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

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Disclosure

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Background

- Bilateral laryngeal paralysis (LP) is the absence of normal movement of both vocal folds due to affection of neural supply.
- Different etiological categories.
- Available literature on LP:
  - Iatrogenic LP reportedly persists in many
  - Vincristine induced LP recovers after cessation of treatment.
  - Other categories including bilateral congenital idiopathic laryngeal paralysis (BCILP) have no clear natural history

Clement WA et al. Arch Otolaryngol Head & Neck Surg. 2008 Jan;134(1):28-33
Kuruvilla G. Arch Otolaryngol Head & Neck Surg. 2009 Jan;135(1):101-5
Berkowitz RG. Otolaryngol Head Neck Surg. 2007 Apr;136(4):649-52

Background

- General management approach is not well defined
  - Conservative vs. tracheostomy
- Decision making and family counseling require knowledge on natural history
- Recent systematic review on newborns with bilateral LP reported a recovery rate of 61% for all etiological categories

Brigger MT et al. Otolaryngol Head Neck Surg. 2002 Apr;126(4):349-55
Nisa L et al. Int J Pediatr Otorhinolaryngol. 2013 Jan;77(1):13-8

Objectives

- To systematically review the English literature and determine the rate and time to recovery of BCILP

Method
Inclusion criteria

- Types of studies:
  - Inclusive case series of children or children and adults
  - English language

- Types of participants:
  - Neonates or children (≤18 years)
  - Confirmed diagnosis of BCILP
    - Age at diagnosis (< 60 days)
    - Direct laryngoscopy (DL) with palpation of arytenoid
    - Work-up to rule out other etiologies

Method
Inclusion criteria

- Follow up:
  - Sufficiently long to detect outcome

- Exclusion Criteria:
  - Recovery based only on symptom resolution
  - Studies aimed at evaluating surgical or medical interventions
  - Single case reports
  - Narrative reviews and expert opinion

Method
Electronic search

- Cochrane Library, EMBASE, Medline, SCOPUS, CINAHL, and Proquest Dissertations
- 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.)
- Limited to English language and Adult, Human populations.
- No date limit applied.

Method
Other search methods

- Bibliography check of selected studies.

- References were managed using RefWork.

Method
Data extraction and quality assessment

- Two independent authors
- Variable collected
  - Authors, publication year
  - Design
  - Sample size and method
  - *Diagnosis: method, mean age at diagnosis*
  - Follow up: length and percentage
  - *Spontaneous recovery: rate, time to, and assessment method*
  - Tracheostomy rate
- Quality assessment:
  - The Center for Evidence Based Medicine diagnostic study critical appraisal tool

Method

Outcome measures

- Partial or complete spontaneous resolution based on:
  - Direct laryngoscopy (DL)
  - Flexible fiberoptic laryngoscopy (FFL)
  - ± Laryngeal electromyography (LEMG)

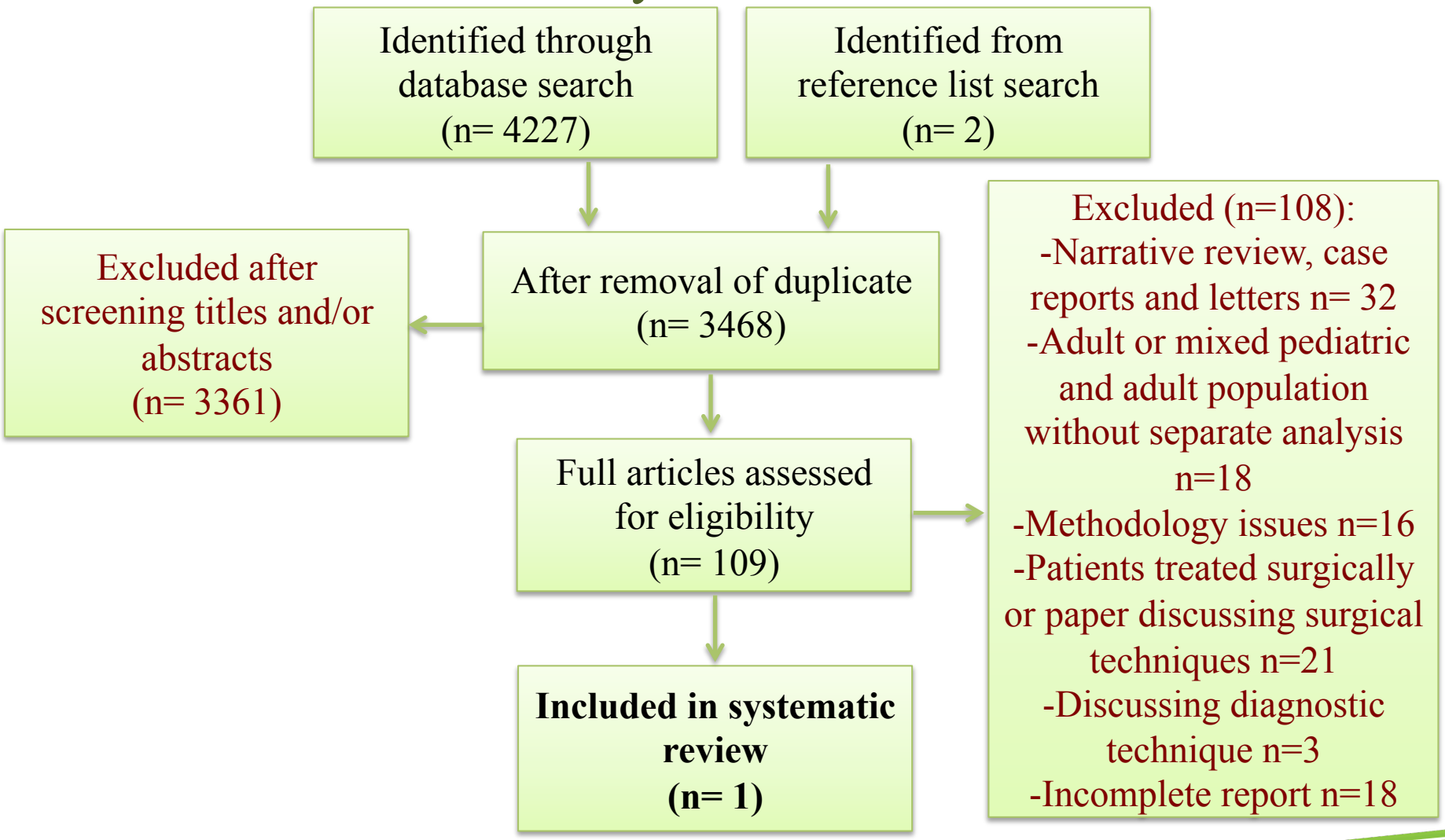
Method

Statistical analysis

- Unsuitable for meta-analysis
- Descriptive statistics
- Qualitative analysis

Results

Study selection



Results

Quality assessment

Study	Sampling	Follow up	Outcome Objectivity	Subgroup analysis	Outcome reporting	Precision of estimate	Applicability
Miyamoto et al. 2005	√[1]	√	√	√[2]	√[3]	–	√

1 Some patients with possible neurological causes labeled as idiopathic, three patients excluded without giving the reason (vague inclusion criteria).
2 Analysis is based on tracheostomy status of the patients.
3 Some data are missing from the paper

Miyamoto RC et al. Otolaryngol Head Neck Surg 2005 Aug;133(2):241-5

Results
Collected data

Reference	Level of evidence	BCILP n	Diagnostic method	Avg. age at diagnosis	Avg. F/U months	Percentage of followed patients	Spontaneous recovery n	Avg. time to recovery months	Recovery assessment method
Miyamoto et al. 2005	IV	17	DL/ FFL/ SS	13.7 days	50	100%	11	25.3	FFL

FFL: Flexible fiberoptic laryngoscopy, DL: Direct laryngoscopy, SS: Symptoms and signs, F/U: Follow up

Miyamoto RC et al. Otolaryngol Head Neck Surg 2005 Aug;133(2):241-5

Conclusion

- The available literature is of low quality and provides a weak level of evidence on the natural history of pediatric BCILP.

Significance

- This is the first study to systematically review the literature on BCILP.
- The available figures should be cautiously used for counseling.
- Future work include performing prospective multicenter study on LP.

Thank you