International Journal of Pediatric Otorhinolaryngology 79 (2015) 202–209

Contents lists available at ScienceDirect

International Journal of Pediatric Otorhinolaryngology

journal homepage: www.elsevier.com/locate/ijporl

Spontaneous recovery of bilateral congenital idiopathic laryngeal paralysis: Systematic non-meta-analytical review

Mohammed Jomah [a,1], Caroline Jeffery [b], Sandra Campbell [c], Aleksandra Krajacic [d], Hamdy El-Hakim [e,f,*]

[a] Department of Surgery, Faculty of Medicine and Dentistry, University of Alberta, Edmonton, Alberta, Canada
[b] Division of Otolaryngology – Head & Neck Surgery, The Stollery Children's Hospital & The University of Alberta Hospitals, Faculty of Medicine and Dentistry, University of Alberta, Edmonton, Alberta, Canada
[c] John W. Scott Health Science Library, University of Alberta, Edmonton, Alberta, Canada
[d] Department of Surgery, The Stollery Children's Hospital & The University of Alberta Hospitals, Faculty of Medicine and Dentistry, University of Alberta, Edmonton, Alberta, Canada
[e] Pediatric Otolaryngology Service, Division of Otolaryngology – Head & Neck Surgery, Department of Surgery, The Stollery Children's Hospital & The University of Alberta Hospitals, Faculty of Medicine and Dentistry, University of Alberta, Edmonton, Alberta, Canada
[f] Pediatric Otolaryngology Service, Division of Pediatric Surgery, Department of Pediatrics, The Stollery Children's Hospital & The University of Alberta Hospitals, Faculty of Medicine and Dentistry, University of Alberta, Edmonton, Alberta, Canada

ARTICLE INFO

Article history:
Received 9 September 2014
Received in revised form 7 December 2014
Accepted 8 December 2014
Available online 15 December 2014

Keywords:
Larynx
Paralysis
Congenital
Idiopathic
Bilateral
Recovery

ABSTRACT

Objectives: To systematically review the frequency and time to spontaneous recovery in pediatric patients with bilateral congenital idiopathic laryngeal paralysis (BCILP).
Methods: The databases of Medline, EMBASE, Scopus, CINAHL, Cochrane Library and Proquest Dissertations were searched for English language articles reporting on laryngeal paralysis in pediatric patients. A bibliography search of the selected studies was done to identify additional articles. We included prospective or retrospective case-series studies of children and neonates diagnosed with BCILP at age <60 days and confirmed by direct laryngoscopy, with sufficient follow up and objective assessment for recovery.

Two authors independently extracted the data and assessed the quality of each study. Discrepancies were resolved by consensus and adjudication by a third author.
Results: Of the 4229 articles identified by the search, only one study met our inclusion criteria. The study was a retrospective case series, and was of low quality. The mean age at diagnosis was fourteen days. Sixty-five percent of the patients recovered spontaneously, and the mean time to recovery was twenty-five months. Tracheostomy was performed in 71% of the patients.
Conclusions: The available literature is of low quality and provides weak evidence on the natural history of BCILP in pediatric population.

1. Introduction

The natural course and pathophysiology of laryngeal paralysis (LP) contain significant knowledge gaps [1] this is particularly so with respect to the etiology and epidemiology of bilateral congenital idiopathic laryngeal paralysis (BCILP) [2]. Overall, case series reported variable rates of spontaneous recovery, from as early as the first four weeks to as late as eleven years [3–7]. However, most of these studies suffer significant methodological limitations. Often, objective measures of resolution were not used as the primary endpoint. Instead, rates of decannulation or subjective judgment on symptomatic improvement/or resolution had been used. In addition, several studies failed to include long-term follow up with endoscopic examination [8].

Currently management decisions are neither based on likelihood of recovery [2] nor guided by evidence-based information.

Tracheostomy is considered the reference standard for managing LP in neonates and children [9,10]. However, it is associated

* Corresponding author at: Pediatric Otolaryngology, 2C3.57 Walter MacKenzie Health Science Centre, 8440-112 Street, Edmonton, Alberta, Canada T6G 2B7. Tel.: +1 780 407 8629; fax: +1 780 407 2004.
E-mail address: Hamdy.ElHakim@albertahealthservices.ca (H. El-Hakim).
[1] Department of Otolaryngology, College of Medicine, King Saud University, Riyadh, Saudi Arabia.

http://dx.doi.org/10.1016/j.ijporl.2014.12.007
0165-5876/

with significant morbidity and occasional mortality [11–13]. Also, although tracheostomy is effective in securing the airway, it does not address directly the etiology and pathogenesis of the disease. This often makes subsequent surgical interventions in order to achieve decannulation necessary [9]. These interventions (e.g. cordotomy, arytenoidectomy, and arytenoidopexy) are not uniformly utilized, and there is no consensus on their optimal techniques, indications or timing [9]. Since most of the described procedures result in some compromise to the laryngeal function and/or structure, counseling the child and family on their choice without epidemiological data compounds the process even more.

Recently a systematic review on newborn patients with bilateral abductor LP concluded that full or partial recovery occur in 61% of cases [14]. The review however, included all etiological classes of LP.

Our study aims to conduct a systematic review of the current literature on the natural history of pediatric BCILP and rates of spontaneous resolution.

2. Methods

2.1. Data source and study selection

2.1.1. Electronic searches

In January, 2013 a specialized health science librarian (S.C.) searched Medline, EMBASE, Scopus, CINAHL, Cochrane Library and Proquest Dissertations databases using combinations of keywords and MESH headings such as ((Vocal Cord Paralysis/or laryngeal paraly*.mp. or laryngeal paresis.mp. or (vocal cord* adj2 (paresis or paraly*)).mp.) NOT unilateral.mp.) to identify all published studies reporting the natural resolution of laryngeal paralysis. The search was adapted to each database. The search was limited to original research papers published in English language. Reference lists from the identified studies were scanned for additional studies. The article references were managed using RefWorks 2.0 (ProQuest LLC) online software. Duplicates were removed using the same program.

2.1.2. Study selection

We screened all the titles and abstracts from the primary electronic search after removing duplicates and excluded non-eligible literature (e.g. adult, acquired, post-thyroidectomy, skull base). Full manuscripts of the relevant papers were obtained.

Two authors (C.J. and M.J.) assessed the manuscripts fully for eligibility of inclusion. Any disagreement about the study selection was resolved by consensus and adjudication by the third author (H.E.).

2.2. Study eligibility criteria

2.2.1. Types of studies

1. Human studies.
2. Published in English language.

2.2.2. Types of participants

1. Neonates or children ($\leq$18 years).
2. Confirmed diagnosis of BCILP.

2.2.3. Diagnosis

1. Initial diagnosis made prior to two months of age, either clinically or by bedside fiber-optic flexible laryngeal endoscopy (FFLE).
2. Confirmed with direct rigid laryngoscopy (DRL) with palpation of arytenoids (PA) to rule out fixation and other airway anomalies [15].
3. Idiopathic etiology assigned based on clearly described investigative work-up to rule out other etiologies.

2.2.4. Types of outcome measures

Partial or complete spontaneous resolution based on one of:

1. FFLE
2. DRL

$\pm$ Laryngeal electromyography (LEMG).

2.2.5. Follow up

Follow up for sufficient time after diagnosis until the outcome "recovery" has taken place or at least for 6 months to label the case as "not recovered".

2.2.6. Exclusion criteria

1. Previous surgical procedure to thorax or head and neck area.
2. Studies aiming at assessing the effectiveness of a surgical or medical intervention in treating LP (other than tracheostomy).
3. Insufficient or absent information on follow up.
4. Case reports.
5. Narrative reviews and expert opinion.
6. Absence of imaging studies ruling out central or neurological etiologies.

2.3. Data extraction and quality assessment

2.3.1. Data extraction

Two independent authors (C.J. and M.J.) extracted the data. We entered the following data into a spread sheet (Microsoft Excel® 2011): authors and year, sample size, sampling method, diagnostic method(s), age at diagnosis, follow-up length and percentage, spontaneous recovery rate and timing, method of assessing recovery, and tracheostomy rate. For each article, a level of evidence score was assigned based on Oxford Centre for Evidence-based Medicine Levels of Evidence [16].

2.3.2. Quality assessment

Two independent authors (C.J. and M.J.) completed the quality assessment using The Center for Evidence Based Medicine diagnostic study critical appraisal tool [17]. Variables assessed included sampling method, follow up duration, objectivity of outcome assessment, subgroup stratification, outcome course over time, precision of outcome estimate and applicability to other patients. Variables were evaluated according to the following criteria:

- Sampling:
  We evaluated:
  i. Consecutiveness.
  ii. Primary establishment of diagnosis at onset.
  iii. Accounting for excluded or missing patients.
  iv. Uniformity and detail of diagnostic modality. The minimum standard was FFLE, DRL with PA to rule out fixation.
  v. Confirmation of congenital idiopathic etiology by history and investigations.
- Follow up:
  We assessed length, and completeness of follow up. Unless the patients had achieved the outcome of interest, a follow up for at least six months was the minimum acceptable.
- Outcome objectivity:
  We included studies that used either FFLE, or DRL, with or without LEMG. Neither symptomatic improvement nor decannulation was accepted.