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Feeding Issues and Interventions in Infants and Children with Clefts Lip and Palate: A Meta-Analysis

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ABSTRACT

The feeding process in infants with cleft lip and palate is greatly influenced by the ability to suck and swallow. Infants with this condition often have difficulty in sucking and swallowing effectively, which can lead to longer feeding duration, fatigue in the infant, risk of aspiration, and decreased nutritional intake. Inefficient oral feeding can inhibit the growth and development of the infant. Therefore, feeding infants with cleft lip and/or palate requires great care and knowledge from the mother. If not treated quickly and appropriately, this condition can cause malnutrition, growth retardation, and other health complications. This study aims to determine the description of feeding and intervention in infants with cleft lip and palate. The method used is meta-analysis, with a range of articles selected from 2015 to 2025. Article searches were carried out systematically and comprehensively through the PubMed, Science Direct, Elsevier, and Google Scholar databases, using the PICO (Population, Intervention, Comparison, Outcome) model to determine the eligibility of the article. Articles that met the qualitative criteria were reviewed again, and from this process six articles were obtained that were eligible for quantitative analysis. This meta-analysis showed that infants and children with cleft lip and palate had lower developmental outcomes than those without the condition (effect value -0.41; 95% CI = -0.29 to 0.01). The six primary studies were from countries such as South Africa, the United States, Brazil, and Italy, and were cross-sectional in design.

Keywords: *Eating Problems, Eating Disorder Interventions, Children with Cleft Lip and Palate*

1. INTRODUCTION

Cleft lip and palate (CLP) is a congenital disorder that can have a significant impact on the developmental aspects of the baby. Cleft lip and palate is a birth defect that affects the structure of the lip, palate or both caused by problems in the process of embryonic facial fusion. Although cleft lip and palate is a single anomaly, there are still consequences that affect several systems and functions of the child and cause social and psychological problems. Therefore, early intervention for cleft lip and palate is very important. The ideal time for lip closure is at 3 months and palate closure is at 9 months. Determining age is very important to achieve language and speech quality standards. In addition to language and speech aspects, the main problem experienced by babies with cleft lip and palate is feeding issues. Before being advised to close the cleft lip and palate, parents will experience several obstacles in successful feeding. This disorder arises due to anatomical problems that affect the baby's ability to produce the oral pressure needed in the sucking process(Todorović et al., 2022)

The process of feeding babies with cleft lip and palate is greatly influenced by the ability to suck and swallow. Feeding babies with cleft lip and/or palate cases requires great concern from parents. Children who experience cleft lip and/or no palate usually also experience growth delays compared to other children who do not experience cleft lip and/or no palate. According to(Singhal, 2022)explains the provision of feeding assistance in cases of cleft lip and palate through the nasogastric method with 29% of cleft lip cases requiring

one month or more to regulate regular eating patterns with 77% of cleft lip babies, while 20% never have regular eating patterns. Inefficient oral feeding can result in stunted growth and development. Babies with CLP often have difficulty sucking and swallowing effectively, which can lead to longer feeding durations, fatigue in babies, risk of aspiration, and decreased nutritional intake. This has the potential to cause stunted growth and development, malnutrition and other health complications if not treated quickly. In addition, difficulties in feeding can put emotional and psychological pressure on parents or caregivers, who must find solutions to meet the baby's nutritional needs(Singhal, 2022).

Along with the development of medical and technological, intervention management in children with CLP has been developed to overcome feeding problems. Interventions include the use of bottles/nipples designed to facilitate feeding, nasal veolar molding to help close the gap temporarily, and education for parents regarding proper feeding techniques. In addition, the role of a multidisciplinary team, including nutritionists, pediatricians, plastic surgeons and speech therapists is very important to provide effective treatment. However, although various interventions have been introduced, there are still various results in the effectiveness and success of these interventions. One factor is the lack of a comprehensive understanding of the factors that influence feeding problems, as well as limited research on the long-term impact of the interventions carried out with further research needed.(Jar et al., 2023).

This study aims to identify feeding issues specific to infants with CLP, evaluate the effectiveness of existing interventions, and provide research-based recommendations for future clinical improvement. In doing so, this study is expected to contribute to improving the quality of life of infants with CLP, as well as providing insights for health care providers and families to address these challenges more effectively.

2. METHOD

This study used a meta-analysis design, namely by combining and analyzing data from various relevant primary studies. The analysis was carried out systematically to obtain a comprehensive picture of the problems and interventions for feeding infants and children with cleft lip and palate. This study was conducted by searching and selecting articles from various ethnicities and races spread throughout the world. The time span of the selected articles covers 2015 to 2025, while the process of searching and selecting articles was carried out for two weeks.

Geographically, the articles used in this study came from various countries, namely South Africa, the United States, Brazil, and Italy. The six articles were selected because they met the predetermined inclusion criteria. The data search strategy was carried out through several international databases that have reliable indexing, including PubMed, Science Direct, Elsevier, and Google Scholar. In the search process, several keywords were used such as "children", "baby", "eating problem", "cleft lip and palate", "eating disorder intervention", and equivalents in Indonesian such as "bibir sumbing" and "permasalahan serta pelayanan makanan pada bayi dan anak dengan cleft lip and palate". To determine the eligibility of the article, the researcher used the PICO (Population, Intervention, Comparison, Outcome) model approach, with the population being infants and children, the intervention being cleft lip and palate, the comparator being children without the condition, and the results (outcomes) being eating problems and the interventions given.

The data collection technique follows the meta-analysis principle according to Davies and Crombie in Murti (2018). The first step is to formulate inclusion and exclusion criteria. Inclusion criteria include full-text articles that use a cross-sectional design with multivariate analysis, involve infant and child subjects, and focus on feeding problems and their interventions. Meanwhile, exclusion criteria include articles in languages other than English, using bivariate analysis, and published before 2015. A comprehensive literature search was conducted, including published and unpublished articles, as well as additional articles obtained from references to the main articles that have passed the selection.

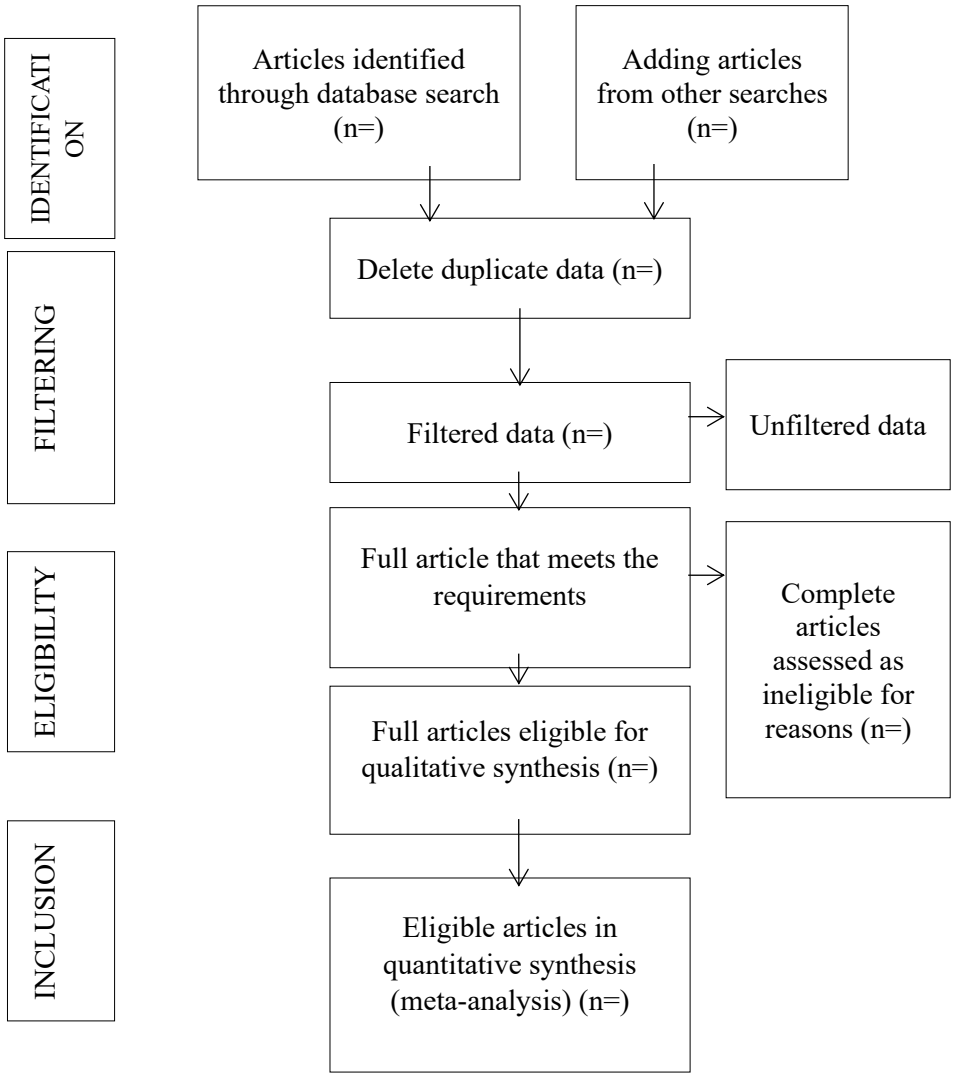
In assessing the quality of research, Articles that have been identified and then each study is assessed based on eligibility criteria. The quality of research and reporting of findings. The quality and design of research analyzed in meta-analysis are very important because they affect the results. Assessment of study quality is carried out quantitatively and qualitatively. The assessment criteria in this study are, checklist for observational cross-sectional studies(Murti, 2018).

In Combining Results, Findings from each study are combined (aggregation) to produce basic conclusions about feeding problems and feeding interventions in infants and children with Cleft Lip and Palate. Aggregation in this study was carried out qualitatively and quantitatively. Researchers used a continuous scale to obtain the same final results for all studies to be analyzed. The next step is to calculate the overall effect of

combining the data. Data analysis was carried out using Review Manager (RevMan) 5 software issued by the Cochrane Collaboration. RevMan is used to describe the 95% confidence interval (CI) using the effect model, and also I2 or data heterogeneity (Murti, 2018).

Research Steps

The steps of this research are to follow the PRISMA 2009 flow diagram guidelines.



Flowchart 1. PRISMA 2009 flow diagram

In this study, the variables studied consist of independent variables and dependent variables. The independent variable is Cleft Lip and Palate, which is a congenital abnormality characterized by a gap or split in the upper lip, palate, or both. This abnormality occurs due to failure of tissue fusion during embryogenesis. Meanwhile, the dependent variable is the problem and intervention of feeding in infants and children with Cleft Lip and Palate. The eating problems in question are related to difficulties in nutritional intake, such as lack of interest in eating or obstacles in receiving adequate nutrition.

The operational definition of Cleft Lip and Palate in this study refers to congenital abnormalities that can be measured using commonly used classification systems, such as the LAHSHAL, Veau, or WHO classifications. The measurement scale for this variable is continuous. As for feeding problems and interventions, the definition refers to nutritional barriers experienced by infants and children, the data of which are obtained through interviews with parents. This variable is also measured using a continuous scale.

The instruments used in this study were published scientific articles that raised issues related to problems and interventions in feeding infants and children with cleft lip and palate. Data processing was carried out through a series of stages, starting from identifying articles obtained from various reputable journal databases.

The articles found were then selected through a screening process to ensure compliance with the research criteria. Articles that passed the eligibility stage were then further analyzed using special software for meta-analysis, in order to obtain systematic and valid results.

In this Research Analysis, Variation between studies according to Dahlan in 2012 is used to determine the analysis model which is divided into two models, namely the fixed effect model (FEM) and the random effect model (REM): If there is no variation between studies, the p value in the heterogeneity test is greater than 0.05 or I2 is small, then the analysis model used is the fixed effect model (FEM). The combined effect is calculated by ignoring the variation between studies (Dahlan, 2012). Calculation of the combined effect using FEM (EG) is:

$$\mathbf{EG}=\frac{\Sigma WY}{\Sigma W}$$

If the p value varies between studies in the heterogeneity test of less than 0.05 or I2 is large, then the analysis model used is the random effect model (REM). The combined effect is calculated by involving variations between studies (Dahlan, 2012). The calculation steps using REM are as follows:

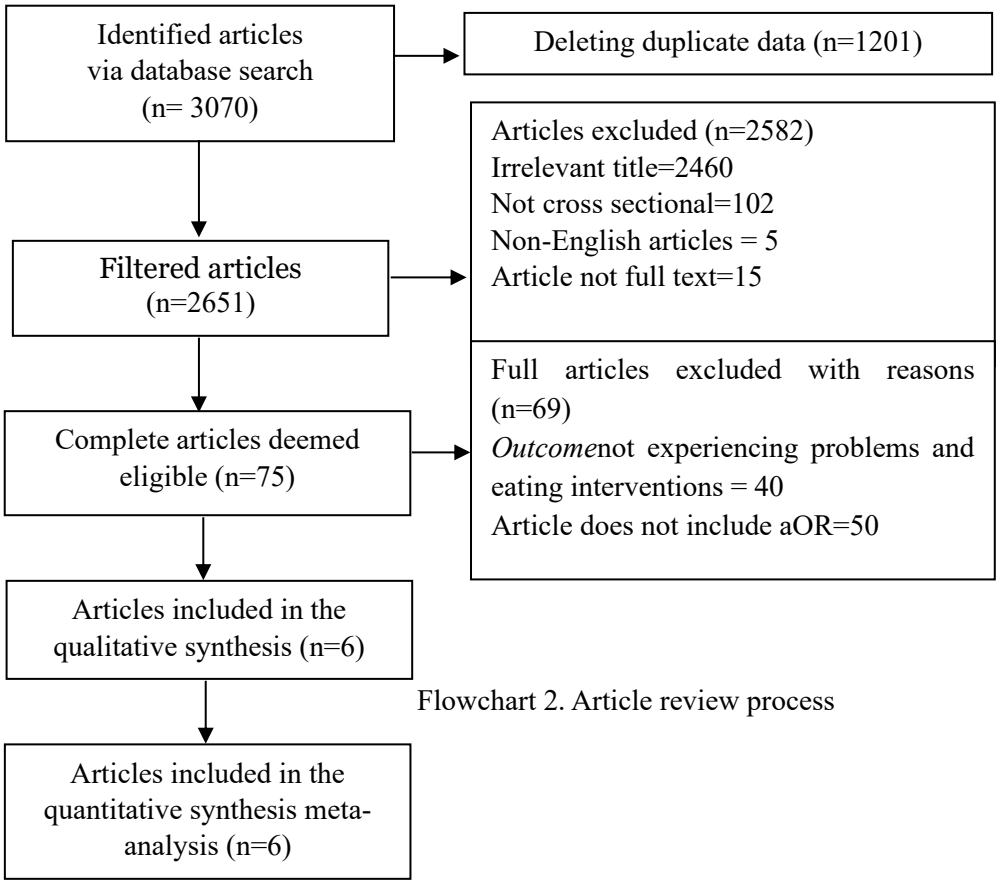
Calculate the total variation Vt: $\mathbf{Vt = V+ T2}$

Calculate the research weight (Wt and Wt%) $\mathbf{Wr = Wt\% = x\ 100\ \%}\frac{1}{Vt}\frac{Wt}{\Sigma Wt}$

Calculate the combined effect with the Random Effect Model (EGr): $\mathbf{Egr} =\frac{\Sigma WtY}{\Sigma Wt}$

3. RESULTS AND DISCUSSIONS

The search process by searching through journal databases including PubMed, Science Direct, Elsevier, and Google Scholar. The search keywords used are "children" AND "baby" AND "eating problem" AND "Cleft lip and palate" AND "eating disorder intervention" AND "Problems and Interventions in Feeding In Infants and Children With Cleft Lip And Palate", "Cleft Lip", "Problems and Interventions in Feeding In Infants and Children With Clef Lip and Palate". The article review process can be seen in the search flow as follows:



Flowchart 2. Article review process

The initial search process yielded 3070 articles, after going through the process of deleting duplicate articles, 2582 articles were obtained with 75 of them meeting the requirements for further full text review. Full text articles that fall into the exclusion criteria are due to the following:

1. Not using observational studies
2. *Outcome*from observational studies not problems and interventions of feeding in infants and children
3. Independent variables are not cleft lip and palate
4. The intervention given is not cleft lip and palate
5. Does not include aOR values from multivariate analysis results

Articles that meet the qualitative requirements were reviewed again and the final results of the article review process were obtained, there were 6 articles that met the quantitative requirements for conducting a meta-analysis of problems and interventions for feeding infants and children with cleft lip and palate.

*Assessment*Research Quality

The assessment of study quality was carried out quantitatively and qualitatively, this study used the Critical Appraisal Checklist for Cross-sectional (Center for Evidence Based Management, 2014).

Table 1. Assessment of Research Quality on Problems and Interventions for Feeding Infants and Children with Cleft Lip and Palate

No	Research Questions	Rando et al., 2018	Montes et al., 2018	Visser et al., 2018	Madhoun et al., 2021	Defabianis et al., 2022	Prezelski et al., 2024
1.	Does this objective clearly address the focus/problem of the research?	1	1	1	1	1	1
2.	Is the research method (research design) suitable for answering the research question?	1	1	1	1	1	1
3.	Is the method of selecting research subjects written clearly?	1	1	1	1	1	1
4.	Can the sampling method introduce bias (selection)?	0	0	0	0	0	0
5.	Does the research sample taken represent the designated population?	1	1	1	1	1	1
6.	Is the sample order based on pre-study considerations?	1	1	1	1	1	1
7.	Was a satisfactory response achieved?	1	1	1	1	1	1
8.	Are the research instruments valid and reliable?	1	1	1	1	1	1
9.	Is statistical significance assessed?	1	1	1	1	1	1
10.	Are confidence intervals provided for the main results?	1	1	1	1	1	1
11.	Are there any confounding factors that have not been taken into account?	0	0	0	0	0	0
12.	Can the results be applied to your research?	1	1	1	1	1	1
	Total score	**10**	**10**	**10**	**10**	**10**	**10**

Article Summary Source

There are 6 observational study articles as sources for the meta-analysis of Problems and Interventions for Feeding in Infants and Children with Cleft Lip and Palate, namely:

Table 2. Description of primary studies included in the meta-analysis

Writer (year)	Country	Study design	Sample	P	I	C	O
(Rando et al., 2018)	Brazil	*Cross-sectional*	121	Children	Child cleft lip and palate	Child does not have cleft lip and palate	*Eating Disorder Problems and Interventions*
(Montes et al., 2018)	Brazil	*Cross-sectional*	108	Children	Child cleft lip and palate	Child does not have cleft lip and palate	*Eating Disorder Problems and Interventions*
(Visser et al., 2018)	South Africa	*Cross-sectional*	25	Baby	Child cleft lip and palate	Child does not have cleft lip and palate	*Eating Disorder Problems and Interventions*
(Madhoun et al., 2021)	American	*Cross-sectional*	60	Baby	Child cleft lip and palate	Child does not have cleft lip and palate	*Eating Disorder Problems and Interventions*
(Defabianis et al., 2022)	Italy	*Cross-sectional*	142	Children	Child cleft lip and palate	Child does not have cleft lip and palate	*Eating Disorder Problems and Interventions*
(Prezelski et al., 2024)	American	*Cross-sectional*	300	Babies	Child cleft lip and palate	Child does not have cleft lip and palate	*Eating Disorder Problems and Interventions*

Forest plot

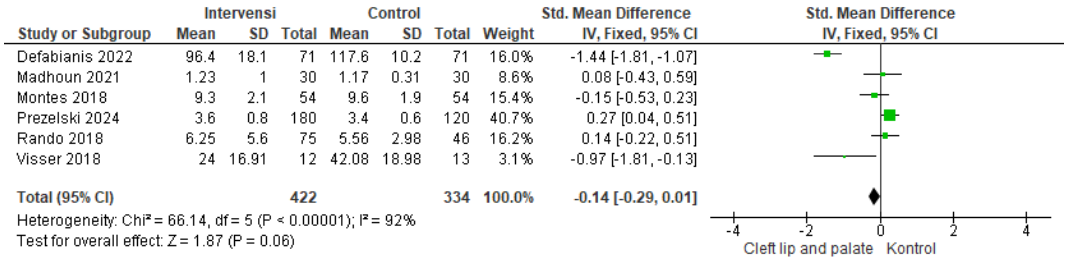


Figure 3. Forest plot of the effect of stunting on child development

Interpretation of the results of the meta-analysis process can be seen through the forest plot. Figure 4.3 shows that negative cleft lip and palate have a negative effect (-0.14; 95% CI -0.29 to 0.01) meaning that there is an increase in problems and feeding interventions compared to children without cleft lip and palate, and is statistically significant (p <0.001). The heterogeneity of the research data shows I2 = 92% so that the distribution of data is stated as heterogeneous (random effect model).

Funnel Plot

*Funnel plot*is a plot that plots the estimated effect size from each study against an estimate of its precision, usually the standard error.

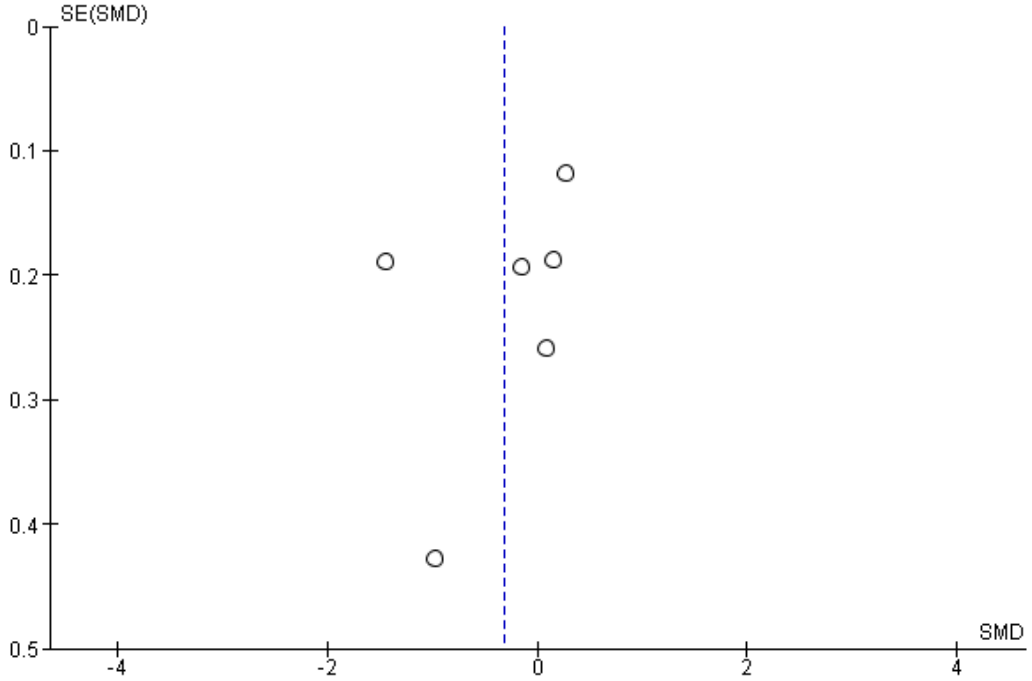


Figure 4. Funnel plot of the effect of stunting on child development

Based on Figure 4.4, it usually shows a standard error. Based on Figure 4.4, the results of the meta-analysis study show that there is a publication bias indicated by the non-symmetrical plot distribution. The estimated effect size in infants and children with cleft and lip palate who experience problems and feeding interventions exceeds the actual size (overestimated). The plot on the right of the graph has a standard error between 0.1 and 0.3 and the plot on the left of the graph has a standard error of 0.2 and 0.6.

Discussion

Cleft lip or often called cleft lip and palate (CLP) is one of the most common types of birth defects. Orofacial clefts manifest as a cleft lip, either accompanied by or independent of a cleft palate, or as a cleft palate that is not associated with a cleft lip. This condition can be found in about 1 in every 500 to 1000 babies born worldwide(Rando et al., 2018; Ucar et al., 2015).

The way babies with cleft lip and palate are fed is greatly influenced by their ability to suck and swallow. Caring for a baby born with a cleft lip and/or palate requires a great deal of attention from caregivers. Ineffective oral feeding can hinder proper growth and development. Babies with cleft lip and palate often struggle to suck and swallow efficiently, resulting in prolonged feeding sessions, fatigue, potential risk of aspiration, and lower nutrient intake.(Ashby, 2011; Denadai & Lo, 2024; Dudding et al., 2023).

The consequences on communication, auditory perception, appearance, and mental health can affect the well-being and social engagement of those affected. Typically, children with this disorder will require comprehensive care from infancy through adulthood and tend to face increased rates of illness and death compared to those without the underlying condition.(Merino et al., 2024; Ogundele, 2018).

This study is a systematic review and meta-analysis. The purpose of this study is to obtain conclusions from various similar studies regarding the problems and interventions of feeding in infants and children with cleft lip and palate. Research that discusses data on cleft lip and palate with problems and interventions of feeding is considered important because there are problems of inconsistency/controversy regarding the results of various primary studies/research that have been conducted. The number of relevant studies published and accessible is still small and also has data access problems (data duplication) (Murti, 2018).

*Confounding factors*affect the relationship or effect of exposure to the occurrence of the disease estimated by the study is not the same as the relationship or effect that actually occurs in the target population, aka invalid study results (Murti, 2018). The combined estimate of children with cleft lip and palate for feeding problems

and interventions was processed using the RevMan 5.3 application with the generic inverse variance method. This method is used to analyze data in the form of rates, time-to-event, hazard ratio, ordinal scale, adjusted estimate, difference of mean or ratio of mean.

The results of systematic reviews and meta-analyses are presented in the form of forest plots and funnel plots. Forest plots show an overview of information from each study studied in the meta-analysis, and estimates of the overall results (Murti, 2018). Forest plots visually show the magnitude of variation (heterogeneity) between study results (Akobeng in Murti, 2018).

*Funnel plot*is a diagram in meta-analysis used to demonstrate the possibility of publication bias. The funnel plot shows the relationship between the study's effect size and the sample size or standard error of the effect size from the various studies studied (Murti, 2018).

*Funnel plot*visually shows the magnitude of variation (heterogeneity) (Akobeng, 2005 in Murti, 2018). The funnel plot shows the relationship between the effect size of the study and the sample size of the various studies studied, which can be measured in various different ways (Murti, 2018).

The results of a meta-analysis of 6 articles on problems and interventions in feeding infants and children with cleft lip and palate on child development are summarized in a forest plot. The forest plot in Figure 4.3 shows that infants and children with cleft lip and palate have a negative value which will increase the risk of experiencing abnormal development compared to infants and children without cleft lip and palate (-0.41; 95% CI = -02.9 to 0.01). As a result, infants and children with cleft lip and palate may be a strong risk factor for problems and interventions in feeding infants and children.

This is supported by a quote from Danelz (2019) in the American Cleft pLate- Craniofacial Association (ACPA) that babies with cleft lip and palate cannot breastfeed properly because they cannot create the pressure needed to suck the nipple. Trying to give with a regular bottle to provide food can cause poor weight gain to growth failure in children.

Babies born with cleft lip and palate have the same nutritional needs as babies without the condition, provided there are no additional health problems. Ensuring adequate nutrition remains important, while identifying a feeding method that closely mimics normal feeding is essential. Key challenges include poor sucking ability, excessive water intake, risk of choking, nasal reflux, fatigue, inadequate milk intake, poor weight gain and long breastfeeding sessions. Difficulty breastfeeding effectively can also increase stress and anxiety for the mother, potentially leading to a weakened mother-child bond.(Istien Wardani & Dyah Ayu, 2022; WHO, 2020).

The main characteristics of feeding techniques discussed in the study were different feeding routes (such as nasogastric tube), suction-based methods (such as bottle and breast feeding), and non-suction approaches (including cup, spoon, syringe, and paladai). Aspects assessed in the study included food acceptance and amount consumed, feeding efficiency, duration and problems encountered during feeding, and growth and nutritional progress.(Moxon, 2028; UNICEF, 2016).

Other aspects related to different feeding approaches and growth metrics, as well as challenges with feeding, are presented in the results. Parameters related to sucking, including suction and compression during bottle feeding, were evaluated. Assessments of sucking ability, oral motor skills, and feeding effectiveness occurred at 2 weeks, 3 months, and 14 months of age. Findings showed that infants with smaller clefts (eg, CL or minor soft palate cleft) tended to demonstrate normal levels of suction and compression more often than those with larger clefts. Newborns with cleft palate and CLP often had feeding difficulties. The incidence of inadequate feeding skills decreased to 19% at 3 months of age and 15% at 14 months of age. Oral motor problems and ongoing symptoms, especially nasal regurgitation, were more common in infants who demonstrated poor feeding skills, regardless of any additional conditions. Infants with nonsyndromic complete unilateral CLP or a cleft affecting the soft palate and at least two-thirds of the hard palate demonstrated less effective sucking patterns compared with infants without a cleft.(Chen & Antonelli, 2020; Kesari et al., 2023; Leach et al., 2020; Pados et al., 2017).

Feeding infants and children with cleft lip and palate should be the next concern of parents and caregivers, a factor that can cause significant stress for the mother and family. Prompt and accurate diagnosis and visits by health workers are carried out immediately after the birth of the baby so that the family can be supported and taught feeding skills. It is important to realize that parents with infants and children with cleft lip and palate need significant support so that the infants and children can grow and develop normally.

4. CONCLUSION

This meta-analysis concluded that infants and children with cleft lip and palate had a negative value where it would increase the risk of experiencing abnormal development compared to infants and children without cleft lip and palate (-0.41; 95% CI = -02.9 to 0.01). This meta-analysis combined 6 primary studies using cross-sectional designs from countries from South Africa, America, Brazil and Italy.

Research Contribution

Anggi Resina Putri as the main researcher plays a role in identifying articles, screening articles, article eligibility, inclusion and analysis of research data. Roy Romey Daulas Mangunsongplay a role inbackground, formulation of the frameworkthoughts, supporting theories and research discussion.

Conflict Of Interest

There is no conflict of interest whatsoever in this research.

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