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Review article

Paralyzed neonatal larynx in adduction. Case series, systematic review and analysis

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ABSTRACT

Objective: Bilateral vocal cord abductor paralysis (BVCAbP) is considered a rare cause of stridor in the newborn. The goal of this work is to present a case series and to review systematically the literature on bilateral vocal cord abductor paralysis in the newborn to better characterize the current knowledge on this entity.
Methods: We performed a systematic literature review with Medline (1950–2011). The authors screened all cases of BVCAbP reported and selected those affecting newborns.
Results: Out of the 129 articles screened, 16 were included. A total of 69 cases could be retrieved and analyzed. Associated co-morbidities were found in 54% of the patients, most notably malformative conditions (intracranial or other), or a positive perinatal history (trauma/asphyxia, prematurity). Tracheostomy placement was required in 59% of children, and of these 44% were successfully decannulated. In terms of functional outcome full recovery or improvement were seen in 61% of patients. Major underlying co-morbidities affected negatively the functional outcome ($p = .004$), but not the need for tracheostomy ($p = .604$) or the decannulation success rate ($p = .063$).
Conclusion: BVCAbP in the newborn is a serious cause of airway obstruction. It can be seen either in a context of multisystem anomalies or as an isolated finding. Newborns with major co-morbidities affecting their normal development are more likely to have poor functional outcomes and to remain tracheostomy-dependant.

Contents

1. Introduction

Stridor in the newborn can be the manifestation of multiple congenital anomalies of the airway. Due to the size and the particular conformation of the upper airway in the newborn stridor

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represents a dreadful alerting sign of a potentially life-threatening anomaly. The primary approach consists of airway stabilization and must be followed by a secondary diagnostic approach [1].

In children, vocal cord paralysis (unilateral or bilateral) represents the second most common cause of congenital anomaly of the larynx, after laryngomalacia and preceding congenital subglottic stenosis [2].

The term vocal cord paralysis encompasses a broad range of clinical pictures. In any case, the 2 initial and most important considerations in the diagnostic approach of the pediatric patient with vocal cord paralysis is determining the onset mode (congenital versus late-onset), and the unilateral versus bilateral involvement.

Bilateral abductor paralysis (BVCAbP) represents a unique subgroup within this entity, posing the additional challenge of upper airway obstruction during inspiration. Two etiologic groups of bilateral vocal cord paralysis in children have been classically considered: idiopathic and secondary to a neurologic disorder. The main disorders found in the second group are Arnold–Chiari malformation, hydrocephalus, myelomeningocele, hypoxic cerebral palsy, and hemorrhage [3].

Even though the management of congenital bilateral vocal cord paralysis has become more conservative, it is not clear whether or not this statement can be fully applied to the abduction subtype [4]. Indeed, since there is a potential for spontaneous recovery the ideal approach should permit to relieve the symptoms of respiratory obstruction with minimal disruptive effects on swallowing and speech. For the children who develop severe respiratory obstruction, tracheotomy has been the classically used approach. This method fulfills the functional preservation concerns but carries significant morbidity and mortality in the pediatric group [5].

In this systematic review we analyzed the cases of congenital BVCAbP with onset in the neonatal period in order to provide an overview on the current knowledge, the evolution of management approaches and the outcome of this entity. We sought to identify potential factors that influence disease outcomes. Finally, we add three illustrative cases to the existing literature.

2. Methods

2.1. Search strategy and selection of publications

We performed a literature search with Medline as main research engine (1950–2011), using the following Boolean operator combinations: "[(vocal cord OR vocal fold OR laryngeal) AND ("abductor")] AND ("paralysis OR palsy")". The term "newborn" or "infant" was not included within the search in order to avoid selection bias.

We screened the abstracts of all the articles produced and retrieved the full text of the relevant ones. Language restriction was done for articles in English, French, German, Spanish, and Italian. References from selected articles were also screened in order to identify further relevant publications.

We selected case reports and case series (prospective and retrospective), describing cases of newborns with bilateral abductor vocal cord paralysis. Onset within the first 48 h following birth had to be explicitly reported for a case to be included.

We describe an institutional series of illustrative cases that were included for analysis purposes.

2.2. Data extraction

We extracted the following data: age, gender, co-morbidities, presentation, management, and outcome.

2.3. Data analysis

We calculated summary statistics in order to characterize patients' features, and then performed several comparisons of subgroups using the Fisher's exact test. Comparison of means was performed with Student's *t*-test. Assessment of correlation between 2 variables was done using the logistic regression model. Statistical significance was set at p-value <0.05 (two-sided).

2.4. Case series

2.4.1. Case 1

Male, born at 32 weeks of gestation with forceps, presented immediate stridor and episodes of desaturation treated with non-invasive ventilation (continuous positive airway pressure - CPAP).

Endoscopic workup revealed Hollinger type II laryngomalacia, bilateral vocal cord paralysis in paramedian position without any abduction movements, and grade I subglottic stenosis according to Myer and Cotton.

Neurological workup allowed diagnosing Sotos syndrome and Chiari malformation type I, with the patient undergoing ventriculocysternostomy. After this procedure the patient could be weaned off the CPAP.

The follow-up at 17 months of life showed full recovery of laryngeal motor function. The subglottic stenosis was not evident anymore.

2.4.2. Case 2

Female, born prematurely at 26 weeks of gestation, underwent immediate orotracheal intubation for respiratory distress with stridor. After multiple failed attempts of extubation she could be finally started on CPAP at 2 months of age.

Endoscopic workup revealed type I laryngomalacia and bilateral vocal cord abductor paralysis. The rest of the diagnostic workup was non-contributory.

Non invasive ventilation could be gradually stopped by age 4.5 months.

The patient did well clinically and repeat endoscopy at 12, 18 and 24 months revealed full recovery of the right vocal cord paralysis, but persistent paralysis of the left side.

2.4.3. Case 3

Male, born at 41 weeks of gestation with forceps delivery, presented immediate respiratory distress with stridor and hypotonia. Non-invasive treatment with CPAP was started and continued for 3 weeks.

Endoscopic workup revealed type I laryngomalacia and bilateral vocal cord paralysis (Fig. 1). Imaging of the central nervous system showed a supratentorial subdural hematoma with no other anomalies.

The patient's clinical evolution was excellent.

Endoscopic follow-up at 6 months showed complete recovery of the right vocal cord paralysis and only slight abduction on the left side (Fig. 2), without persistent signs of laryngomalacia.

3. Results

The literature search produced 129 articles. Of these 11 were excluded for language reasons (articles in Japanese, Chinese and Dutch language), 48 concerned adults, 91 were produced by keyword matching but were not relevant to the subject of this review, and 3 were excluded because not enough patients' features could be retrieved to make sure inclusion criteria were fulfilled. An additional 2 articles were excluded as they reported late-onset BVCAbP (stridor not presenting within the first 48 h after birth). After the selection process, 16 articles [6–22] reporting 66 cases