

# Adult laryngomalacia: A systematic scoping review with meta-analysis

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## BACKGROUND

- New onset laryngomalacia diagnosed in the adult population is uncommon and poorly defined. It has been reported to occur in patients with severe neurological compromise.<sup>1</sup>
- In a similar category of disease, exercise-induced laryngeal obstruction (EILO) presents with stridor and dyspnea with exertion.<sup>2</sup> This is diagnosed with a continuous laryngoscopy exercise test (CLE-test).<sup>3</sup>
- While this variant is better defined as aerodynamic supraglottic collapse, it mimics laryngomalacia.
- We believe this condition is likely underrecognized, characterized with inconsistent nomenclature, and in need of dedicated research.

## METHODS

- The review followed the five-stage framework outlined by Arksey and O'Malley (2005) and was performed in accordance with the Preferred Reporting Items for Systematic reviews and Meta-Analysis extension for Scoping Reviews (PRISMA-ScR) checklist.<sup>4</sup>
- Stage 1, Research Question: What are the characteristics of adults diagnosed with new onset laryngomalacia?
- Stage 2, Relevant Literature: A comprehensive literature search was performed from inception until May 1, 2023, in CINAHL (EBSCO), Cochrane Review, PubMed, and Scopus (Elsevier) databases.
- Stage 3, Study Selection: Two authors independently screened articles based on title and abstract, and then by full text.
- Stage 4, Charting the Data: Data was extracted by the primary author and appraised by the second author. All the included reports were critically appraised to assess the level of evidence using the Oxford Center for Evidence-Based Medicine criteria.<sup>5</sup>
- Stage 5, Collating, Summarizing, and Reporting Results: Meta-analysis of continuous measures and meta-analysis of proportions was performed. Each measure (mean/proportion and 95% confidence interval (CI) was weighted according to the number of patients affected. Finally, potential publication bias was evaluated by visual inspection of the funnel plot and Egger's regression test, which statistically examines the asymmetry of the funnel plot.

## RESULTS

Figure 1: PRISMA Diagram of Included Studies

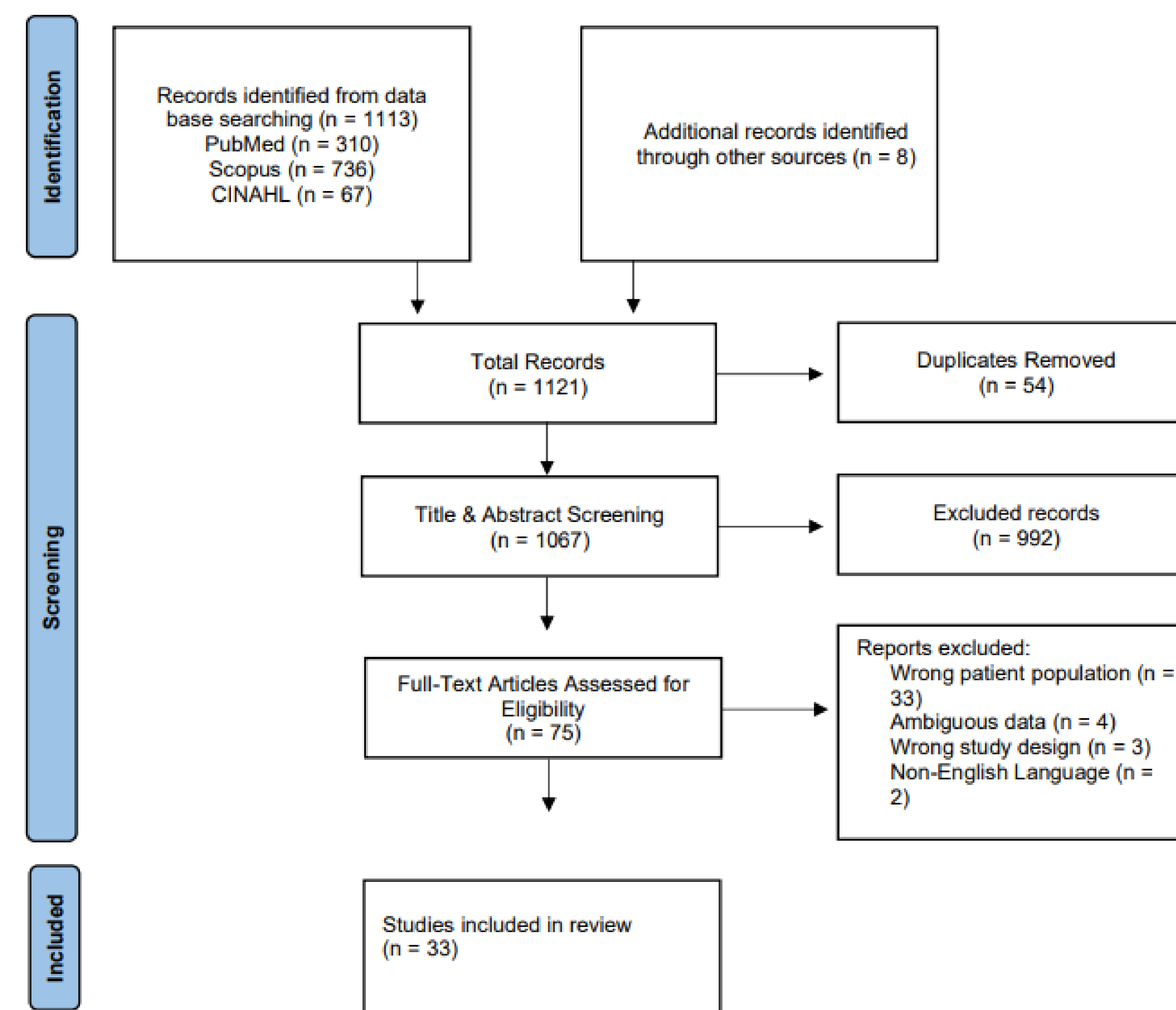


Figure 2: Funnel Plot Assessing Publication Bias

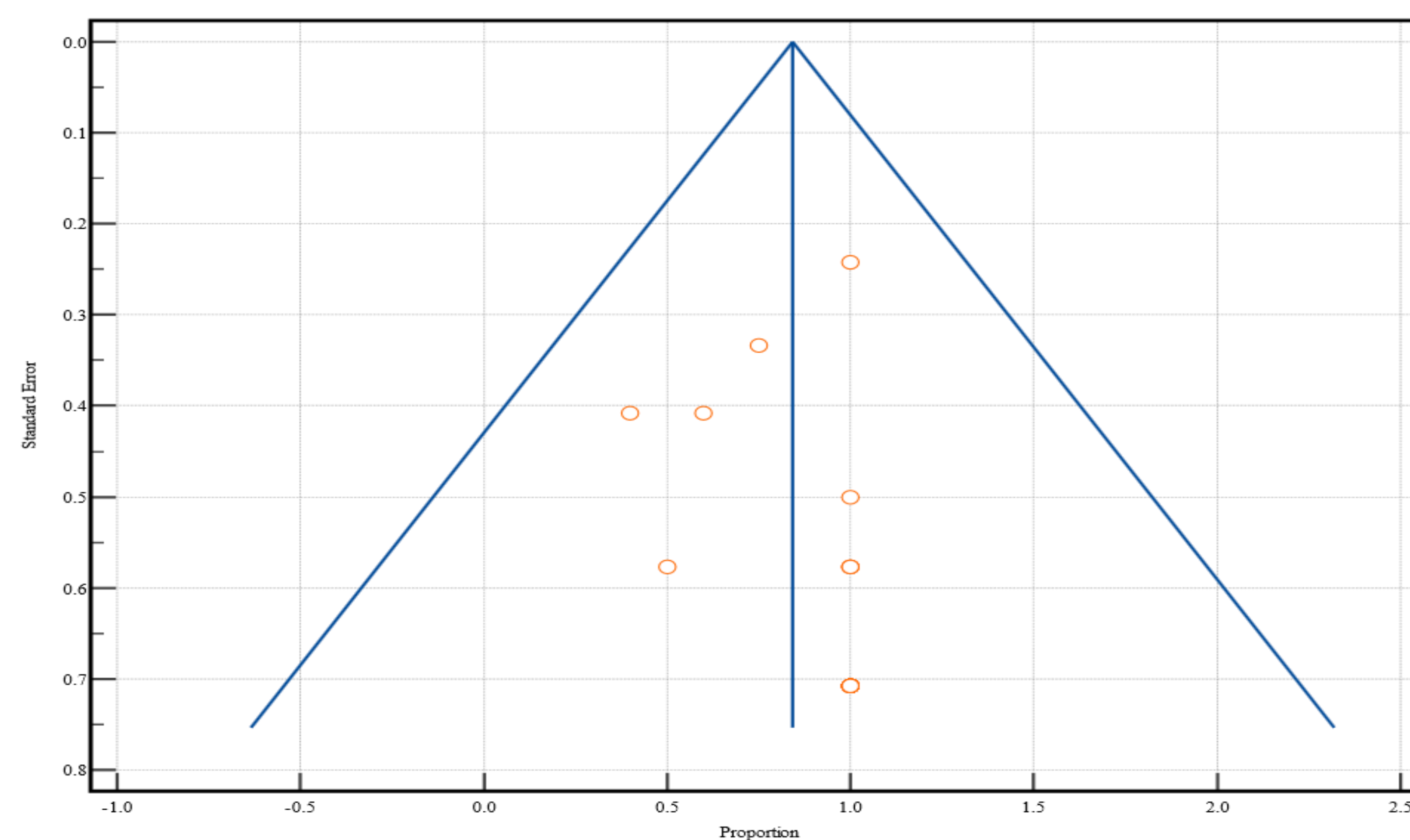


Table 2: Meta-proportions of Outcomes

Outcome	n	%	95% CI	I <sup>2</sup>
Complete resolution of symptoms after therapy	41	61.9	47.9 to 74.5	17.21%
Improvement in symptoms after therapy	43	68.9	55.3 to 80.5	42.78%
Recurrence of symptoms after therapy	20	64.5	10.1 to 99.9	82.06%
Persistence of symptoms without therapy	6	53.4	17.9 to 86.4	26.95%
Spontaneous resolution	9	45.7	17.9 to 75.6	0.00%
Permanent tracheostomy	4	71.6	29.2 to 96.4	0.00%

## CONCLUSIONS

- Adult onset laryngomalacia is difficult to characterize, and its existence might not be agreed upon in the field of laryngology.
- When it is reported, it typically presents with stridor at rest and dyspnea on exertion in patients during exercise, with neurological injury, or idiopathically. In adults, this disease is most likely multi-factorial.
- The need for a universal nomenclature is highlighted in this review, as ALM is most likely underrecognized or inconsistently classified.

Table 1: Summary Table

Author Year	Total Patients (n)	Age (Years)	Suspected Etiology (n)	Management
Akimoto 2022	1	68	Neurological	Tracheostomy
Arora 2005	1	18	Persistence from childhood	SG
Bartolomeo 2015	1	25	Idiopathic	Partial EG
Chetty 1994	1	42	Post surgical	Patient refusal
Conaway 2004	1	33	Neurological	Conservative
Connor 2013	1	34	OSA	Initial SG, followed by tracheostomy with repeat SG
Dion 2012	5	34	OSA (2), exercise-induced (3)	SG (3), patient referred elsewhere (1), lost to follow up (1)
Echternach 2008	1	47	Post surgical	Partial EG
Famokunwa 2020	16	32.1	Traumatic (1), exercise-induced (15)	SG (16)
Ferri 2020	1	36	Idiopathic	Conservative therapy followed by SG
Gessler 2002	1	27	Idiopathic	SG
Harries 1995	1	73	Idiopathic	Epiglottic fixing
Hey 2014	2	46.5	Idiopathic (2)	Multiple SG with tracheostomy (1), multiple laser epiglottopexy with tracheostomy (1)
Kanemaru 2007	1	37	Idiopathic	Partial EG
Kawamoto 2013	1	18	Idiopathic	Partial EG
Keshwani 2020	1	40	Compression	Tracheostomy
Kletzker 1990	3	65	Idiopathic (3)	SG (3)
Li 2002	1	24	Osteogenesis Imperfecta	SG with partial EG
Lim 2011	1	21	Exercise-induced	Conservative
Maat 2007	2	18	Exercise-induced (2)	SG (2)
Mima 1996	1	62	Post surgical	Tracheostomy with forward hyoid traction
Ng 2022	1	50	Neurological	Tracheostomy with partial EG and epiglottic fixing
Nonomura 1996	1	55	Idiopathic	Tracheostomy with SG
Panchasara 2015	2	21	Exercise-induced (2)	Conservative (2)
Peron 1988	2	19.5	Neurological (2)	SG (1), observation (1)
Rutt 2014	1	61	Idiopathic	SG (2)
Siou 2002	2	69	Idiopathic (2)	SG (2)
Takeshita 2017	1	86	Post surgical	Tracheostomy, then transfer to other hospital
Templer 1981	1	18	Congenital abnormality	Tracheostomy with removal of epiglottis, ventricular bands, and aryepiglottic folds
Veiga 2021	1	49	Idiopathic	Not reported
Vezina 2020	1	55	Exercise-induced	Conservative
Wiggs 1995	5	44.6	Neurological (5)	Observation (3), tracheostomy (2)
Woo 1992	8	37.4	Neurological (6), post-surgical (1), traumatic (1)	EG (4), EG with SG (1), observation (2), laryngectomy (1)

SG = supraglottoplasty; EG = epiglottectomy; OSA = obstructive sleep apnea

## REFERENCES

Please scan QR code for supplemental data.

