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Congenital Airway Anomalies in Children: A Decade of Evidence on Pulmonologic Challenges, Surgical Management, and Speech Rehabilitation — A Systematic Review and Pooled Cohort Analysis of Published Data (2015–2024)

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ABSTRACT

Background: Congenital airway anomalies (CAA) are rare but serious structural malformations of the upper and lower airways in children, frequently presenting with respiratory distress, feeding difficulties, and impaired speech development. The past decade has witnessed major advances in diagnosis, surgical correction, and multidisciplinary rehabilitation, yet global outcome data remain fragmented.

Objective: To systematically review literature published between 2015 and 2024 on congenital airway anomalies in children, evaluating pulmonologic outcomes, surgical management strategies, and speech/voice rehabilitation, and to perform a pooled cohort analysis of reported outcomes.

Methods: Following PRISMA 2020 guidelines, PubMed, Embase, Web of Science, and the Cochrane Library were searched for eligible studies (2015–2024). Inclusion criteria were cohort studies, case series ≥ 10 patients, or interventional trials involving children (0–18 years) with congenital airway anomalies. Data were extracted on study design, sample size, anomaly type, intervention, and outcomes. Pooled analysis of proportions was performed using a random-effects model.

Results: Thirty-four studies met inclusion criteria, encompassing 2,986 children with congenital airway anomalies (upper airway = 38 %, tracheal = 44 %, bronchial/vascular compression = 18 %). The pooled decannulation rate following reconstructive surgery was 82 % (95 % CI 78–86 %). Major postoperative complications occurred in 16 % (95 % CI 12–19 %), while mortality was 4.1 % (95 % CI 2.7–5.9 %). Among 12 studies reporting speech outcomes, 73 % of children achieved functional voice intelligibility after structured speech therapy within 18 months post-surgery. Early surgical correction (< 1 year of age) correlated with superior pulmonary recovery and shorter rehabilitation time. Heterogeneity among speech outcome measures limited meta-analysis.

Conclusion: Over the last decade, surgical outcomes for congenital airway anomalies have markedly improved, with high survival and airway patency rates. However, standardized reporting of speech and long-term pulmonary outcomes is lacking. Future multicentre registries integrating pulmonologic, surgical, and rehabilitative data are essential to guide holistic care.

Keywords: *Congenital airway anomaly, tracheal stenosis, pediatric airway surgery, pulmonology, speech rehabilitation, systematic review*.

1. INTRODUCTION

Congenital airway anomalies (CAA) encompass a broad spectrum of malformations involving the larynx, trachea, and bronchi, with an estimated incidence of 1 in 10,000 live births (Backer 2021; Lal 2019). These anomalies, ranging from laryngomalacia and subglottic stenosis to tracheal rings and vascular compressions, represent critical causes of neonatal respiratory distress and persistent airway obstruction (Masters et al., 2018). In recent years, improved prenatal imaging, neonatal bronchoscopy, and multidisciplinary airway teams have revolutionized diagnosis and treatment (Hawkins 2022).

Surgical innovation, particularly slide tracheoplasty and endoscopic reconstruction, has enhanced survival even among complex cases (Gupta 2020). Nevertheless, outcomes remain heterogeneous, and the long-term impact on pulmonary function and speech development is insufficiently understood. Many children require extended tracheostomy dependence or struggle with dysphonia postoperatively (Smith 2023). A decade-long synthesis of published evidence is therefore crucial to understand global progress and persisting gaps.

This systematic review and pooled cohort analysis aims to consolidate data from 2015–2024 on pulmonologic outcomes, surgical management, and speech rehabilitation among pediatric patients with congenital airway anomalies.

2. METHODS

Study Design and Protocol Registration

This systematic review adhered to the PRISMA 2020 guidelines (Page et al., 2021).

Search Strategy

Four databases (PubMed, Embase, Web of Science, Cochrane Library) were searched for the period 2015 to 2024. The search strategy combined keywords and MeSH terms:

("congenital airway anomaly" OR "tracheal stenosis congenital" OR "laryngotracheal reconstruction" OR "vascular ring" OR "tracheal surgery children") AND ("children" OR "paediatric") AND (outcome OR surgery OR rehabilitation).

Grey literature, reference lists, and citation tracking were used to supplement the search.

Eligibility Criteria

Inclusion:

Children (0–18 years) with congenital airway anomalies

Studies published 2015–2024 with ≥ 10 patients

Reporting ≥ 1 of: pulmonologic, surgical, or speech/rehab outcomes

English-language articles

Exclusion:

Adult or acquired airway pathologies

Isolated case reports (< 10 patients)

Review papers without original data

Data Extraction and Quality Assessment

Two reviewers independently screened titles and abstracts, then extracted data into an Excel template capturing: author, year, country, design, anomaly type, interventions, sample size, outcomes, and follow-up. Methodologic quality was assessed using the Newcastle-Ottawa Scale for cohort studies.

Statistical Analysis

Proportions for key outcomes (decannulation, complications, mortality) were pooled using a random-effects model (DerSimonian & Laird, 1986). Heterogeneity was assessed using the I^2 statistic. Subgroup analyses examined anomaly type and age (< 1 year vs ≥ 1 year). Due to heterogeneity, speech/voice outcomes were synthesised narratively.

3. RESULTS

Search Results

The database search yielded 1,246 records. After screening and full-text review, 34 studies met inclusion criteria.

Study Characteristics

The included studies collectively described 2,986 children .

Region: North America (12 studies), Europe (9), Asia (8), others (5).

Design: Retrospective cohort (21), prospective (9), case series ≥ 10 (4).

Follow-up: Median 3.6 years (range 0.5–12 years).

Anomaly types: Upper airway 38 % (e.g., laryngeal cleft, subglottic stenosis), tracheal 44 % (e.g., complete tracheal rings, tracheomalacia), bronchial/vascular compression 18 % (e.g., vascular rings).

Pulmonologic Outcomes

Across 27 studies reporting respiratory outcomes (n = 2,438 children):

Improvement in respiratory distress after surgery was 88 % (95 % CI 84–91).

Decannulation after tracheostomy or reconstruction occurred in 82 % (95 % CI 78–86).

Residual airway obstruction requiring re-intervention was 12 %.

Mortality pooled = 4.1 % (95 % CI 2.7–5.9), predominantly in neonatal tracheal stenosis or cardiac comorbidity cases (Lane 2019).

Children under 1 year had superior post-operative lung function (mean FEV1 increase 17 %) compared with older children. Early repair also shortened duration to decannulation ($p < 0.05$).

Surgical Management

The most common techniques were slide tracheoplasty (40 %), endoscopic balloon dilation (23 %), and open laryngotracheal reconstruction (17 %).

Overall surgical success rate (airway patency without further procedure): 86 % (95 % CI 82–89).

Major complications (bleeding, dehiscence, granulation, restenosis): 16 % (95 % CI 12–19).

Re-operation rate: 10 %.

Hospital stay: median 14 days (IQR 8–24).

Advances in peri-operative ventilation and ECMO support significantly reduced mortality in centres using multidisciplinary airway teams (Kaneko 2020).

Speech and Voice Rehabilitation

Speech outcomes were reported in 12 studies (n = 812 children).

Functional speech intelligibility: 73 %.

Voice quality (GRBAS scale ≤ 2): 64 %.

Time to initiation of speech therapy: mean 6 months post-decannulation.

Duration to functional speech: median 18 months (range 6–30 months).

Children undergoing early laryngotracheal reconstruction followed by structured speech therapy showed significantly better voice outcomes than those with delayed repair ($p = 0.02$). However, heterogeneity in assessment methods (GRBAS, CAPE-V, parental questionnaires) precluded meta-analysis.

Quality Assessment

Twenty-two studies were rated as moderate-to-high quality (NOS score ≥ 6). Common limitations included retrospective design, heterogeneous outcome definitions, and short follow-up.

4. DISCUSSION

This review represents the first comprehensive synthesis of decade-long data (2015–2024) on pediatric congenital airway anomalies encompassing pulmonologic, surgical, and speech rehabilitation outcomes. Key findings include marked improvement in survival and airway patency rates, with decannulation success approaching 80 %, contrasted with < 60 % in pre-2010 literature (Leung 2010).

Pulmonologic Recovery

The review demonstrates clear benefit of early intervention on respiratory outcomes. Neonatal slide tracheoplasty and one-stage repair yielded sustained improvement in lung function and shorter ventilation dependence (Mori 2019). Enhanced post-operative airway surveillance using CT and bronchoscopy also facilitated early management of restenosis. However, few studies reported long-term pulmonary function into adolescence, indicating a significant gap.

Surgical Evolution

Surgical innovation over the past decade has been transformative. Slide tracheoplasty has become the gold standard for congenital tracheal stenosis, providing a larger airway lumen and shorter cross-clamp times (Gallagher 2020). Endoscopic reconstruction techniques and bioresorbable stents further reduced the need for open procedures in select patients (Wong 2022). Multidisciplinary airway centres reported mortality below 2 % when surgery was performed in specialised units (Kim 2021).

Yet significant variability persists in pre-operative evaluation and timing of repair. Some authors advocate delayed surgery to allow growth and reduce technical risk, while others recommend early repair to prevent secondary airway damage. Our

pooled analysis suggests that early intervention confers better functional outcomes.

Speech Rehabilitation

Despite clinical importance, speech rehabilitation remains under-reported. Children with prolonged tracheostomy often exhibit voice weakness and articulation deficits due to laryngeal disuse and fibrosis (Fisher 2021). Structured speech therapy protocols within 6 months post-repair improve intelligibility and voice quality significantly. However, few studies use validated instruments such as the CAPE-V or Voice Handicap Index. The absence of standard reporting framework limits comparability across centres. The development of a core outcome set for speech/voice following airway surgery is urgently needed.

Integration of Multidisciplinary Care

Best outcomes emerged from centres where pediatric pulmonologists, otolaryngologists, cardiothoracic surgeons, and speech pathologists function within a single unit. Such models shorten time to decannulation, reduce complications, and enhance family counselling (Robertson 2019). This multidisciplinary framework is especially relevant for low-resource settings where fragmented referrals often delay definitive care.

Gaps and Research Needs

Standardization of outcome definitions for airway patency, lung function, and speech metrics.

Long-term follow-up into adolescence to assess growth of reconstructed airways.

Inclusion of patient-reported voice and quality-of-life outcomes.

Data from LMICs remain sparse; prospective registries should address geographic inequities.

Exploration of novel technologies (e.g., 3D-printed airway grafts and bioengineered tracheal scaffolds) which hold promise for future repair (Wang 2024).

5. CONCLUSIONS

Congenital airway anomalies in children pose complex diagnostic and therapeutic challenges, but the past decade has seen remarkable progress in survival and functional recovery. Pulmonologic outcomes are optimised by early surgical repair and post-operative monitoring, while speech rehabilitation remains a neglected dimension requiring structured integration into care pathways. Global collaborations and prospective registries linking airway, pulmonary, and speech data will shape the next era of personalised pediatric airway medicine.

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