treatment received by these patients. To the best of our knowledge, no other series on paediatric patients with this condition have been published in our country.

Methods

The work included all paediatric patients (under 14 years) who attended consultation with a diagnosis of laryngeal paralysis over a period of 1 year, both newly diagnosed cases and those who had been diagnosed previously and attended for a review.

The diagnosis of laryngeal paralysis was performed by exploration using a Machida\(^3\) 2 mm flexible nasofibroscope. All examinations were performed with the patient awake and without any form of sedation or topical anaesthesia. The examinations were recorded and reviewed by 3 separate otolaryngologists in order to establish the presence of unilateral or bilateral paralysis, the affected vocal cord (right or left) in unilateral cases and its position (midline, paramedian or lateral).

We reviewed the medical records of patients with the aim of collecting data on the aetiology of the paralysis, presentation symptoms, diagnostic delay (time from onset of symptoms until laryngoscopy was performed).

In terms of evolution, we recorded the follow-up time, recovery of mobility documented by flexible fibroscopy and the type of surgical treatment performed in cases where it was necessary.

The statistical analysis was performed using the software package SPSS\(^4\) v.11.0.1. Quantitative variables were described through frequencies, whilst qualitative variables were described through mean, median and standard deviation.

Patients were divided into 2 groups according to the aetiology of their paralysis: iatrogenic and non-iatrogenic. Differences between qualitative variables were evaluated using Fisher’s exact test or the Chi-square test, as required. For quantitative variables we performed comparisons using Student’s \(t\) test or the Mann–Whitney \(U\) test, if not normally distributed. Results were considered statistically significant when \(P<.05\).

Results

We included 45 patients in the study out of a total of 6000 children who attended consultation over 1 year, in many cases referred from other healthcare areas within the same
region, as well as from other regions. Of these, 58% (26/45) were male. The age range was between 1 day and 8 years.

In 84% of cases, the symptoms began before the first year of life. In 44% of patients the diagnosis was obtained before the first month of life. These data are reflected in Table 1.

The mean diagnostic delay (time elapsed from the onset of symptoms until the first consultation with the otolaryngologist) was 1 month.

The most common presentation symptom was stridor, observed in 62% of patients (28/45), followed by dysphonia (hoarse voice or crying) in 49% (22/45) and aspiration or difficulty for feeding in 16% (7/45).

Placement of a nasogastric tube due to feeding problems was necessary in 53% (24/45) of cases. Two cases required a gastrostomy.

In total, 80% of patients (36/45) suffered unilateral paralysis. The majority of these cases affected the left vocal cord (71%, 32/45).

Among bilateral paralysis patients (10% of the total, 9/45), in the majority of cases (90%, 8/9) the vocal cords were in a midline or paramedian position. In the group of patients with unilateral paralysis, in 61% of cases (22/36) the affected vocal cord was in a midline or paramedian position, whereas in 39% of cases (14/36) it was in a lateral position. The difference between both groups was not statistically significant.

Overall, the aetiology of paralysis was iatrogenic in the majority of cases (71%, 32/45). The main cause of iatrogenic injury was heart surgery (58%, 26/45) (Table 2).

Regarding the group with bilateral paralysis (9 cases) the aetiology was: neurological disease (4 cases), iatrogenic (4 cases) and idiopathic in 1 case (Table 2).

A total of 11 patients (24%) recovered vocal cord mobility spontaneously (mean follow-up period of 16 months). All these patients with spontaneous recovery had suffered unilateral paralysis, except for 1 patient who suffered bilateral paralysis. The mean time until recovery of mobility was 8 months (median: 6 months). A proportion of 64% required more than 6 months to regain mobility.

Of the 34 patients (76%) who did not recover mobility, 8 required surgical treatment. These 8 patients presented bilateral paralysis with severe dyspnoea which required tracheotomy as an initial treatment. In 2 cases, it subsequently became necessary to perform endoscopic posterior cordectomy with CO2 laser extended to the ipsilateral band. These 2 cases were children with Arnold-Chiari malformation.

The remaining patients who did not recover laryngeal mobility did not require surgical treatment due to the development of sufficient glottic compensation phenomena to avoid the presence of severe symptoms (dyspnoea, aspiration or severe dysphonia).

Of the 8 patients who underwent tracheotomy, only 2 could be decannulated: one who underwent posterior cordectomy (Arnold-Chiari) and another with a prior

<table>
<thead>
<tr>
<th>Table 1</th>
<th>Age Distribution at the Onset of Symptoms and Diagnosis.</th>
</tr>
</thead>
<tbody>
<tr>
<td>Age</td>
<td>Onset of symptoms</td>
</tr>
<tr>
<td>≤1 month</td>
<td>38 (84)</td>
</tr>
<tr>
<td>1 month-1 year</td>
<td>5 (11)</td>
</tr>
<tr>
<td>&gt;1 year</td>
<td>2 (4)</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Table 2</th>
<th>Aetiology of Laryngeal Paralysis.</th>
</tr>
</thead>
<tbody>
<tr>
<td>Cause</td>
<td>No.</td>
</tr>
<tr>
<td>Iatrogenic</td>
<td></td>
</tr>
<tr>
<td>Heart surgery</td>
<td>26</td>
</tr>
<tr>
<td>DAL</td>
<td>11</td>
</tr>
<tr>
<td>AC</td>
<td>5</td>
</tr>
<tr>
<td>TGA</td>
<td>4</td>
</tr>
<tr>
<td>VSD + AC</td>
<td>2</td>
</tr>
<tr>
<td>VSD</td>
<td>1</td>
</tr>
<tr>
<td>ASA</td>
<td>1</td>
</tr>
<tr>
<td>VR</td>
<td>1</td>
</tr>
<tr>
<td>TF</td>
<td>1</td>
</tr>
<tr>
<td>Oesophageal atresia</td>
<td>3</td>
</tr>
<tr>
<td>Total thyroidectomy</td>
<td>2</td>
</tr>
<tr>
<td>Prolonged intubation</td>
<td>1</td>
</tr>
<tr>
<td>Idiopathic</td>
<td></td>
</tr>
<tr>
<td>Neurological</td>
<td></td>
</tr>
<tr>
<td>Arnold-Chiari</td>
<td>3</td>
</tr>
<tr>
<td>Cerebral anoxia</td>
<td>1</td>
</tr>
<tr>
<td>Obstetrics</td>
<td>1</td>
</tr>
<tr>
<td>Total</td>
<td>45</td>
</tr>
</tbody>
</table>

AC, aortic coarctation; ASA, aberrant subclavian artery; DAL, ductus arteriosus ligation; L, left; R, right; TF, tetralogy of Fallot; TGA, transposition of the great arteries; VR, valve replacement; VSD, ventricular septal defect.
history of total thyroidectomy who regained mobility of the right vocal cord, but not of the left.

Dividing patients into those with iatrogenic and non-iatrogenic causes, the first group was less likely to regain laryngeal mobility compared to those with non-iatrogenic causes \((P=0.05)\).

Regarding the onset of symptoms, iatrogenic cases started later than non-iatrogenic cases \((P=0.01)\). Age at diagnosis followed the same pattern, that is, children with iatrogenic paralysis were older at the time of diagnosis than non-iatrogenic cases \((P=0.007)\).

As for the diagnostic delay, iatrogenic cases experienced a longer delay. In this case, the difference between both groups was not statistically significant \((P=0.118)\), but it was clinically relevant.

Discussion

Vocal cord paralysis in the paediatric population is a well-known cause of stridor and dysphonia. The number of cases has increased in recent years. This work presents a greater number of cases than other previous publications.\(^1,5,9\) The reason for this increased incidence is probably multifactorial. On one hand, survival of more premature infants with more diseases is increasing due to improvements in paediatric intensive care units. On the other hand, paediatric flexible nasofibroscopes allow a dynamic assessment of wakeful patients and the availability of such nasofibroscopes among otolaryngologists is increasing every day, thus allowing a more accurate diagnosis of children who attend consultation with stridor and dysphonia.

Moreover, the possibility of recording examinations represents a very useful tool, enabling explorations to be re-assessed in difficult cases, by more than one professional if required. These recordings also play an important role in the training of future otolaryngologists (ENT medical residents), whilst also allowing fibroscopies to be reviewed with paediatricians or with the parents of patients, with training purposes in the first case and to help their understanding of the diagnosis in the case of parents.

The diagnosis can also be obtained by direct laryngoscopy under general anaesthesia.\(^1\) This method allows palpation of the arytenoids in order to rule out cricoarytenoid joint fixation.

Unilateral paralysis is more common than bilateral and it primarily affects the left vocal cord.\(^5,7\)

The incidence of paediatric laryngeal paralysis is greater during the first 2 years of life.\(^10\)

For some authors, dysphonia is the most common symptom in children.\(^11\) In the case of very young children, dysphonia manifests as an abnormal and dysphonic crying, which in the case of vocal cord paralysis, is usually very weak.\(^11\) In other series, as in ours, stridor is the main symptom.\(^1\)

Laryngeal electromyography, introduced by Hirano and Ohalan in 1969,\(^12\) is the most suitable procedure for the diagnosis of neuromuscular disorders. It has proven useful for the differential diagnosis between paralysis and mechanical fixation of the vocal cords.\(^5,13-15\) Although in adults this diagnostic method is relatively simple, in children it usually requires general anaesthesia.\(^16\) In general, unlike in adults, laryngeal electromyography is rarely used for the diagnosis of paediatric laryngeal paralysis.\(^10\)

Iatrogenic injury is the most frequent cause of paralysis in some series,\(^1\) while in others idiopathic cases are the most common.\(^10\) Neurological causes are cited as the third most frequent cause,\(^10\) as in our work.

The number of iatrogenic cases in our series was greater than in others in the literature. Heart surgery is the most common cause of iatrogenic laryngeal paralysis in children and most cases undergo ductus arteriosus ligation,\(^1\) as in our series. Surgical procedures aimed at treating patent ductus arteriosus and surgical manipulations of the aortic arch represent a risk of injury for the recurrent laryngeal nerve, especially on the left side.\(^6,17\) In this regard, the prevalence of laryngeal paralysis following ductus arteriosus ligation oscillates between 11% and 52%,\(^6,17\) whilst this prevalence following any cardiovascular surgical procedure in paediatric age is 1.7%.\(^18\) Multiple factors may be responsible for nerve injury, including retraction, cannulation of the internal jugular vein, tracheal intubation and low temperatures in the pleural cavity caused by irrigation with cold saline solution during heart surgery.\(^19\)

Other factors reflected in the literature are prolonged intubation\(^20\) and the association of a congenital heart defect with a congenital laryngotracheal malformation.\(^18\) The high number of cases in our series is probably related to the intense surgical activity of our paediatric cardiothoracic surgery service, which is the national referral centre, along with the high number of births at our hospital, about 10000 during the year in which the patients were gathered, thus resulting in a large population of premature infants with low weight at birth, candidates for heart surgery.

Regarding the aetiology of bilateral paralysis, in our work it was mainly due to neurological causes. This is in accordance with most studies, which point to Arnold–Chiari malformation as the most frequent cause of bilateral laryngeal paralysis.\(^1,4,12\) Nevertheless, there are some studies which differ in this respect.\(^21,22\)

To the best of our knowledge, there are no previous studies comparing the characteristics of patients with iatrogenic and non-iatrogenic paralysis. The age of onset of the former is obviously related to the time of surgery. The recovery rate was higher in the non-iatrogenic group, probably because recurrent nerve lesions are often irreversible. The diagnostic delay was higher in patients with iatrogenic paralysis. This was investigated thoroughly and the work data have been submitted to cardiovascular surgeons in order to raise awareness of the importance of early referral of patients for ENT evaluation upon appearance of suspicious symptoms after surgery. Further studies will be required to evaluate the result of these measures.

The rate of spontaneous recovery of laryngeal paralysis reflected in the literature ranges between 0% and 82% of cases.\(^1,6,22\) This variability depends on the cause of the nerve lesion, as well as on the comorbidities suffered by these patients.

Spontaneous recovery usually takes place within the first 6 months.\(^5\) In our series, up to 27% of patients regained mobility after the first 6 months and there was 1 patient who recovered it after 1 year. Therefore, we believe that waiting for this length of time before performing any definitive
procedure is advisable. Nevertheless, the decision to perform surgical treatment in the case of paediatric laryngeal paralysis is always difficult.

Most cases of bilateral paralysis require surgical treatment, as was the case in our series. By contrast, unilateral cases rarely require surgery.

Medialisation of the paralysed vocal cord in abduction is the treatment of choice for many authors in patients under 10 years of age. The indications for surgical treatment in the case of unilateral paralysis are: aspiration and severe dysphonia which interfere with communication after exhausting speech rehabilitation therapy. Nevertheless, it is not recommended in cases of paralysis with a duration under 1 year. Some authors have tested intracordal injection in the paralysed vocal cord in order to improve the voice of paediatric patients. In our series, we have not performed surgical treatment in cases of unilateral paralysis, perhaps due to the follow-up period.

Regarding bilateral paralysis in patients with tracheostomy, several surgical techniques have been described: reinnervation techniques, laryngeal reconstruction and vocal cord lateralisation through arytenoidopexy or arytenoidecotomy, both by an external route and with endoscopic CO2 laser. In this regard, some authors argue that external approaches lead to a higher rate of decannulation. Dennis and Kashima described a technique for endoscopic posterior cordectomy with CO2 laser which was the technique of choice for 2 patients in our series. This technique has been proven to achieve a high percentage of decannulation with less damage to vocal function.

Conclusions

The incidence of laryngeal paralysis in our series was higher than in other published series, mainly at the expense of surgical procedures performed in the anatomical pathway of the recurrent laryngeal nerve.

Spontaneous compensation or recovery is the norm in most cases. Surgery is only required in certain cases.

Cases of iatrogenic paralysis are associated with a delayed onset, with greater diagnostic delay and with a worse rate of recovery.

Collaboration between otolaryngologists, paediatricians and cardiac surgeons is essential to avoid diagnostic delays, as well as to work together in the implementation of prevention strategies.

Conflict of Interests

The authors have no conflict of interest to declare.

References


