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Clinical Evaluation vs Endoscopic Findings in Laryngomalacia at a Tertiary Institution in Northern Nigeria

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Abstract

Background: *Laryngomalacia is the commonest congenital abnormality of the larynx. It the most important cause of stridor in neonates and infants stridor in the main presenting symptoms. In majority of the patient seen especially in peripheral health centers clinical symptoms and signs are relied upon for the diagnosis of the condition. Flexible laryngoscopy is however the mainstay of diagnosis.* ***Objective:*** *The aim of this study is to compare clinical findings and endoscopic findings of laryngomalacia in department of Ear, Nose and Throat of Aminu Kano Teaching Hospital Northern Nigeria.* ***Material and Methods:*** *We undertook a prospective cross – sectional review of all patient with larynomalacia who presented at the department of ENT Aminu Kano Teaching Hospital between June 2012 to May 2014. Bio – data and clinical information were obtained and stridor graded. Then flexible fiber optic laryngoscopy was done on the entire patient and the stage of endoscopic grading documented.* ***RESULT:*** *A total of 35 children were seen during the study period and 26 met the inclusion criteria and were enrolled. There were 17 (65.4%) males and 9 (34.6%) females with a male female ratio of 2.1:1. There ages ranged from 1day to1year. The mean age at presentation was 5 ± 2.71 months. Twenty six (100%) of the children presented with different degrees of stridor. Based on, the clinical staging of stridor the degree of stridor found was 21(80.8%) had grade 1 to 2 stridor, and 5 (19.2%) had grade 3 none was found to have grade 4 stridor. Clinical assessment of the severity of the condition by the same investigators, 17 (65.4%) of the patients had mild laryngomalacia, 5 (19.2%) had moderate laryngomalacia and 4 (15.4%) had severe laryngomalacia. Objective endoscopic grading of findings in these patients showed 18 (69.2%) of the patients had grade 1 to 2, 3 (11.5%) had grade 3 and 5 (19.2%) had grade 4 to 5 laryngomalacia. Using the Spearman rho correlation coefficient test, we found a significant strong positive correlation between clinical assessment and endoscopic grading of laryngomalacia (r=0.860, P=0.000).****Conclusion:*** *clinical evaluation still has significant reliability in the diagnosis of laryngomalacia. But flexible laryngoscopy remains the mainstay of diagnosis and all patients with laryngomalacia should have flexible laryngoscopy.*

Keywords: *Laryngomalacia, Stridor, flexible laryngoscopy.*

Introduction

Laryngomalacia is a condition in which the laryngeal inlet collapses on inspiration causing stridor. Laryngomalacia is the most common cause of stridor in newborns affecting 45 – 75% of all infants with congenital stridor [(1)].

Congenital flaccid larynx (laryngomalacia) is secondary to flaccidity and in coordination of the supraglottic cartilage and mucosa of the arytenoids, aryepiglottic folds and epiglottis. Patient with this disease have few symptoms at birth, but over the first few weeks of life gradually develop high pitched inspiratory stridor and occasionally feeding difficulties. Diagnosis must be made while observing the functioning larynx [(3)]. Laryngomalacia can be diagnosing based on symptoms and signs, flexible laryngoscopy, and lateral radiographs of

the neck and chest. Rigid endoscopy is done to identify possible synchronous airway lesion such as subglottic stenosis. Flexible laryngoscopy has been shown to be effective in neonates [3]. In an awake patient flexible scope has the advantage of appreciating the dynamic of supraglottis. The disadvantage is that milder form of laryngomalacia may be missed when the child is crying [3]. 233 patients with laryngomalacia were evaluated by Mancuso et al respectively to assess the necessity of performing a complete diagnostic work up in all patient[3], [18].

Reassuring the parent remains the mainstay of treatment in most cases because the condition is usually self – limited. Complete resolution may take as long as 18 month. Rarely intervention may be necessary either by placing a tracheostomy or preforming supraglottoplasty. The range or spectrum of the disease varies in presentation, progression and outcome. The impact or effect of the disease on parents/care givers is sometimes detrimental and their subjective assessment therefore may not tally well with clinical findings.

Literature review aetiology and incidence

Although the exact aetiology of larymgomalacia is unknown. Fort et al however found an autosomal dominance relationship in some children [4]. In a study in which 453 patient were evaluated by Hawkins and clark with flexible fiberscope, found 84 with primary and 29 with secondary laryngomalacia[5]. Respiratory disorders were noted in 68 of 297 children examined with flexible bronchospe [6]. Hain et al in their study on the prevalance of Gastroesophageal reflux in children with tracheomalacia and laryngomalacia found 70% of the children with laryngomalacia had GER [7]. Thompson in his paper on the theory of etiology estimated the incidence of associated congenital anomalies/genetic disorder as between 8 – 20%. [1],[8]

Signs and symptoms

The most common symptoms in patient with laryngomalacia include inspiratory stridor, feeding problems and Gastric reflux[7]. Increase in respiratory effort such as crying or feeding usually worsens the respiratory stridor. Symptoms appear shortly after birth in most patient [9]. It increase in severity until 6 – 8 month of age, plateau at 9 months and steadily improves thereafter [10][11]. There is no correlation noted to exist between the duration of stridor and the severity or time of onset [9].

There are two theories proposed to be accounting for the narrowing of the laryngeal inlet the first is that a neuromascular abnormality leads to improper support of the supraglottis causing increased flaccidity [12]. Children with laryngomalacia were found to have a higher incidence of Neuromascular abnormality[10].

The other proposed theory responsible for laryngeal inlet narrowing is anatomic from (1) a flaccid epiglottis that folds against posterior pharyngeal wall or into the airway (2) a long, tubular epiglottis that curl on itself (3)short and redundant aryepiglottic folds with varying sizes of cuneiform cartilage that rotate medially into the airways [13]. The W – Shaped epiglottic occurs in 30 – 50% of patient and is thought not be significant [14] [16].

In another study radiologic evidence of gastric reflux occurs in 80% and regurgitation in 40% [15]. Zalzal and associate found 5 out of 21 patients (24%) with laryngomalacia have feeding problem [17]. Aspiration pneumonitis was seen 7% [12]. Obstructive sleep apnoe and central apnoe were noted in 23% and 10% respectively.

Lauren et al in their study noted that prospective quality of life assessment of children with laryngomalacia and their families caused significant burden of disease. In addition to quality of life improves in all patient but may improve more significantly in patient manage surgically. [19] Prasad et al found that epiglottic prolapse correlated with severity of laryngomalacia and cuneiform prolapse with swallowing dysfunction. They noted swallowing dysfunction had a significant emotional impact on parental life[20]

Aims and objective

General aim: to compare clinical findings with flexible endoscopic findings in patient with laryngomalacia.

Specific

1. To assess clinical findings in patient with laryngomalacia.
2. To assess endoscopic findings in patient with laryngomalacia.
3. To compare clinical findings with endoscopic finding in patient with laryngomalacia.

Methodology

This study was prospective cross-sectional survey and involved all consecutive children with features of laryngomalacia that presented to the Ear Nose and Throat clinic of Aminu Kano Teaching Hospital Nigeria between June 2012 and May 2014.

Study design

All patients who presented to the Ear Nose and Throat clinic of Aminu Kano Teaching Hospital during the study period were reviewed by the investigators. And those children who presented with symptoms suggestive of laryngomalacia included into the study. Those whose parents or care givers did not consent and those who did not tolerate flexible laryngoscopy were excluded from the study.

Information regarding patient Bio data and clinical symptoms of laryngomalacia were obtained from the patient or caregivers. Afterwards, all the children had a detailed clinical examination by both investigators at the same sitting.

The degree of stridor was graded using the clinical staging for stridor[21]

1. *Grade 1* (Exertional stridor) : Stridor appears during crying or exercise.
2. *Grade 2* (Continuous stridor or stridor at rest) : Stridor is present at rest & become worse
3. with exertion. *Infants < 1 yr of age should be hospitalized.*
4. *Grade 3* (Stridor with retractions) : Stridor is continuous & accompanied with suprasternal & supraclavicular retractions. The patient looks anxious, irritable, & struggling for
5. breathing. *Hospitalization is indicated for all cases.*
6. *Grade 4* (Stridor with cyanosis) : In addition to continuous stridor & retractions, cyanosis & altered consciousness occur denoting severe respiratory failure. *Urgent hospitalization & ET intubation are indicated.*

In addition, they all had laryngoscopy using a 300mm Pentax flexible fibreoptic laryngoscope with 3-50mm depth of field and tip diameter 3.5mm was use. The Hollinger's classification (23) model was used to grade the endoscopic findings of the patients.

Hollinger classification laryngomalacia was used to grade the endoscopic findings and is as follows[22]

Grade	Description
1	Collapse of A – E folds
2	Tubular Epiglottis with inward collapse
3	Collapse of corniculate and cuneiform cartilage
4	Posterior collapse of epiglottis
5	Shortened A – E folds

This study conformed to the Code of Ethics of the World Medical Association (Declaration of Helsinki) and was approved by the Ethical Review Committee of Aminu Kano Teaching Hospital, Kano, Nigeria.

Data management

Data obtained were analyzed using IBM SPSS version 21 (for windows). The Spearman rho test was used to test correlation between the variables. The statistical level of significance was when $P < 0.05$.

Data were collected and results were tallied and analysed.

Results

A total of 35 children were seen during the study period and 26 met the inclusion criteria and were enrolled. There were 17 (65.4%) males and 9 (34.6%) females with a male female ratio of 2.1:1. There ages ranged from 1day to1year. The mean age at presentation was 5 ± 2.71 months. The duration of symptoms ranged from 1 day to 1 year and the mean duration of symptom was 5 ± 2.71 months

Twenty six (100%) of the children presented with different degrees of stridor. Based on, the clinical staging of stridor the degree of stridor found was 21(80.8%) had grade 1 to 2 stridor, and 5 (19.2%) had grade 3 none was found to have grade 4 stridor. Other findings at presentation included 6 (23.1%) had retarded growth and development, 5 (19.2%) had pectus excavatum, 2 (7.7%) had sleep apnea, 2 (7.7%) had moderate to severe airway obstruction and 2 (7.7%) had other associated co- morbid conditions.

Based on subjective clinical assessment of the severity of the condition by the same investigators, 17 (65.4%) of the patients had mild laryngomalacia, 5 (19.2%) had moderate laryngomalacia and 4 (15.4%) had severe laryngomalacia. Objective endoscopic grading of findings in these patients showed 18 (69.2%) of the patients had grade 1 to 2, 3 (11.5%) had grade 3 and 5 (19.2%) had grade 4 to 5 laryngomalacia.

Using the Spearman rho correlation coefficient test, we found a significant strong positive correlation between clinical assessment and endoscopic grading of laryngomalacia ($r=0.860$, $P=0.000$).

Discussion

Laryngomalacia affecting 45 – 75 of infants with congenital stridor is the most common cause of stridor in the newborn.

In this study the entire patient seen presented with stridor at birth indicating that all of them have the congenital types of laryngomalacia. The onset tallied with findings by Pitcher and Thompson in their study on surgical management of larybgomalacia[3] and also April and Thompson[1]. 65.5% of the patient was males with males to female ratio of 2: 1: 1. This showing slight male preponderance and same was documented by D.M Thompson [8] and April and Thompson [1].

Majority of the patient presented before the age 6 month and 61.5% presented were at the age 2 – 6 month. This is in keeping with time of increasing severity in the natural history of the diseases[1][9]. The increasing symptom before it plateau at 9 month necessitates parent to seek for medical attention.

Out of the 4 patient (15%) that presented at 1 year of age despite the onset of symptoms at birth among which 2 (7.5%) were form rural areas. The other two have associated congenital abnormality which parent pay attention to before considering the stridor.

The entire patients were found to have varying degrees of stridor. This is in keeping with many other studies with stridor being the main symptom at presentation.[1][3],[4][7][10]. None had grade IV stridor out of those patient with higher grade of stridor 2 had associated congenital anomalies. This showed that only few (15.3%) had severe layngomalacia. Finding in many studies indicated that the disease is self limiting and only few presents with severity that may necessitates surgical intervention. [1][2][4][5][14][17]

Gastric and feeding problems were noticed in 34.6%. A higher percentage was found by Haim et al[7] although regurgitation was noticed in 40% of patient and feeding problem in only 15% of patient with laryngomalacia by Zalzal[17].

Abnormal cry as well as growth and developmental abnormities was noticed in 23% of the patient, 3 out of 26(11.5%) had paltus excavatum and was noticed among those who presented late.7.6% had sleep apnoea April M. Landry and Dana M. Thompson list abnormal cry growth abnormalities, sleep apnoea and pectus excavation among less common symptoms of laryngomalacia[(1)]. Belmont JR and Grund Fast K found central apnoea in 10% of patient they studied.[(14)] 2 patient had tracheostomy as a result of severe respiratory distress among which one patient had falling tongue due to microagnathesia and the other had associated lower respiratory to act infection nor seeing the clinical condition. April and Thompson showed that surgical intervention improves symptom in severe cases and severity of laryngomalacia is common among patient with associated genital abnormality.[(1)(8)]

2 patient (15.3%) had associated, microagnathesia and mylomemgocele. The two patient had severe form of the disease. The incidence of associated congenital abnormality/syndromes and genetic disorder was estimated as 8 – 20%. The disease is severe in those associated with congenital abnormality[(1),(2),(8)]

This study found a strong positive correlation between clinical findings and endoscopic assessment. Therefore clinical finding can be relied upon to gauge the endoscopic finding, however, still remains superior to clinical evaluation alone because in this study objective endoscopic grading revealed a higher percentage of patient with mild form of the disease than the clinical evaluation alone. Even though flexible laryngoscopy has the advantage of likely missing some mild form of the disease when the child is crying, [(3)] it still remains superior to clinical evaluation alone[(8)].

Conclusion

Laryngomalacia as an important and most common cause of stridor in neonates can be diagnosed based on clinical symptoms and signs, however flexible laryngoscopy remains superior and is the gold standard on the diagnosis of laryngomalacia despite the strong positive correlation found by this study.

Recommendations

1. Clinicians practicing in rural and peripheral health centre may rely on clinical findings in the management of larngomalacia when facility for flexible laryngoscope is not available.
2. Laryngoscopy remains the gold standard in the diagnosis and treatment of patient with laryngomalacia. Therefore all patient with laryngomalacia should have laryngoscopy as part of their clinical assessment.

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