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Review article

Primary dilatation as a treatment for pediatric laryngotracheal stenosis: A systematic review☆

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ABSTRACT

Objective: To identify and review original studies on balloon and rigid dilatation as primary therapy for laryngotracheal stenosis (LTS) in pediatric patients.
Design: Systematic review.
Methods: A comprehensive search strategy in MEDLINE, EMBASE and the Cochrane Central Register of Controlled Trials was conducted and limited to human studies published in English after 1980. Two independent reviewers identified original studies on primary dilatation therapy for LTS in patients younger than 18 years. Studies on tracheobronchial stenosis or stents for tracheomalacia were excluded. 22 of 369 identified studies (6%) met the inclusion criteria. Two reviewers independently appraised the level of evidence of each study, using the Oxford clinical evidence-based medicine guidelines, and extracted raw data using a standardized form developed a priori.
Results: The patient population consisted of grades I–III LTS. Most studies used adjuvant therapy including laser or topical agents. The primary outcome of success was achieving a functional airway without open laryngo-tracheal surgery or ongoing need for a tracheostomy. In studies using balloon dilatation alone (6 studies, $n = 10$) or rigid dilatation alone (5 studies, $n = 68$), success rates were 50% and 53%, respectively. Success rates ranged from 50% to 78% for balloon dilatation with adjuvant therapy (6 studies, $n = 24$) and 53%–100% for rigid dilatation with adjuvant therapy (5 studies, $n = 61$).
Conclusions: Dilatation was successful as primary therapy in the majority of low-grade pediatric LTS. Given the lack of comparative studies among other study limitations, it could not be determined whether one method of dilatation was superior to another.

Contents

1. Introduction

For the purpose of this study, laryngotracheal stenosis (LTS) describes narrowing of the airway that can be found anywhere from the true vocal cords to the distal trachea. It is usually acquired, but can be congenital. The most common cause of acquired LTS is trauma through prolonged endotracheal intubation. The incidence of post-intubation neonatal LTS has decreased

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over the past few decades. In the 1970s and 1980s, the incidence was as high as 8.3% and this decreased to 2% in the late 1990s. This downward trend may be due to improved techniques in managing the ventilation of neonates and decreased ventilation time [1]. Nevertheless, LTS still remains a major cause of airway obstruction in the pediatric population and, therefore, both endoscopic and open surgical methods have been developed to address this problem.

Historically, LTS was managed with endoscopic antegrade bouginage as early as the 1970s [2]. This method has gradually fallen out of favor since the shearing forces of the bougie predisposes to further scar tissue formation and restenosis. As a result, open surgery with laryngotracheal reconstruction has become the currently accepted as the standard of care for LTS. Endoscopic dilatation has recently regained popularity with the advent of the new angioplasty-type balloon catheter that was proposed to address the concern about restenosis. Modern balloons impart radially-directed force against the tracheal lumen, which is theoretically less traumatic to the laryngotracheal mucosa than the bougie [3]. Adjuvant therapies have included laser surgery, steroid injection, topical mitomycin C, and spray cryotherapy [4–6]. Currently, rigid dilators are still in use and balloon dilators are increasingly being used to treat low-grade LTS.

This study focuses on endoscopic dilatation techniques, which can be broadly classified into rigid dilatation (e.g. bougie, bronchoscope, tracheal dilator) and balloon dilatation (e.g. laryngeal balloon, angioplasty balloon, esophageal balloon). A systematic review on the effectiveness of rigid or balloon dilatation has not previously been conducted. The objective of this study is to investigate the evidence supporting the use of dilatation (with or without adjuvants) as primary therapy for LTS.

2. Methods

2.1. Study selection

A comprehensive search strategy in MEDLINE (from 1946), EMBASE (from 1980) and the Cochrane Central Register of Controlled Trials was conducted in April 2012 (see Fig. 1). The search terms used for dilatation were "dilat*", "balloon" and "bougi*". The search terms used for laryngotracheal were "laryngotrach*", "laryngeal", "glotti*", "subglotti*", "trach*" and "upper airway". The search terms used for stenosis were "stenosis" and "restenosis". The study population was specified using search terms "pediatric", "paediatric", "child*" and "infant*". These terms were combined with Boolean operators and 785 records were identified. These records were then limited to the English language, human subjects and a publication date after 1980. Once these limits were applied and duplicates were removed, 369 records remained.

Two reviewers independently assessed the titles and abstracts of the 369 studies identified by the search and applied specific eligibility criteria. Included studies consisted of original studies pertaining to dilatation as primary therapy for laryngotracheal stenosis in patients younger than 18 years. Studies pertaining to patients with tracheo-bronchial stenosis, the use of stents or Montgomery T-tubes were excluded. Studies on the laser or microdebrider as primary therapy were excluded because these did not exert manually directed forces, and therefore did not represent dilatation techniques. However, the laser or microdebrider, if used at the time of dilatation, were considered adjuvant therapy and these studies were included.

After reviewing 64 full-text articles, 42 were excluded for the following reasons: in twelve studies the patients had stenosis of at least one main bronchus; in twelve studies dilatation was not the primary therapy (dilatation was used in conjunction with primary open surgery); in nine studies the outcomes of the pediatric patients could not be separated from those of the adult patients, or age could not be determined; two studies were not primary; three studies did not specify the type of endoscopic procedure; and 4 studies used stents or Montgomery T-tubes. Therefore, after the inclusion and exclusion criteria were applied, 22 studies were included in the data analysis.

2.2. Data extraction

The same two reviewers independently extracted data from the included articles using a standardized data extraction form developed a priori. This form included data on patient demographics, the nature of the LTS, its management and outcomes. The evidence presented in the selected studies was categorized by level of evidence as defined by the Oxford center for evidence-based medicine (CEBM) [7].

The data was categorized into 4 methods of dilatation: (i) balloon dilatation; (ii) balloon dilatation and adjuvant therapy; (iii) rigid dilatation; (iv) rigid dilatation and adjuvant therapy. If the

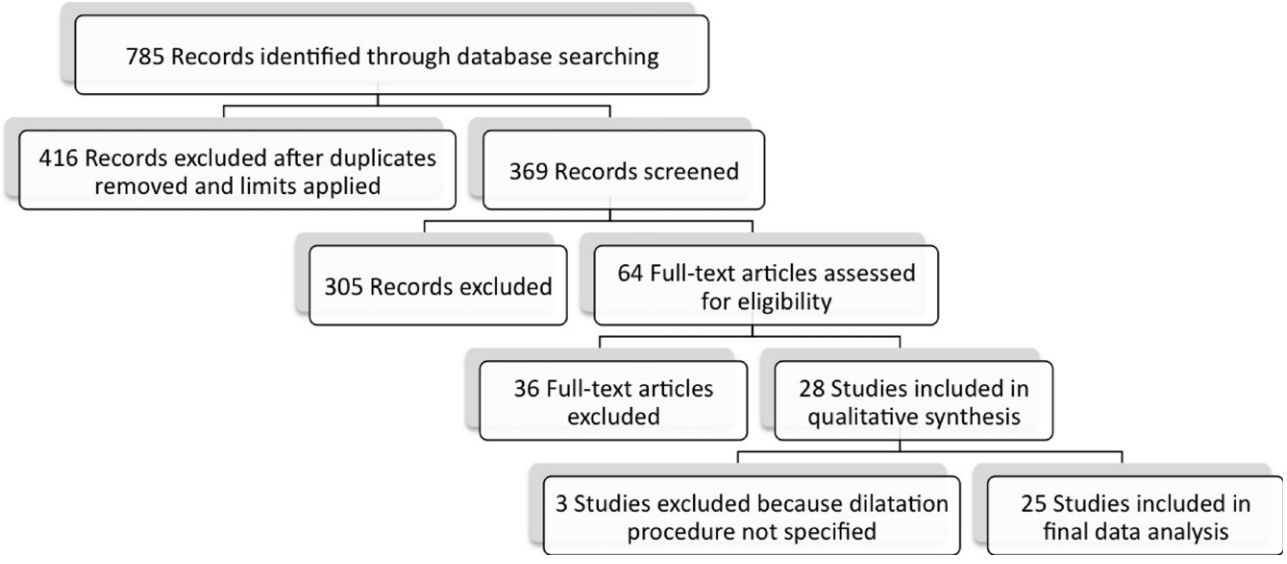

Fig. 1. Study selection process.