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Article

Effects of alveolar bone grafts vs. orthognathic surgery on cleft palate speech nasalance: a meta-analysis

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SUMMARY

Patients born with cleft palate frequently struggle with speech nasalance issues, such as hypernasality and hyponasality. Hypernasality results in patients having nasal speech, whereas hyponasality results in patients having a congested voice. To improve speech function, cleft palate patients may undergo numerous surgical interventions. Two of the most common secondary cleft repair surgeries are alveolar bone grafting and orthognathic surgery. The purpose of this meta-analysis was to determine whether one surgical intervention is more effective in addressing hypo- and hypernasality in cleft palate patients. We assessed three key outcomes: change in nasalance scores post-operation, percent change of patients with excess nasal emission post-operation, and presence of hypo/hypernasality post-operation. We hypothesized that alveolar bone grafts will be superior to orthognathic surgery for addressing speech nasalance. Results from nine studies conducted over 47 years revealed distinct outcomes for bone grafts and orthognathic surgery. Alveolar bone grafts were found to generally improve hypernasality, whereas orthognathic surgery resulted in worsened hypernasality. For each treatment type, both hypo- and hypernasality can be found in patients post-operation, with no statistical indication of immediate changes to hyponasality. Limitations to this analysis included the small sample size used in the component studies, so more research would be helpful for additional understanding. Studies used in the analysis consisted of both cohort studies and randomized controlled trials, resulting in some confounding effects. From a clinical perspective, this meta-analysis has the potential to inspire future studies comparing these surgical methods, which may ultimately enable craniofacial surgeons to enhance nasalance outcomes for individuals with cleft palates.

INTRODUCTION

A cleft palate is a congenital disorder diagnosed at birth when newborns have an opening in their upper palate. Occurring when palatal tissue is not completely developed during fetal growth, this condition impacts roughly 33 in every 100,000 newborns (1). Cleft palates vary in size and severity and can affect both the hard and soft palates. Unilateral clefts affect one side of the mouth, whereas bilateral clefts affect both sides (2). In addition to structural defects, cleft palate patients may encounter functional challenges with hearing, feeding, and speech (2).

The primary surgical intervention for cleft palate repair is palatoplasty, typically performed when patients are between 10 and 12 months old. This procedure aims to close the cleft palate, thereby separating the oral and nasal cavities. Palatoplasty is medically recommended for all patients born with cleft palate, as it sets the foundation for improved feeding and speech development (3).

In many cases, cleft palate patients require additional surgeries for functional purposes (3). Two of the most commonly performed secondary cleft repair procedures are alveolar bone grafts and orthognathic surgery. Alveolar bone grafts are performed in patients with clefts that extend into the alveolar ridge (hard palate). This procedure involves using synthetic or human hip bone to facilitate bone growth in the hard palate, thereby closing alveolar gaps (4, 5). Orthognathic surgery involves repositioning the upper or lower jaw to correct dental misalignment (6). LeFort jaw surgery, used to treat underbite through repositioning of the upper jaw, is the most common type of orthognathic surgery performed in cleft patients (7). Procedures such as bone grafts and orthognathic surgery have the potential to enhance long-term speech function in cleft patients.

Speech proficiency issues commonly observed in cleft palate patients include hyponasality and hypernasality, both of which affect speech nasalance. Hyponasality occurs during speech when there is not enough sound resonating in the nasal cavity. This can result in a patient's voice sounding congested and blocked. Hypernasality occurs during speech when there is too much sound resonating in the nasal cavity, resulting in nasal speech. The primary cause of hypernasality is excessive nasal emission, which occurs when too much air is released through the nose during speech. Among the two, hypernasality is more commonly observed in cleft palate patients (8). These issues can be attributed to velopharyngeal insufficiency, where the sphincter between oral and nasal cavities may not be closed completely (9). Speech nasalance in patients is more directly addressed with bone grafts, whereas nasalance after surgery is considered a side effect of orthognathic surgery (6, 8).

This meta-analysis aimed to compare the effectiveness of bone grafts and orthognathic surgery for improving speech nasalance in individuals with cleft palate. Specifically, our research question aimed to determine whether one surgical treatment is more effective than the other for addressing hypo/hypernasality in cleft palate patients. Because these procedures affect different areas of the oral cavity associated with speech nasalance, we hypothesized that alveolar bone grafts will be superior to orthognathic surgery for addressing nasalance in cleft palate patients. After collecting data from various studies, we focused on three specific outcomes

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for which there was sufficient data to analyze: change in nasalance scores post-operation, percent change of patients with excess nasal emission before and after surgery, and frequency of hypo/hypernasality post-operation. We found that alveolar bone grafts reduce hypernasality while orthognathic surgery increases hypernasality, giving insight into how each procedure affects speech patterns in cleft palate patients.

RESULTS

Change in Nasalance Scores Post-op

The meta-analysis for change in nasalance scores post operation consisted of three studies published between 2001 and 2010 (10-12). In total, the three studies included 50 cleft patients undergoing orthognathic surgery and 20 undergoing bone grafts. Oral nasalance scores measure the level of nasal resonance produced during speech, and range from a percentage value of 0 to 100. A lower nasalance score corresponds with decreased levels of nasal emission, which is generally better for patients with hypernasality. The average nasalance score for all cleft palate patients prior to undergoing surgery was 18.62. After undergoing bone graft, the average nasalance score of patients was 14.41, while after undergoing orthognathic surgery, the average nasalance score of patients was 30.30. The 2001 study by Bureau et al. consisted of two groups of cleft palate patients: patients born with bilateral cleft palate and patients born with unilateral cleft palate (10). Both groups of patients shared similar results for nasalance scores after bone graft. Patients who underwent bone grafts experienced an overall decrease in nasalance scores by an average of 3.84 ($p = 0.14$), whereas patients who underwent orthognathic surgery experienced an overall increase in nasalance scores by an average of 11.3 ($p = 0.05$). Each p-value in **Figure 1** was provided by the original study when determining the change in nasalance score. P-values were then used to calculate confidence intervals using binomial distribution. (**Figure 1**).

Change in Percentage of Patients with Excess Nasal Emission Post-op

The meta-analysis for change in percentage of patients with excess nasal emission post-op consisted of four studies published between 1976 and 2013 (13-16). In total, the studies consisted of 64 cleft patients who underwent bone grafts and 74 who underwent orthognathic surgery. On average, 66.33% of all cleft palate patients had excess nasal emission prior to undergoing surgery. After receiving a bone graft, an average of 52.25% of patients had excess nasal emission, and after undergoing orthognathic surgery, an average of 81% of patients had excess nasal emission. The percentage of patients with excess nasal emission decreased in the group that underwent bone grafts by an average of 23.9%, whereas the percentage of patients with excess nasal emission increased in the group that underwent orthognathic surgery by an average of 24.5%. Trials conducted for both alveolar bone grafts and orthognathic surgery fall within the 95% confidence interval, indicating a statistically significant difference between outcomes for both procedures (**Figure 2**).

Frequency of Hypo/Hypernasality Post-op

The meta-analysis for the frequency of hypo/hypernasality post-op consisted of two studies published in 1998 and 2023 (17, 18). In total, the studies consisted of 56 cleft patients undergoing bone grafts and 40 undergoing orthognathic surgery. Neither study provided information on the number of patients with hypo/hypernasality prior to surgery. After orthognathic surgery, 42.5% of patients had either hypo- or hypernasality, while after alveolar bone grafts, 41.1% had either hypo- or hypernasality. 17.5% of patients had hyponasality after orthognathic surgery, while 12.5% of patients had hyponasality after alveolar bone grafts. 25% of patients had hypernasality after orthognathic surgery, and 28.6% of patients had hypernasality after both graft. Therefore, bone grafts resulted in a higher percentage of patients with hypernasality (3.6% higher), while orthognathic surgery resulted in a higher percentage of patients with hyponasality (5.0% higher). All trials fell within the 5% margin of error, meaning that only trends could be observed from these specific studies (**Figure 3**).

DISCUSSION

In this study, we conducted a meta-analysis to compare the effects of alveolar bone graft and orthognathic surgery on speech nasalance in cleft palate patients. Data collected from nine studies spanning 47 years provided a comprehensive understanding of the post-operative outcomes associated with each surgical intervention.

Through the meta-analysis conducted for change in nasalance scores post-op, we concluded that alveolar bone grafts are better suited for reducing nasal emission in cleft palate patients. In other words, bone grafts improved hypernasality when compared to orthognathic surgery. Across three studies, patients undergoing bone grafts experienced an overall decrease in nasal scores, indicating a reduction in nasal resonance. We can be confident that nasalance scores decreased after alveolar bone graft with 95% confidence. On the other hand, patients who underwent orthognathic surgery experienced a significant increase in nasalance scores, signifying an increase in nasal resonance.

Study	SX Type	Pre	Post	MD
Bureau (10)	Bone Graft	18.64	14.27	-4.37
Bureau (10)	Bone Graft	17.85	14.54	-3.31
Trindade (11)	Orthognathic	21	35	14
Chua (12)	Orthognathic	16.99	25.59	8.6

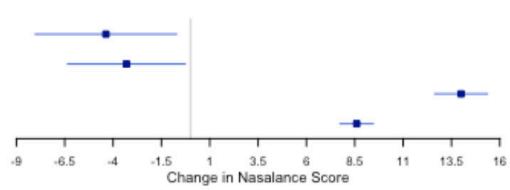

Figure 1: Change in patient nasalance score post operation. Forest plot showing means and 95% confidence intervals for the change in nasalance score post operation for patients undergoing bone grafts and orthognathic surgery. Abbreviations: SX = surgery, Pre = average nasalance score pre-op, Post = average nasalance score post-op, MD = mean change in nasalance score per study.

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Study	SX Type	Pre	Post	PC
Denny (13)	Bone Graft	100	75	-25
Jackson (14)	Bone Graft	52.3	29.5	-22.8
Janulewicz (15)	Orthognathic	58	82	24
Pereira (16)	Orthognathic	55	80	25

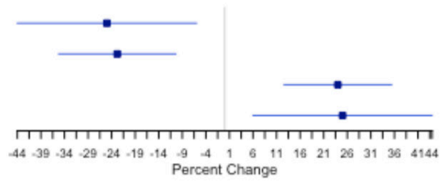


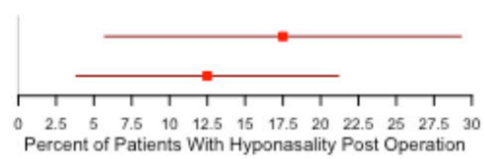
Figure 2: Change in Percentage of Patients with Excess Nasal Emission Post-op. Forest plot showing the percent changes and 95% confidence intervals for patients with excess nasal emission after bone graft and orthognathic surgery. Abbreviations: Pre = percent of patients with excess nasal emission pre-op, Post = percent of patients with excess nasal emission post-op, PC = percent change of patients with excess nasal emission post-op.

Hence, orthognathic surgery actually worsened hypernasality. Across all studies, patients prior to surgery had moderate hypernasality (nasalance scores ranging from 16.99 to 21), consistent with the high prevalence of hypernasality in cleft palate patients (1). Although our results regarding orthognathic surgery indicated that it may help hyponasality by increasing speech nasalance, this increase in speech nasalance is typically considered a negative side effect. Similarly, the meta-analysis conducted for percent change of patients with excess nasal emission post-op found that bone grafts reduce excess nasal emission in populations of cleft palate patients. The percentage of patients with excess nasal emission was reduced in the group that underwent bone graft, whereas the percentage of patients with excess nasal emission was increased in the group that underwent orthognathic surgery. These differences can be attributed to the process of each surgical treatment. Bone grafts in the hard palate close gaps between the nasal and oral cavity, reducing air flow between the two during speech (5). However, orthognathic surgery can destabilize the upper palate, potentially opening palatal gaps that were fixed by palatoplasty (15). As such, orthognathic surgery is normally not used as a treatment solely for treating hyponasality (12). The homogeneity observed between these two meta-analyses indicate that orthognathic surgery may not be well suited for improving nasalance in cleft palate patients.

The meta-analysis conducted for frequency of hypo/hypernasality post-op found that both hypernasality and hyponasality can be found in patients undergoing bone grafts or orthognathic surgery. This means that patients who underwent bone grafts can still have hypernasality, even if our previous results indicated that nasal emission is reduced overall in these patients. One important note, however, is that bone grafts actually resulted in a slightly higher percentage of patients with hypernasality, while orthognathic surgery resulted in a higher percentage of patients with hyponasality. This trend contradicts the findings of our previous analysis, in which bone grafts generally decreased hypernasality. Nonetheless, because this meta-analysis was conducted with only two studies, future studies should be reviewed to further clarify this finding.

A major limitation in our meta-analysis was a general lack of data for specific speech metrics. The analysis of nasalance score used a measured speech metric, while the other two analyses used calculated patient percentages based on respective sample sizes. Confidence intervals calculated using binomial distribution were also overlapping in **Figure 3**, meaning that only trends can be observed. Another limitation was the unavailability of information specific to speech for each surgical procedure. Certain analyses, such as the one comparing hypo/hypernasality, were compiled using fewer studies due to a lack of available data. Overall, each meta-analysis consisted of 2-4 studies, each with less than 100

Study	SX Type	n	Hypo
Maegawa (17)	Orthognathic	40	17.5
Brudnicki (18)	Bone Graft	56	12.5



Study	SX Type	n	Hyper
Maegawa (17)	Orthognathic	40	25
Brudnicki (18)	Bone Graft	56	28.6

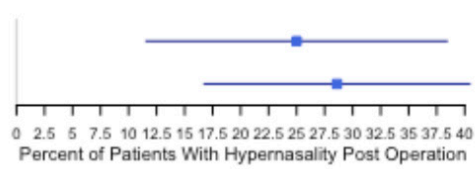


Figure 3: Percent of patients with hypo/hypernasality post-op. Forest plots showing the percentages and 95% confidence intervals for patients with hypernasality and hyponasality after bone graft and orthognathic surgery. The red forest plot includes studies for hyponasality, and the blue forest plot includes studies for hypernasality. Abbreviations: n = sample size, Hyper = percent of patients with hypernasality post-op, Hypo = percent of patients with hyponasality post-op.

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patients. These small sample sizes may partially explain the contradictory results present between different analyses. Additionally, one uncontrolled variable present in these studies was the mixture of patients with bilateral and unilateral cleft palate. Unilateral cleft palate is less severe than bilateral cleft palate, and patients with bilateral cleft usually require more speech therapy. The varying severity of cleft palate present in these patients may contribute to statistical differences because bilateral cleft patients tend to exhibit higher levels of speech nasalance (1).

All studies that we used consisted of adolescent patients. This is understandable, because most cleft palate patients undergo surgical care during adolescence (2). Most studies focused on individual metrics within speech nasalance, meaning that there was no overlap of studies that we used for the three separate analyses. The only studies that utilized one speech metric (nasalance scores) were those used in the analysis for **Figure 1**. Because the other studies do not use nasalance scores as the primary metric for speech nasalance, this did lead to some contradictory results when analyzing the data presented in **Figure 3**. Furthermore, studies used in this paper ranged over 47 years, with studies used for alveolar bone grafts being slightly older. However, there have been no significant changes in either orthognathic surgery or bone graft within this time period, meaning that the time of study is likely not the cause of contradictory metadata (5, 7).

Most cleft palate patients will undergo both surgical procedures to treat their condition (3). Understanding the effects of these procedures on speech can prepare patients for potential outcomes that may need to be addressed. Based on these predicted effects, medical providers can make additional recommendations to improve a patient's long-term speech outcomes.

In summary, our results indicated that alveolar bone grafts are better than orthognathic surgery for improving speech nasalance in cleft palate patients. Bone grafts generally reduced hypernasality, while orthognathic surgery increased hypernasality. For each treatment type, both hypo- and hypernasality could be found in patients post-operation, with no statistical indication of immediate changes to hyponasality. From a clinical perspective, this meta-analysis may inspire future studies and give additional perspective on how to improve nasalance outcomes for individuals with cleft palates. By analyzing the implications of different surgical procedures, personalized medicine approaches can be better tailored for cleft palate patients.

MATERIALS AND METHODS

To perform the meta-analysis, the PubMed and Google Scholar databases were searched using the following keywords: cleft palate AND speech AND (surgery OR craniofacial) AND bone graft OR (jaw OR orthognathic OR maxillary advancement). Additional searches specific to speech metrics were also manually inputted (i.e. "nasalance scores AND bone graft AND cleft palate).

Study Selection Criteria

The criteria for the inclusion of studies were as follows: a) the study focused on speech in cohorts of cleft palate patients, b) the study focused on bone grafts or orthognathic surgery, and c) the study utilized statistical speech metrics relevant to nasalance, hyponasality, or hypernasality.

Reference	Author	Year	SX Type	Sample Size	Speech Metric
10	Bureau	2001	Bone Graft	10	Nasalance Score
11	Trindade	2003	Orthognathic	29	Nasalance Score
12	Chua	2010	Orthognathic	21	Nasalance Score
13	Denny	1993	Bone Graft	20	Percent Patients w/ NE Post Op
14	Jackson	1976	Bone Graft	44	Percent Patients w/ NE Post Op
15	Janulewicz	2004	Orthognathic	54	Percent Patients w/ NE Post Op
16	Pereira	2013	Orthognathic	20	Percent Patients w/ NE Post Op
17	Maegawa	1998	Orthognathic	40	Percent of Patients Hypo/Hypernasality Post Op
18	Brudnicki	2023	Bone Graft	56	Percent of Patients Hypo/Hypernasality Post Op

Table 1: Studies selected for meta-analysis.

The criteria for the exclusion of studies were as follows: a) the study lacked data relevant to nasalance, hyponasality, and hypernasality, b) the study focused on the surgical procedures rather than its effects on speech, and c) the study was a meta-analysis or literature review.

In the initial search, 36 studies were obtained. After applying inclusion and exclusion criteria, numerical data was extracted from nine studies (**Table 1**).

Variable Extraction

The following variables were manually extracted from the selected studies: (a) author, (b) year of publication, (c) title of study, (d) orthognathic surgery or bone graft, (e) sample size, and (f) measured speech metric. Metrics that utilized patient percentages were calculated separately.

The standardized speech metric collected in this meta-analysis was speech nasalance score. In the studies used for **Figure 1**, speech nasalance scores were calculated using a nasometer, a headset with two microphones on either side of a metal plate that sits on the speaker's upper lip (10-12). As patients speak, the nasometer measures the amount of acoustic energy emitted from the nose by calculating the ratio of nasal to oral sound pressure level. Numerical values indicating the degree of speech nasalance are then collected.

Meta-analysis

The meta-analysis was performed using RStudio Version 2023.06.2+561. Data recorded on Google Sheets was imported to RStudio via csv files, and the forestplot, metafor, dplyr, and tidyverse packages were used to plot data. The XQuartz application was used in tandem with R to edit data frames without having to re-upload csv files. ChatGPT 3.5 was also used to fix coding errors. Confidence intervals for each data point were calculated using binomial distribution in R, and all p-values used in the binomial distribution were provided by the respective study. Mean values for trials were extracted from each paper, and were used in the binomial distribution equation to calculate upper and lower confidence intervals. All p-values below 0.05 were considered significantly different from zero.

For **Figure 1**, data was manually inputted for average pre- and postoperative nasalance scores, and the difference among these scores was calculated and plotted. For **Figure 2**, data was manually inputted for percentage of patients with excess nasal emission pre- and postoperatively, and the percent change was calculated and plotted. For **Figure 3**, two separate forest plots were created. Values for percentage of patients with hypo- and hypernasality were manually inputted and plotted.

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APPENDIX

RStudio Programming Scripts:

```
### packages used
library(tidyverse)
library(metafor)
library(forestplot)
library(dplyr)

### change in nasalance score post operation

#create confidence intervals
df <- read_csv("~/Documents/Justin_data.csv")
df$mean <- df$mean_naso_post - df$mean_naso_pre
df$mean_diff <- df$mean
df$variance <- (df$sd_pre^2 / df$n) + (df$sd_post^2 / df$n)
df$lower <- df$mean - qt(1-0.025,df$n)*sqrt(df$variance/df$n)
df$upper <- df$mean + qt(1-0.025,df$n)*sqrt(df$variance/df$n)

#create forestplot
df |>
  forestplot(labeltext = c(study, sx_type, mean_naso_pre, mean_naso_post, mean_diff),
             xlab = "Change in Nasalance Score",
             boxsize=0.2)|>
  fp_set_style(box = "darkblue",
               line = "royalblue",
               summary = "royalblue")

write.csv(df,file="Nasalance Scores Table.csv")

### percentage of patients with excess nasal emission post-op

#create confidence intervals
percentNE$change <- (percentNE$percent_nasal_emission_post -
percentNE$percent_nasal_emission_pre)/100
```

```
alpha <- 0.95
z <- qnorm(1-(1-alpha)/2)
percentNE$lower <- (percentNE$change - z*sqrt(abs(percentNE$change) * (1-
abs(percentNE$change)) / percentNE$n)) * 100
percentNE$upper <- (percentNE$change + z*sqrt(abs(percentNE$change) * (1-
abs(percentNE$change)) / percentNE$n)) * 100
percentNE$mean <- percentNE$change * 100

# create forestplot
percentNE %>%
  forestplot(
    labeltext = c("study", "sx_type", "percent_nasal_emission_pre",
"percent_nasal_emission_post", "mean"),
    xlab = "Percent Change",
    boxsize=0.2) %>%
  fp_set_style(box = "royalblue",
               line = "darkblue",
               summary = "royalblue")

write.csv(percentNE, file="Percent Excess Nasal Emission.csv")

### percentage of patients with hypernasality post-op

#create confidence intervals
percentHYPER <- read_csv("~/Desktop/percent_hypernasality_post.csv")
percentHYPER$change <- percentHYPER$mean/100
percentHYPER$lower <- (percentHYPER$change - z*sqrt(abs(percentHYPER$change) * (1-
abs(percentHYPER$change)) / percentHYPER$n)) * 100
percentHYPER$upper <- (percentHYPER$change + z*sqrt(abs(percentHYPER$change) * (1-
abs(percentHYPER$change)) / percentHYPER$n)) * 100

#create forestplot
percentHYPER %>%
  forestplot(
    labeltext = c("study", "sx_type", "n", "percent_hypernasality_post"),
```

```
    xlab = "Percent of Patients With Hypernasality Post Operation",
    boxsize=0.2) %>%
  fp_set_style(box = "royalblue",
               line = "darkblue",
               summary = "royalblue")

write.csv(percentHYPER, file="Percent Hypernasality.csv")

### percentage of patients with hyponasality post-op

#create confidence intervals
percentHYPO <- read_csv("~/Desktop/percent_hyponasality_post.csv")
percentHYPO$change <- percentHYPO$mean/100
percentHYPO$lower <- (percentHYPO$change - z*sqrt(abs(percentHYPO$change) * (1-
abs(percentHYPO$change)) / percentHYPO$n)) * 100
percentHYPO$upper <- (percentHYPO$change + z*sqrt(abs(percentHYPO$change) * (1-
abs(percentHYPO$change)) / percentHYPO$n)) * 100


#create forestplot
percentHYPO %>%
  forestplot(
    labeltext = c("study", "sx_type", "n", "percent_hyponasality_post"),
    mean = percent_hyponasality_post,
    xlab = "Percent of Patients With Hyponasality Post Operation",
    boxsize=0.2) %>%
  fp_set_style(box = "red",
               line = "darkred",
               summary = "red")

write.csv(percentHYPO, file="Percent Hyponasality.csv")
```