Open Access Indonesian Journal of Medical Reviews

Journal Homepage: https://hmpublisher.com/index.php/OAIJMR

Characteristics of Laryngomalacia Patients at Department of Otorhinolaryngology-Head and Neck Surgery Dr. Mohammad Hoesin General Hospital, Palembang, Indonesia Period 2021-2022

Fatharani Azka Toer[1*], Puspa Zuleika[2], Adelien[2], Lisa Apri Yanti[2], Fiona Widyasari[2]

[1]Medical Education Study Program, Faculty of Medicine, Universitas Sriwijaya, Palembang, Indonesia

[2]Department of Otorhinolaryngology-Head and Neck Surgery, Faculty of Medicine, Universitas Sriwijaya/Dr. Mohammad Hoesin General Hospital, Palembang, Indonesia

ARTICLE INFO

Laryngomalacia
Characteristics
Palembang

***Corresponding author:**
Fatharani Azka Toer

E-mail address:
azkatoer12@gmail.com

All authors have reviewed and approved the final version of the manuscript.

https://doi.org/10.37275/oaijmr.v3i1.273

A B S T R A C T

Laryngomalacia is a congenital laryngeal disorder that is commonly found as a cause of stridor in infants and children. This disorder causes the collapse of the structures in the larynx resulting in impaired breathing and eating conditions in patients. This study aimed to present the characteristics of laryngomalacia patients at the Department of Otorhinolaryngology-Head and Neck Surgery (ORL-HNS) Dr. Mohammad Hoesin General Hospital, Palembang, Indonesia, period 2021-2022. This was a descriptive observational study that used medical record data as secondary data and primary data in the form of questionnaires in the medical record section and ORL-HNS at Dr. Mohammad Hoesin General Hospital, Palembang, period 2021-2022. Data evaluated included age, gender, clinical symptoms, classification based on Olney distribution, degree of severity, comorbid factors, and therapy. This study showed that most patients were in the age group 3-12 months (52.9%). More were found in males (73.5%). The most common clinical symptoms were eating disorders (51.4%). The most common classification is type 1 (52.9%). Most patients come with a mild degree of severity (61.8%). The most common comorbid factors were neurological diseases (35.1%). The type of medical therapy was given to the majority of patients (94.1%).

1. Introduction

Laryngomalacia is a congenital laryngeal disorder that is commonly found as a cause of stridor in infants and children.[1] Laryngomalacia accounts for 60-70% as a cause of stridor cases in neonates and infants, also affecting 75% of infants in the United States.[2,3] Although the exact incidence is not known in the entire population, it is estimated that around one in 2,100-2,600 children suffers from laryngomalacia, with the prevalence of males being higher than females.[4] Although the cause of laryngomalacia is still uncertain. Many theories have been proposed for the etiology of laryngomalacia, including anatomical, cartilage, and neurological/neuromuscular theories.[2,5] The most credible theory is that of changes in the neuromuscular tone of the larynx and resulting prolapse of the supra-arytenoid tissue and supraglottic collapse causing airway obstruction.[6] There is no classification system that is widely used to classify laryngomalacia, but many categorizations have been developed to classify this disease. One of the categorizations was carried out by Olney by classifying laryngomalacia into three types based on laryngoscopy appearance. In the Olney classification system, laryngomalacia is divided into type 1—prolapsed appearance of the mucosa over the arytenoid

cartilages, type 2—shortening of the aryepiglottic folds, and type 3—collapse of the epiglottis posteriorly.[2,7]

There is no standard method that is generally used to classify the severity of laryngomalacia. However, an overview of the severity of this disease can be seen from the severity of respiratory symptoms and eating disorders which are divided into mild, moderate, and severe categories.[8] Symptoms that can be used as an assessment of the severity of this disease include stridor, oxygen saturation level, cyanosis or apnea, coughing, choking, regurgitation, aspiration, and failure to thrive. The severity of laryngomalacia is also influenced by the presence of comorbid factors in the patient. Medical comorbidities certainly affect the respiratory condition or ability to eat in patients, so they have an impact on the course of laryngomalacia patients.[3] Several comorbidities that may accompany laryngomalacia include gastroesophageal reflux disease (GERD) or laryngopharyngeal reflux (LPR), neurological disease, congenital heart disease, congenital anomalies or syndromes, and secondary airway lesions.[6] This study aimed to determine the characteristics of laryngomalacia patients at the Department of Otorhinolaryngology-Head and Neck Surgery (ORL-HNS) Dr. Mohammad Hoesin General Hospital Palembang. The characteristics studied included age, gender, clinical symptoms, classification, degree of severity, comorbid factors, and type of therapy.

2. Methods

The study was conducted in the form of a descriptive observational study to determine the characteristics of laryngomalacia patients at ORL-HNS Dr. Mohammad Hoesin General Hospital Palembang for the 2021-2022 period. This study used secondary data in the form of medical record data and primary data in the form of questionnaires to complement medical record data according to the variables used in this study. The sample of this study was laryngomalacia patients at ORL-HNS Dr. Mohammad Hoesin General Hospital Palembang in 2021-2022. The inclusion criteria include 34 laryngomalacia patients at ORL-HNS Dr. Mohammad Hoesin General Hospital Palembang in 2021-2022 with complete medical record data and FOL examination (fiber optic laryngoscopy) in the diagnosis of laryngomalacia. This study was approved by the health and research ethics committee of the Faculty of Medicine, Universitas Sriwijaya, Palembang, Indonesia.

Data were collected and recorded according to the variables required for the study, namely age, sex, clinical symptoms, classification, degree of severity, comorbid factors, and type of therapy. Then, the data is processed using SPSS statistics software 24. Then, the data is presented in tabular form and explained in narrative form.

3. Results and Discussion

Based on the data shown in Table 1, it was found that most patients were in the age group 3-12 months, with a total of 18 patients (52.9%) followed by 9 patients (26.5%) in the age group <3 months, 5 patients (14, 7%) in the 1-2 year age group, and as many as 2 patients (5.9%) in the 2-3 year age group. Similar results were obtained in a study conducted at the ORL-HNS RSHS/FKUP polyclinic for the 2012-2015 period, which showed that of the 107 patients involved in the study, the average patient age was 4.19 months, with the largest age group being children aged 3. -6 months a number of 35 people.[9] The infant itself is considered an obligate nasal breather due to immaturity of coordination between respiratory efforts, oropharyngeal motor systems, and sensory input. The coordination ability to breathe and swallow improves with the maturation process in children. During the period of postnatal development, enlargement of the cranium accompanied by flexion of the cranium base results in a downward displacement of the larynx. This causes the separation between the uvula and epiglottis and the elongation of the pharyngeal cavity, which functions in respiration and swallowing. This maturation or transitional period occurs at the age of 3 to 5 months. Because of the precise timing of the development and coordination of

the respiratory and swallowing systems, the presence of structural or neurological disorders in children can easily interfere with the function of both.[10,11] This is what then happens to children with laryngomalacia, where the anatomical structural abnormalities of the larynx affect breathing and swallowing functions in children. The results of this study also found no patients aged over 3 years. These results are in line with the theory of infraglottic airway development in pediatrics. Where this theory states that although there are differences between the pediatric larynx and the adult larynx (in size, position, consistency, and shape) which predispose to airway obstruction in laryngomalacia, the pediatric larynx experiences twice the acceleration of growth in the first 3 years.[12–15]

Table 1. Frequency distribution of laryngomalacia patients based on age (n=34).

Age	Total
<3 months	9 (26,5%)
3-12 months	18 (52,9%)
1-2 years	5 (14,7%)
2-3 years	2 (5,9%)
>3 years	0 (0%)

Table 2. Frequency distribution of laryngomalacia patients based on gender (n=34).

Gender	Total
Male	25 (73,5%)
Female	9 (26,5%)

Based on the data shown in Table 2, 25 patients (73.5%) were male, and 9 patients (26.5%) were female. So it is known that male patients are more than female patients. These results are similar to a study conducted in Surabaya where out of 76 patients, 41 patients were males and 35 females.[16] Another study conducted at the ORL-HNS polyclinic RSHS/FKUP for the 2012-2015 period showed that there were 69 male patients and 38 female patients.[9] There is no difference in the male and female larynx at birth and in childhood. Other research conducted on the role of gender in the incidence of laryngomalacia stated that there was no certainty about the relationship between gender and the incidence of laryngomalacia, although many previous studies stated that men had more laryngomalacia.[17] Differences in the male and female larynx only occur during puberty due to sex hormones that affect anatomical and physiological changes in the larynx. At puberty, males experience lengthening of the thyroid cartilage without widening, which makes the thyroid lamina narrower and forms a characteristic protrusion. This difference in size and shape then distinguishes the larynx of males and females in adulthood.[18–20]

Table 3. Frequency distribution of laryngomalacia patients based on clinical symptoms (n=34).

Clinical symptoms	Total
Stridor inspiration	16 (47,1%)
Dyspnea	14 (41,2%)
Postprandial regurgitation	2 (5,9%)
Cyanotic	3 (8,8%)
Oxygen desaturation	3 (8,8%)
Difficulty eating	19 (51,4%)
Growth faltering	7 (20,6%)
Pulmonary infection	6 (17,6%)

Based on the data shown in Table 3, it was found that most patients experienced clinical symptoms in the form of difficulty eating a total of 19 patients (51.4%), and the least clinical symptom experienced by patients was postprandial regurgitation which was experienced by 2 patients (5.9%). The results of this study were slightly different from the results of previous studies where the most frequent clinical symptom of 76 patients was inspiratory stridor with a total of 63 patients, followed by 7 patients experiencing dyspnea, 4 patients experiencing snoring, 1 patient having hoarseness, and 1 patient choking easily.[16] The difference in the results of this study was due to differences in the grouping of clinical symptoms of eating difficulties which consisted of four clinical symptoms such as coughing, choking, difficulty swallowing, and oromotor disturbances. Whereas in previous studies, these four clinical symptoms were divided into different groups. Laryngomalacia affects two main domains, namely the respiratory tract and the alimentary canal. Disorders of the airways that affect breathing result in clinical symptoms such as stridor, retractions, cyanosis, and apnea. Meanwhile, disorders of the airways that affect the swallowing process results in clinical symptoms in the form of coughing or choking while eating, growth faltering, and recurrent lung infections due to aspiration.[21] Cough that occurs in children with laryngomalacia as an eating disorder can occur because of this silent aspiration where food or liquid enters the airway. This can result in recurrent respiratory infections or pneumonia in patients.[3] Impaired balance in the coordination of eating and breathing in laryngomalacia patients also results in increased metabolic demands, which can lead to weight loss and growth faltering.[8]

Table 4. Frequency Distribution of laryngomalacia patients based on Olney classification (n=34).

Classification	Total
Type 1	18 (52,9%)
Type 2	6 (17,6%)
Type 3	2 (5,9%)
Combined type	8 (23,5%)

Based on the data shown in Table 4, laryngomalacia type 1 is the most common type of classification experienced with a total of 18 patients (52.9%), followed by the combined type with 8 patients (23.5%), type 2 with 6 patients (17 .6%), and type 3 in 2 patients (5.9%). A similar study at the ORL-HNS polyclinic RSHS/FKUP for the 2012-2015 period showed that, out of 107 patients involved in the study, type 1 was the most common classification obtained. Where 62 out of 107 patients had laryngomalacia type 1.[9] Research in Surabaya showed that of 76 patients, type 1 was the most common type, whereas 69 patients had type 1 laryngomalacia.[16] This study uses a classification division according to what was disclosed by Olney with the addition of a combined classification. Where Olney divides laryngomalacia into 3 types based on laryngoscopy appearance. In type 1 laryngomalacia, there is a prolapsed appearance of the prolapsed mucosa over the arytenoid cartilage. In type 2, the aryepiglottic folds shorten, causing lateral collapse. The hallmark of type 3 laryngomalacia is that the epiglottis collapses posteriorly.[2,7] In the combined type, two or more types are found at the time of laryngoscopy examination. There is no definite theory that can be used to explain the causes of laryngomalacia and the causes of the anatomic variations in this disease, although several theories have been put forward to explain the causes of supraglottic airway collapse, including the theory of impaired neuromuscular control, the rigidity of laryngeal cartilages, and laryngeal inflammation.[22]

Table 5. Frequency distribution of laryngomalacia patients based on severity (n=34).

Degree of severity	Total
Mild	21 (61,8%)
Moderate	3 (8,8%)
Severe	10 (29,4%)

Table 5 shows the frequency distribution of the severity of laryngomalacia patients. Laryngomalacia patients mostly came with a mild degree of severity, with a total of 21 patients (61.8%). Although there is no widely used standard to assess the severity of laryngomalacia patients, this degree of severity can be assessed by the severity of the patient's respiratory symptoms and eating disorders. In mild symptoms, patients usually experience inspiratory stridor accompanied by difficulty eating, but the patient's coordination of eating and breathing is still good, so it does not lead to hypoxic conditions. Even so, there is still a possibility that patients will experience a worsening of symptoms to a moderate degree. At moderate degrees, more patients come with complaints of difficulty eating, which, if not treated immediately, can result in aspiration and weight loss. In severe symptoms, patients may experience cyanosis, respiratory arrest, difficulty eating, aspiration, and growth faltering, which, if not treated immediately, can threaten the patient's safety.[6,8] However, this degree of severity cannot be used to estimate the final outcome and improvement of the patient's disease. Several factors influence the severity and improvement of this disease, such as the number of medical comorbidities, the presence of secondary airway lesions, and the average oxygen saturation.[6,8]

Table 6. Frequency distribution of laryngomalacia patients based on comorbid factors (n=34).

Comorbid	Total
LPR (laryngopharyngeal reflux) or GERD (gastroesophageal reflux disease)	1 (2,9%)
Congenital syndrome	6 (17,6%)
Neurological disease	13 (35,1%)
Heart disorder	7 (20,6%)
Craniofacial abnormalities	5 (14,7%)
Malnutrition	10 (29,4%)
Pneumonia	8 (23,5%)
No comorbid factors	9 (26,5%)

In this study, it was found that a laryngomalacia patient could have more than one medical comorbidity. Based on the data shown in Table 6, it is known that most laryngomalacia patients have medical comorbidities in the form of neurological diseases, with a total of 13 patients (35.1%). The fewest medical comorbidities accompanying laryngomalacia patients were LPR or GERD, with a total of 1 patient (2.9%). A similar study showed that, out of 76 patients, the most common comorbid factors were neurological diseases in 16 patients.[16] Other studies have shown results where children with cerebral palsy are 12 times more at risk of having persistent laryngomalacia compared to other neurological dysfunctions.[24] So far, there is no definite relationship between co-morbidities in the form of neurological disease and laryngomalacia. However, the high incidence of neurological disease in laryngomalacia is believed to have a relationship in accordance with the existing neurological theory as the most trusted etiological theory.[6] Neurological theory suggests that laryngomalacia may be a consequence of an underdeveloped or abnormally integrated central nervous system, especially the peripheral nerves and brainstem nuclei which are responsible for breathing and airway patency.[16] Neurological disease is also

thought to affect the function of the vagus nerve and laryngeal tone at the central nervous system or brainstem level, thereby contributing to the severity of symptoms.[6]

Table 7. Frequency distribution of laryngomalacia patients based on the type of therapy (n=34).

Types of therapy	Total
Medicamentosa	32 (94,1%)
Medicamentosa and surgery	2 (5,9%)

Table 7 shows the frequency distribution of the types of therapy performed by laryngomalacia patients. The majority of laryngomalacia patients were treated with medical therapy, with a total of 32 patients (94.1%). In comparison, the other 2 patients (5.9%) were given medical therapy accompanied by surgery. A similar study showed that of 76 patients, 73 of them underwent conservative therapy, and 3 others had a tracheostomy. Conservative applied in the form of dietary modifications, installation of a nasogastric tube, anti-reflux medication, nebulizer, and follow-up routine every 1-6 months with FOL examination. Surgical therapy is performed with a tracheostomy to widen the airway.[16] Generally, laryngomalacia patients can be given observational and medical therapy as a treatment. However, special attention is still needed in patients with dyscoordination in breathing and swallowing.[3] Administration of pharmacological treatment in the form of acid suppression has been shown to improve symptoms of eating disorders such as aspiration, choking, or regurgitation.[2,6] Pharmacological management is carried out by administering omeprazole with a dose calculated according to the child's age and weight. Administration of medicinal preparations (omeprazole) to children can be done by mixing the contents of the capsule with fruit juice such as cranberry syrup or yogurt. In addition to providing medical therapy, the patient's parents need to be given education regarding actions when choking, feeding procedures and patterns, and monitoring weight gain.[25] It is also necessary to make changes to the patient's diet, including condensing milk and positioning the child upright while eating at a slower tempo.[26] Meanwhile, in patients with severe symptoms with clinical features of cyanosis or apnea when eating or sleeping and in patients who have severe airway obstruction, surgery can be performed. This operative action aims to improve the patient's airway and feeding canal to prevent growth faltering and aspiration.[6]

4. Conclusion

Laryngomalacia patients at ORL-HNS Dr. Mohammad Hoesin General Hospital Palembang for the 2021-2022 period mostly came from the age group 3-12 months (52.9%), and most of the patients were male (73.5%). The most common clinical symptom in patients was difficulty eating (51.4%). Based on the appearance of FOL, the type that was found in most patients was type 1 (53.9%). Most patients came with a mild degree of severity (61.8%). The comorbid factors that most often accompany laryngomalacia patients are neurological diseases (35.1%), and the majority of laryngomalacia patients are treated using medical therapy (94.1%).

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