Surgical Management of Velopharyngeal Dysfunction in Patients with Cleft Palate: A Systematic Review

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Abstract:
Aim: A wide array of surgical techniques is used to treat post-palatoplasty VPI; each technique has its own advantages, limitations and drawbacks. The aim of this study is to review current literature regarding the speech outcome of different techniques for treatment of VPD in non-syndromic cleft patients.

Methods and Material: A systematic review was carried out according to PRISMA-P guidelines by searching Medline, Scopus and Web of Science Core Collection electronic databases. Main outcome measured: pre-operative and post-operative speech assessment.

Results: 25 relevant studies were retrieved, consisting of data on 1,175 patients. Overall there was 73% improvement in postoperative speech scores. A variety of scales were used for speech assessment. There was no notable difference between techniques with respect to speech outcome.

Discussion: There is a lack of high quality studies in this field. A standardized way for reporting speech outcome is needed.

Keywords: Velopharyngeal dysfunction, Cleft palate surgery, Speech.

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Introduction:
Cleft palate is one of the most common causes of velopharyngeal dysfunction (VPD). VPD is defined as the inability of the velopharyngeal valve (VPV) to close properly, due to inadequate function of dynamic structures that work to control it. The velopharyngeal valve is created by the soft palate or velum, the lateral pharyngeal walls, and the posterior pharyngeal wall. VPD affects multiple functions, with speech being the most critical. (1)(2) Despite best attempts, primary palatal repair only achieves normal speech in 70%–80% of individuals with cleft palate. That is, 20%–30% of children born with cleft palate will require secondary surgeries to manage VPD. (3) A wide array of surgical techniques is used to treat post-palatoplasty VPI; each technique has its own advantages, limitations and drawbacks. These techniques can be classified into two major groups; palatal surgery and extra-palatine surgeries. (7) Treatment options are chosen based on the condition of the palate,velopharyngeal function as shown by nasendoscopy and/or videofluoroscopy, and surgeon preference. (4) Palatoplasties are considered dynamic procedures as they result in an enhanced palatal mobility and function. (5)(6) Up to this point, there is no one operative procedure that is suitable for all cases, this is Last, articles yielded by abstract due to the various clinical presentations of VPI screening was reviewed by one reviewer in cleft patients. So far, no attempt was done to formulate a treatment algorithm that would consider all these decisive factors, and decide upon them the appropriate surgical treatment. The aim of this study is to review current literature regarding the speech outcome of different techniques of surgical treatment of VPD in cleft patients, in an attempt to reach a treatment algorithm.

MATERIALS & METHODS
This review followed the PRISMA- P guidelines for conduct of a systematic review, and protocol registered on the PROSPERO register of systematic reviews. (7) Search strategy
A thorough literature review was done by searching Medline, Scopus and Web of Science Core Collection electronic databases using the following keywords “cleft palate surgery”, “velopharyngeal insufficiency” and “speech”.

Study selection
Studies were included according to these criteria: English language, studies on patients undergoing surgery for management of VPD. A minimum of six months’ postoperative follow-up at least. Pre and post-operative speech assessment. Surgeries performed on patients three years old or above. Case reports, letters or commentaries were excluded, as well as, studies that did not meet the inclusion criteria.

Data Extraction
Data extraction was done manually using Microsoft Excel. Surgical techniques were categorized into two major different techniques of surgical treatment of VPD in cleft patients, in an attempt to reach a treatment algorithm.

Studies’ screening was done in three considered dynamic procedures as they result stages: title screening, second, one in an enhanced palatal mobility and function. reviewer (A. A.) reviewed all abstracts. (5)(6) Up to this point, there is no one operative with application of inclusion criteria procedure that is suitable for all cases, this is Last, articles yielded by abstract due to the various clinical presentations of VPI screening was reviewed by one reviewer in cleft patients. So far, no attempt was done to (A. A.) and their references were searched for any relevant articles.
records for analysis, therefore the 25 articles included increased to 28 records.

**Data Synthesis**
Tables were used to summarize study characteristics and outcomes. Surgical outcomes and complications were calculated manually as a ratio of the number of subjects with the defined outcome and the total number of patients in each study. To report outcome of different techniques, mean of the outcome of individual studies was manually calculated.

**Assessment of Risk of Bias**
The quality of each study was assessed for appraisal of potential risk of bias using the methodological index for non-randomized studies (MINORS) tool (8).

**Results**

**Study selection**
Study selection process followed PRISMA flowchart.

**Study design and participants**
The majority of studies reported prospective cohort, followed by retrospective series and cross-sectional studies. The total number of patients extracted from the 25 studies included in the review was 1,175.

**Study Participants**
All included articles reported secondary surgeries for speech in non-syndromic patients with history of cleft palate repair. Age at surgery ranged from 3-58 years.

**Study procedures**
Surgical techniques included in this review were; palatal re-repair, palatal lengthening with buccal myo-mucosal flap, double opposing z-plasty, PPW augmentation, pharyngeal flaps, and combined techniques; palate re-repair with buccal myo-mucosal flap and double opposing z-plasty combined with superiorly based pharyngeal flap.

**Surgical outcome**
The palatal re-repair group showed 85% and 66% improvement in resonance and nasal emission post-operatively respectively. (5) The BMMF group showed 82% and 70% improvement in resonance and nasal emission respectively (9) Double opposing z-plasty group showed 70%, 77% and 65% improvement in resonance, nasal emission and articulation respectively. Pharyngeal wall augmentation group showed 50% and 60% improvement in resonance and nasal emission respectively. Pharyngeal flap and pharyngoplasty group showed 85%, 69% and 72% improvement in resonance, nasal emission and articulation respectively.

**Risk of bias assessment:**
Regarding selection bias, none of the studies included were randomized; they comprised prospective cohort, retrospective series and cross-sectional studies, all Level IV evidence. As for detection bias, only 44% studies reported blinded assessment. All studies selected patients according to a specified inclusion criteria and none included all patients admitted to the center. Furthermore, attrition bias, all studies included subjects to whom they had complete records, therefore percentage of patients lost to follow up was not reported. Quality assessment was measured by assigning scores to each study according to MINORS items.

**Discussion**
The current review aimed to enhance care provided to our cleft palate patients.
This was done by reviewing articles discussing different surgical techniques used for treatment of VPD. Factors affecting technique choice and success are of paramount importance. These can be categorized into factors related to patients’ demographics and others related to VP function. According to the available literature cleft type and gender showed no strong correlation with the outcome of different techniques. (10)(11)(12)

When it comes to age the following aspects should be considered; age at which the primary surgery was performed, age of speech assessment and timing of the secondary speech surgery. There was no significant correlation between timing of the primary surgery and speech results of secondary procedures, although delay in primary repair would consequently result in more compensatory errors, the post-operative nasality score of the VPI surgeries was not related to it. (12)

Timing of the VPI surgery is a controversial point. The decision to perform the surgery depends on balancing a lot of factors. Firstly, the social life of the child; in cases where the child is near school age it is sometimes crucial for his self-confidence to perform the surgery as early as possible to allow for training and speech enhancement before school. Another factor is the maxillary growth, in patient with hypoplastic maxillae, it is better to postpone surgery to allow for non-restricted growth. Thirdly, some techniques have proven to be more successful in older ages, like the pharyngeal flap and the palatal re-repair. This could be attributed to the bigger anatomy and ease of surgical manipulation(13)(11)(14) (15) In case of a scarred palate with anteriorly tethered muscle, muscle re-repair with scar release can suffice, where in a more scarred and shorter palate, the addition of a buccal flap could be mandatory. This is also indicated if intraoperatively the bulk of the dissected muscles was found to be insufficient. (12,16) In cases of a short palate with adequately repaired muscles and minimum scar tissue, a more aggressive treatment by an extra-palatine flap would be employed. The VP gap is a crucial factor to consider when planning the VPI surgery.

Conclusion:
Choice of surgical procedure for VPD presents an ongoing challenge to cleft surgeons, no significant difference in outcome across the various procedures currently in standard practice. There is an urgent need for the multidisciplinary VPD community to develop a universally applicable, standardized minimum data set to record postoperative speech.

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Conflict of Interest: None.

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