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The Rate of Secondary Speech Surgery After Cleft Palate Repair: A Systematic Review

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Background: Reported rates of velopharyngeal insufficiency (VPI) after primary palatoplasty for cleft palate (CP) range from 5% to 30%. Although some cases are managed with speech therapy, many patients with VPI require surgical intervention. In this study, we investigate the rate of VPI surgery in patients with CP.
Methods: A systematic review was undertaken following PRISMA guidelines. PubMed, Scopus, and Cochrane databases were used. Studies reporting the rate of VPI surgery in patients with CP were included. Studies containing patients with submucous clefts and/or lacking 6 months follow-up were excluded. Rates of VPI surgery were calculated through weighted means.
Results: Fifty-eight articles were included. The overall rate of VPI surgery was 17.5% ± 9.2% (range, 0%–59%). When stratified by phenotype, the rate of VPI surgery was 20.0% ± 13.1% for unilateral cleft lip and palate (range, 0%–39.6%), 27.1% ± 17.2% for bilateral cleft lip and palate (range, 0%–59%), and 14.4% ± 7.2% for isolated CP (range, 0%–47.4%, $P > 0.05$). When segregated by the palatoplasty technique, the surgical rate was 7.2% ± 3.7% for Furlow, 20.3% ± 19.8% for 2-flap, 5.0% ± 2.8% for Sommerlad, and 23.4% ± 8.0% for 2-stage ($P > 0.05$). Of studies reporting VPI assessment criteria, speech pathology assessment alone (n = 11, 34.4%) was the most common.
Conclusions: Significant variability exists in reported rates of VPI surgery after CP repair. Initial results suggest a higher rate of VPI surgery in association with certain phenotypes and repair techniques, but data are insufficient for robust conclusions.
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INTRODUCTION

Cleft lip and palate (CLP) is the most common congenital anomaly of the head and neck.[1–3] Primary repair of the palatal defect in patients with CLP is aimed toward normalizing speech and eliminating oronasal regurgitation.[4,5] Velopharyngeal insufficiency (VPI) is a major complication of primary palatoplasty, manifesting as hypernasal speech and compensatory articulation errors.[6] Because intelligible speech is a fundamental component of normal childhood development, minimizing the risk of VPI is of utmost importance to cleft surgeons.[7,8]

Rates of VPI after primary palatoplasty are well reported but highly variable in the current literature, ranging from 5% to 30%.[9–12] Several risk factors for VPI have been identified, including cleft severity, palatoplasty technique, and surgical timing.[9–12] Variations in the published rates of postpalatoplasty VPI are largely due to heterogeneity in the clinical criteria and evaluation methods for its diagnosis. Diagnostic tools range from subjective speech assessment to videofluoroscopy to nasoendoscopy, each of which has distinct criteria for the assessment of VPI.[13,14]

The lack of standardization in VPI diagnostic criteria has translated to a lack of standardization in VPI management. Strategies for the management of VPI include speech therapy and numerous surgical interventions,

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including revision palatoplasty, fat grafting, sphincter pharyngoplasty, and pharyngeal flaps. However, algorithms for pursuing surgical versus nonsurgical management of VPI vary widely by institution.[15,16] As such, the overall rate of surgical intervention for postpalatoplasty VPI is highly variable and poorly understood. Understanding this rate has important implications for clinical care and resource management, especially in low-income settings where secondary surgery may not be routinely available.

The primary objective of this study was to identify the incidence of VPI surgery after primary palatoplasty in patients with CLP. The secondary objective was risk stratification by cleft phenotype and palatoplasty technique.

Takeaways

Question: What is the overall rate of secondary surgery to correct velopharyngeal insufficiency among patients with cleft palate?

Findings: We included 58 articles in this systematic review. The overall rate of secondary surgery was 17.5% with a range of 0%–59%. Patients with bilateral cleft lip and palate had the highest rate of surgery at 27.1%, but this was not significant compared with other phenotypes.

Meaning: Current literature reports a wide range of rates for speech surgery, potentially due to different assessment criteria. More data are required to reach solid conclusions.

METHODS

A systematic literature review was conducted according to Preferred Reporting Items for Systematic Reviews and Meta-analyses 2020 guidelines.[17] The PubMed, Scopus, and Cochrane databases were reviewed for primary and secondary sources of evidence. The following keywords and terms were used in multiple combinations: "VPI," "surgery," "rate," "velopharyngeal insufficiency," "cleft palate," "speech surgery," "revision rate," "cleft palate repair," "long term," "speech outcomes," and "protocol." Studies that reported the rate of revision surgery for VPI after primary palatoplasty were included. Studies that included patients with submucous clefts and/or lacked 6-month follow-up were excluded. Furthermore, when 2 studies that otherwise met the criteria were found to report on the same cohort of patients, the study with shorter follow-up was excluded. (**See appendix, Supplemental Digital Content 1,** which displays the Preferred Reporting Items for Systematic Reviews and Meta-analyses checklist, **http://links.lww.com/PRSGO/D793**.)

Studies were first screened by title and abstract by 2 authors (A.T.P. and D.G.C.). The same 2 authors then screened full-text articles and periodically cross-referenced work to ensure the accuracy of data collection. No disagreements about study inclusion occurred. Data extracted from selected articles included: year of publication, country of publication, income status (World Bank), type of study, rating according to the Newcastle-Ottawa scoring system, study time period, and total number of patients.[18] The length of follow-up, age at primary palatoplasty, number of revision procedures, number of patients per cleft phenotype, primary palatoplasty technique, criteria for VPI surgery, rate of palatal fistulas, percentage of patients who had speech therapy, and VPI revision surgery technique were also collected.

All data were curated in Microsoft Office Excel Version 2406 (Microsoft Corporation Redmond, WA). A weighted analysis was performed to calculate the overall VPI surgery rate across all included studies. A 2-sided Student *t* test was used to compare surgical rates between phenotypes, and analysis of variance was used for comparison among primary palatoplasty techniques. The Pearson correlation coefficient and simple linear regression were used to assess the relationship between age at primary palatoplasty and the rate of VPI surgery. All statistical analysis was conducted in R Statistical Software (version 4.3.2; R Core Team 2024).

RESULTS

The database search strategy identified 6353 unique publications. Of these, 58 publications met the selection criteria and were included for review (Fig. 1).[9,17,19–75]

The studies were published between 1995 and 2024. The country with the highest number of publications was the United States (n = 17, 29.3%) followed by Sweden (n = 8, 13.8%; Table 1). Grouped by World Bank income status, 94.8% (n = 55) of publications were from high-income countries and 5.2% (n = 3) were from low- and middle-income countries.[76] The median Newcastle-Ottawa score was 6 (interquartile range, 0; range, 5–8).

The studies accounted for a total of 16,725 patients, with a median of 103 patients per study (IQR, 223; range, 21–7325). Of the 42 studies that reported age at primary palatoplasty, patients had a mean age of 10.8 ± 4.1 months. No relationship was found between the age of primary palatoplasty and the rate of VPI surgery on the calculation of correlation coefficient (0.05) or linear regression ($P = 0.76$). The mean duration of follow-up was 8.6 ± 6.0 years.

The overall rate of VPI surgery among all patients was 17.5% ± 9.2% (range, 0%–59%). Only 14 (37.8%) studies included a single technique for VPI surgery, whereas 23 (62.2%) studies included multiple techniques. Among the studies including a single VPI surgery technique, the most common was pharyngeal flap (n = 11, 85.7%), followed by Furlow palatoplasty (n = 1, 7.1%). Across studies reporting multiple, data were not reported in enough detail to further stratify by individual techniques.

The rate of palatal fistulas reported by 49 (84.5%) studies, the overall fistula rate across all studies was 8.7% ± 7.1% (range, 0%–70%). Only 3 studies (5.2%) detailed their indications and protocol for speech therapy, whereas 12 (20.7%) studies reported the percentage of patients who underwent speech therapy. Of these 12 studies, the overall rate of speech therapy was 36.5% ± 23.4% (range, 5.6%–81.8%).

Evaluation methods for diagnosing VPI were reported by 32 (55.2%) studies. The most frequently reported

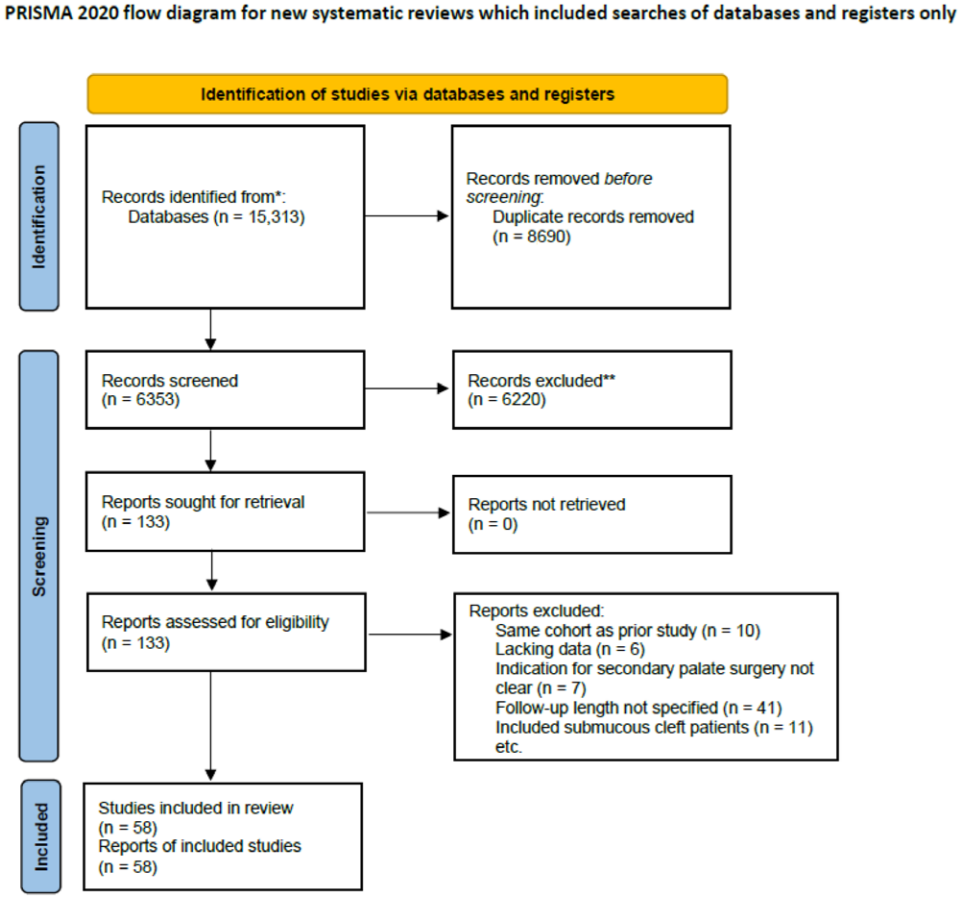

Fig. 1. Preferred Reporting Items for Systematic Reviews and Meta-analyses flow diagram. *Databases used: Pubmed, Cochrane, and Scopus. **Records excluded following removal of duplicates.

Table 1. Number of Included Studies by Country of Publication

Country	No. Studies	Income Status (World Bank)	Country	No. Studies	Income Status (World Bank)
Australia	1	HIC	New Zealand	1	HIC
Belgium	1	HIC	Norway	1	HIC
Canada	2	HIC	Poland	2	HIC
Denmark	1	HIC	Singapore	1	HIC
Egypt	2	LMIC	South Korea	3	HIC
Finland	6	HIC	Sweden	8	HIC
Germany	1	HIC	Switzerland	2	HIC
India	1	LMIC	Taiwan	3	HIC
Italy	1	HIC	United Kingdom	2	HIC
Japan	1	HIC	United States	17	HIC
The Netherlands	1	HIC	—	—	—

HIC, high-income country; LMIC, low- and middle-income country.

method was subjective speech evaluation alone (n = 11, 34.4%). The second most common method was subjective speech evaluation with nasoendoscopy (n = 6, 18.8%; Table 2).

In terms of cleft phenotype, 21 (36.2%) studies reported on patients with unilateral CLP (UCLP) (Table 3), 16 (27.6%) studies reported on patients with bilateral CLP (BCLP) (Table 4), 22 (37.9%) studies reported on patients with isolated cleft palate (ICP) (Table 5), and 12 (20.7%) studies reported on multiple phenotypes. Aggregated by phenotype, there were a total of 1944 (11.6%) patients with UCLP, 851 (5.1%) patients with BCLP, and 6666 (39.9%) patients with ICP, leaving 7264 (43.4%) patients as unclassified. The overall rates of VPI surgery by cleft phenotype were calculated as weighted means as follows: 20.0% ± 13.1% for UCLP (range, 0%–39.6%), 27.1% ± 17.2% for BCLP (range, 0%–59%), and 14.4% ± 7.2% for ICP (range, 0%–47.4%). Statistical testing revealed no significant differences between patients with UCLP and BCLP (t = 1.82, P = 0.08), UCLP and ICP (t = –0.41, P = 0.68), or BCLP and ICP (t = 1.85, P = 0.07).

Table 2. Describing the VPI Assessment Protocols of Included Studies

VPI Assessment Protocol	No. Studies
SLP	11
Nasoendoscopy	4
SLP + nasoendoscopy	6
SLP + nasometry + videofluoroscopy	3
SLP + videofluoroscopy	2
SLP + nasometer ± nasoendoscopy	1
SLP + nasometry	1
SLP + nasoendoscopy + videofluorography + aerodynamic assessment + nasometry	1
SLP + nasoendoscopy + videofluoroscopy	1
Team consensus	1
Team consensus + nasoendoscopy	1

SLP, speech and language pathology assessment.

In terms of the primary palatoplasty technique, 31 (53.4%) studies reported using a single technique and 25 (43.1%) studies reported using multiple techniques. Of the institutions using a single technique, the most popular was the Furlow (n = 8, 25.8%) followed by the Bardach technique (n = 5, 16.1%; Table 6). Of the techniques amenable to analysis, 2-stage palatoplasty demonstrated the highest rate of VPI surgery (23.4% ± 8.0%), whereas the Sommerlad technique demonstrated the lowest rate of VPI surgery (5.0% ± 2.8%). No significant difference was found in the VPI surgery rate between primary techniques ($P = 0.09$).

DISCUSSION

Developing a gold-standard protocol for mitigating complications after cleft palate repair has been a long-standing global pursuit. Understanding the predictors of VPI surgery is central to driving consensus toward such a protocol. In this study, we found that an average of 17.5% of patients require VPI surgery after primary palatoplasty. Previously published systematic reviews report a VPI surgery rate ranging from 0% to 29.2%.[77,78] Our findings suggest a high risk of surgical VPI, which must be incorporated into the preoperative counseling of patients undergoing primary palatoplasty.

The overall rate of postpalatoplasty VPI reported in the literature is 20%–30%.[10,11,79,80] As some patients will not require surgical management for their VPI, this figure is appropriately higher than the 17.5% rate of VPI surgery found in this study. Taken together, these figures suggest that a vast majority of postpalatoplasty VPI is managed surgically. However, because the VPI surgery rate is often used as a proxy for the VPI rate, previously published figures are likely to be subject to significant confounding.[79] We recommend that future investigations prioritize the involvement of speech pathologists and use data from clinical speech assessment to improve the accuracy of reported VPI rates within the literature. Furthermore, because the reporting speech therapy and nonsurgical management of VPI was poor within the included studies, we were unable to perform a more rigorous analysis of the relative risks of surgical versus nonsurgical VPI in patients with CLP.

We found no association between the age at primary palatoplasty and the rate of VPI surgery. However, this result needs to be interpreted with the context, that of the studies reporting mean age at palatoplasty, 39 (92.9%) reported repair before the 18-month cutoff currently recommended by the American Cleft Palate Craniofacial Association. Our results support this current cutoff as they suggest minimal difference between VPI surgery rates,

Table 3. Detailed VPI Surgery Rate and Surgical Protocols Among Studies for Patients With UCLP

First Author	Total Patients With UCLP	Total VPI Procedures	Percentage VPI Procedures	Specified Protocol Repair Timing	Mean Age at Repair, mo	Surgical Technique
Aboul-Wafa[20]	8	0	0	No	15.5	Modified Bardach
Black[28]	10	0	0	No	12.7	Multiple
Botticelli[29]	106	2	1.9	Yes, 3–4 mo	—	Multiple
Brudnicki[30]	56	2	3.6	No	8.1	Own technique
Brusati[31]	40	1	2.5	Yes, 6 mo	6	Pigott
Damalachervu[32]	66	1	1.5	Yes, <60 mo	—	Modified Furlow
de Buys Roessingh[33]	34	3	8.8	Yes, 3 mo	3.2	Malek
Gustafsson[39]	290	88	30.3	Yes, 10–12 mo	10.9	Multiple
Ha[42]	41	11	26.8	Yes, 12 mo	12	Multiple
Hattori[43]	205	24	11.7	Yes, 9–12 mo	—	Modified Furlow
Hortis-Dzierzbicka[46]	28	1	3.6	No	8.8	Modified intra-velar veloplasty
Kappen[48]	48	19	39.6	No	7.8	Two-stage
Klinto[49]	33	3	9	Yes, 12 mo	—	Sommerlad
Lohmander[55]	55	6	10.9	No	7.5	Two-stage
Mommaerts[59]	21	0	0	No	12	Modified Furlow
Moren[60]	73	11	15.1	No	21	Multiple
Okhiria[61]	86	21	24.4	Yes, 6 mo	7.2	Two-stage
Park[64]	56	13	23.2	Yes, 18 mo	19.3	Pushback
Phua[67]	61	11	18.0	No	13	Multiple
Rautio[68]	448	160	35.7	No	—	Multiple
Salyer[70]	179	12	6.7	No	9.4	Two-flap

Table 4. Detailed VPI Surgery Rate and Surgical Protocols Among Studies for Patients With BCLP

First Author	Total BCLP Patients	Total VPI Procedures	Percentage VPI Procedures	Specified Protocol Repair Timing	Mean Age at Repair, mo	Surgical Technique
Aboul-Wafa[20]	16	0	0	No	15.5	Modified Bardach
Al-Dourobi[21]	30	11	36.7	Yes, 3 mo	3	von Langenbeck
Black[28]	10	1	10	No	12.7	Multiple
Damalachervu[32]	18	1	5.6	Yes, <60 mo	—	Modified Furlow
Gustafsson[38]	81	38	46.9	Yes, 10–12 mo	10.6	Multiple
Ha[42]	94	26	27.7	Yes, 12 mo	12	Multiple
Hattori[43]	71	9	12.7	Yes, 9–12 mo	—	Modified Furlow
Hattori[44]	122	72	59.0	Yes, 12 mo	12.3	Bardach
Klinto[49]	17	3	17.6	Yes, 12 mo	—	Sommerlad
Koh[50]	52	8	15.4	No	11.9	Multiple
Mommaerts[59]	11	0	0	No	12	Modified Furlow
Okhiria[61]	37	10	27.0	Yes, 6 mo	7	Two-stage
Park[64]	20	7	35.0	Yes, 18 mo	19.3	Pushback
Phua[67]	32	4	12.5	No	13	Multiple
Salyer[70]	79	8	10.1	No	9.4	Two-flap
Trotter[74]	161	33	20.5	Yes, 10–12 mo	13.7	Multiple

Table 5. Detailed VPI Surgery Rate and Surgical Protocols Among Studies for Patients With ICP

First Author	Total Patients With BCLP	Total VPI Procedures	Percentage VPI Procedures	Specified Protocol Repair Timing	Mean Age at Repair, mo	Surgical Technique
Abdel-Aziz[19]	21	0	0	Yes, <6 mo	4	Furlow
Aboul-Wafa[20]	12	0	0	No	15.5	Modified Bardach
Andersson[22]	351	68	19.4	Yes, 12–36 mo	—	Modified von Langenbeck
Bae[24]	100	1	1	No	12.3	Busan modification
Becker[26]	66	14	21.2	Yes, 7–18 mo	—	Multiple
Black[28]	15	0	0	No	12.7	Multiple
Damalachervu[32]	39	0	0	Yes, <60 mo	—	Modified Furlow
Elander[34]	94	16	17.0	Yes, 6 mo	7.7	Pushback
Evans[35]	273	13	4.8	Yes, 6–12 mo	13	Furlow
Goudy[37]	63	13	20.6	No	—	Three-flap
Gustafsson[41]	423	141	33.3	Yes, 9–12 mo	9	Multiple
Gustafsson[40]	78	37	47.4	No	10	Multiple
Ha[42]	157	19	12.1	Yes, 12 mo	12	Multiple
Hattori[43]	200	7	3.5	Yes, 9–12 mo	—	Modified Furlow
Jodeh[47]*	4239	559	13.2	No	11.4	Multiple
Klinto[49]	30	4	13.3	Yes, 12 mo	—	Sommerlad
Mommaerts[59]	13	0	0	No	12	Modified Furlow
Okhiria[61]	90	15	16.7	Yes, 6 mo	7.2	Two-stage
Park[64]	64	8	12.5	Yes, 18 mo	19.3	Pushback
Patel[65]	96	16	16.7	No	11	—
Phua[67]	118	13	11.0	No	12	Multiple
Salyer[70]	124	14	11.3	No	9.4	Two-flap

*Study was based on national database.

granted the threshold is met. Additional data, not available in included studies, would be required to determine the effect of delayed primary palatoplasty on VPI surgery rate.

In this study, we identified an association between VPI surgery rate and cleft phenotype (Tables 3–5). Specifically, patients with ICP had the lowest overall rate of VPI surgery (14.4% ± 7.2%), followed by patients with UCLP (20.0% ± 13.1%), followed by patients with BCLP (27.1% ± 17.2%). This is consistent with previously reported rates of VPI, which are also higher for patients with Veau III and IV clefts as compared with those with Veau I and II clefts.[53,81–83] However, upon statistical testing, no significant difference was found between phenotypes, likely due to the wide range of rates reported; more data are required to evaluate the initial differences demonstrated in this study. Despite the lack of statistical significance, our findings mirror clinical outcomes reported for other secondary surgical procedures in patients with cleft. For example, a review by Choi et al found that the likelihood of orthognathic surgery among patients with cleft was highest among patients with BCLP and lowest among patients with ICP.[77,84] These patterns are all consistent with the greater technical difficulty of repairing Veau III and IV clefts, in which a paucity of soft tissue complicates the restoration of palatal form and function. Consideration of cleft phenotype is

Table 6. Detailed Rates of VPI Surgery From Studies That Reported Use of a Singular Surgical Technique

Technique	No. Studies	Total Patients	Range VPI Surgery Rates, %	Weighted Mean VPI Surgery Rates, %	Weighted SD
Bardach/2-flap	5	694	0–59.0	20.3	19.8
Furlow	8	1481	0–14.7	7.2	3.7
Intra-velar veloplasty	2	425	2.8–3.6	2.8	0.2
Malek	1	34	—	8.8*	—
Pigott	1	40	—	2.5*	—
Pushback	2	234	17.0–20.0	18.8	1.5
Sommerlad	3	913	2.6–12.5	5.0	2.8
Three-flap	1	63	—	20.6*	—
Two-stage	4	337	10.9–39.6	23.4	8.0
von Langenbeck	2	381	19.4–36.7	20.7	4.7
Own modification					
Bae	1	100	—	1.0*	—
Brudnicki	1	56	—	3.6*	—

*Reporting of singular study VPI surgery rate because the weighted mean was not calculable.

therefore critical to the surgical planning and risk assessment of patients who underwent palatoplasty.

In addition to cleft phenotype, we also identified an association between VPI surgery rate and palatoplasty technique (Table 5). Only 4 primary palatoplasty techniques were reported more than twice within the included studies: Furlow, Bardach, Sommerlad, and 2-stage. The most popular among these was the Furlow (n = 8), which was associated with a VPI surgery rate of 7.2% ± 3.7%. The Sommerlad technique (n = 3) demonstrated the lowest rate of VPI surgery (5.0% ± 2.8%), whereas the 2-stage technique (n = 4) demonstrated the highest (23.4% ± 8.0%). No significance was found upon comparison, but this may be attributed to the low number of studies available. Previous literature corroborates our findings, reporting lower VPI revision rates for both Furlow (5.1%) and Sommerlad (4.6%–10.2%) palatoplasties.[9,85,86] The palatal lengthening achieved by the Furlow technique and the meticulous muscle dissection involved in the Sommerlad technique are both thought to contribute to their favorable risk profiles.[87] On the other hand, centers that use 2-stage palatoplasty postulate that staging leads to improved facial growth and decreased risk of midface hypoplasia.[81–83] Our findings suggest that their supposed preservation of midface growth might come at the expense of speech outcomes. However, because exact palatoplasty techniques were not specified by some studies using the 2-stage approach, the exact mechanism of why this protocol is associated with an increased need for VPI surgery remains unclear.

Our findings must be interpreted within the context of numerous study limitations. First and foremost, only 55.2% of included studies reported their methods for evaluating VPI. Among those that reported VPI evaluation methods, significant variability was observed, with 11 studies using subjective speech exam alone. Furthermore, among those studies that used subjective speech exam alone, a multitude of validated speech assessment scales were reported.[88,89] The included studies were similarly lacking in standardization with regard to their consideration of primary palatoplasty techniques, cleft phenotypes, and follow-up duration.

Our study is also limited by its lack of consideration for cleft severity, a major predictor of postpalatoplasty complications such as VPI.[90–92] Although the data in this review were insufficient to assess the effect of cleft severity on VPI surgery, we expect that the greater risk of VPI associated with wider clefts portends an accordingly greater risk of VPI surgery. Another critical factor we were unable to assess was that of surgeon experience. The included studies did not report the length of time in surgical practice or the number of palatoplasties surgeons performed yearly, both of which undoubtedly affect outcomes.

Finally, an overwhelming majority of studies included in this review were derived from high-income country patient populations, limiting the applicability of our findings to low- and middle-income countries. Because low- and middle-income countries bear most of the global burden of cleft conditions, the reported rate of VPI surgery presented in this study represents just a small subset of cleft patients worldwide. Future studies that include multinational patient populations as well as more rigorous standardization of data reporting are required to improve our understanding of the risk factors for VPI surgery after primary palatoplasty.

CONCLUSIONS

The reported rate of VPI surgery after primary palatoplasty is highly variable within the current literature. Our study found that the rate of VPI surgery is 17.5% overall. Factors associated with higher rates of VPI include more severe cleft phenotypes and 2-stage palatoplasty when compared with other palatoplasty techniques. Future studies featuring larger and more diverse samples are required to improve our understanding of the need for secondary speech surgery.

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DISCLOSURE

The authors have no financial interest to declare in relation to the content of this article.

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