



## Prevalence of expiratory central airway collapse in children undergoing flexible fiberoptic bronchoscopy

Rehab Zaki Elmeazawy, MSc.<sup>1</sup>, Nader Abdelmonem Fasseeh, MD<sup>2</sup>, Amr Ahmed Mubarak, MD.<sup>3</sup>, Ahmad Mohammed Abdelrazik, MD.<sup>1</sup>, Nabil Moustafa Elesawy, MD.<sup>1</sup>

<sup>1</sup>Department of Pediatrics, Faculty of Medicine, Tanta University, Tanta, Egypt

<sup>2</sup>Department of Pediatrics, Faculty of Medicine, Alexandria University, Alexandria, Egypt

<sup>3</sup>Department of Diagnostic radiology, Faculty of Medicine, Tanta University, Tanta, Egypt

[rehab\\_zaki2013@yahoo.com](mailto:rehab_zaki2013@yahoo.com)

**Abstract:** Objectives: To determine the prevalence of expiratory central airway collapse in children undergoing flexible bronchoscopy. **Methods:** This study was carried out on 113 patients below the age of 16 years who have symptoms and signs suggestive of expiratory central airway collapse. Thirty eight patients (20 male and 18 female) were diagnosed with expiratory central airway collapse by flexible bronchoscopy. End-inspiratory and dynamic expiratory cross-sectional airway areas at aorta, carina and bronchus intermedius were recorded in order to assess the degree of dynamic airway collapse by cine CT in 23 of the 38 patients included in the study and the percentage of luminal collapse was calculated. Results: Thirty eight patients (20 male and 18 female; median age 13 months with range 3-39 months) were actually diagnosed with expiratory central airway collapse, indicating a prevalence of 33.6% of the disease. The most common presenting symptoms were recurrent chest infection in 14 patients (36.8%), stridor in 13 patients (34.2%), wheeze in 9 patients (23.7%), cough in 9 patients (23.7%), rattling in 6 patients (15.8%) and cyanosis in 1 patient (2.6%). Conclusion: Flexible bronchoscopy is the “gold standard” for diagnosing and assessing changes in airway luminal size associated with expiratory central airway collapse.

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**Keywords:** Expiratory central airway collapse, Bronchoscope, Children

### 1. Introduction:

Expiratory Central Airway Collapse (ECAC) is defined by excessive airway collapse during expiration either from cartilaginous weakening or redundancy of the posterior membranous wall and includes two entities tracheobronchomalacia (TBM) and excessive dynamic airway collapse (EDAC) (1).

Excessive Dynamic Airway Collapse (EDAC) refers to abnormal and exaggerated bulging of the posterior membrane within the airway lumen during exhalation causing a reduction in cross sectional area by 50 % or more (2).

Tracheobronchomalacia (TBM) is weakness of the tracheal and / or bronchial walls and supporting cartilages in a diffuse or segmental distribution with the resultant collapsibility of central airways (3).

The term tracheobronchomalacia (TBM) and excessive dynamic airway collapse (EDAC) are often used interchangeably, but they are different in pathophysiology and morphology. Although sometimes they coexist in the same patient (4).

The prevalence of TBM and EDAC depends on the patient age, sex, associated co-morbidities and

underlying etiologies, diagnostic tools and criteria used to define the airway collapse while prevalence of stand-alone EDAC still unclear (5).

Expiratory Central Airway Collapse presents clinically with nonspecific respiratory symptoms. In neonates clinical features include harsh cough, central wheeze, dyspnea, gasping, stridor, feeding difficulties and blue spells. In older infants expiratory wheeze, chronic cough, apneic episodes and recurrent respiratory tract infections due to impaired clearance of secretions are common symptoms (6).

Bronchoscopy is a valuable tool in the diagnostic work up of TBM and EDAC. Bronchoscopy allows for the real time evaluation of the tracheal and bronchial tree with tidal respirations and with forced expiratory maneuvers. Bronchoscopy with airway exam is considered the gold standard to diagnose TBM and EDAC (7).

### **Methods:**

#### **Study design:**

This is a cross-sectional prospective study that enrolled 113 patients below the age of 16 years who had symptoms and signs suggestive of expiratory

central airway collapse and were referred to Tanta university hospitals and Alexandria university hospitals between July 2016 to July 2018.

#### Study population:

All children  $\leq 16$  years of age who were clinically suspected to have expiratory central airway collapse and presented with one or more of these symptoms; persistent stridor, persistent wheeze, barking cough, anoxic spells, recurrent pulmonary infections, apnea and persistent respiratory noisy breathing; rattling, grunting were subjected to informed consent, thorough history taking and clinical examination. They were also subjected to flexible bronchoscopy.

#### Bronchoscopy:

Flexible fiberoptic bronchoscopy was carried out on 113 pediatric patients as a part of their routine workup diagnosis. We used two sizes of flexible bronchoscopy (Olympus BF-3C160 video bronchoscope with external diameter 3.7mm & working channel 1.2mm) or (KARL STORZ with external diameter 2.8mm & working channel 1.2mm) according to the patient's age under general anesthesia laryngeal mask ventilation with spontaneous respiration.

During bronchoscopy bowing of the cartilage wall as well as excessive invagination of the posterior wall of the trachea and bronchi to the point where the bronchoscope cannot be advanced any further were observed and expiratory central airway collapse was defined as  $\geq 50\%$  reduction of the airway lumen.

#### Statistical Analysis

Data were collected, revised and edited into a master table using Microsoft Excel 2013. Data were then revised, coded and entered to the statistical package for social science (SPSS) version 22. Patient demographic characteristics were presented as mean and standard deviation for continuous variables with normal distribution, median and interquartile range (IQR) for continuous variables with non-normal distributions and as proportions (percentages) for categorical variables. The distributions of quantitative variables were tested for normality using Kolmogorov-Smirnov test, Shapiro-Wilk test and D'Agstino test, also Histogram and QQ plot were used for vision test. If it reveals normal data distribution, parametric tests were applied. If the data were abnormally distributed, non-parametric tests were used. Related sample testing was done to compare results of both diagnostic modalities. The confidence interval was set to 95% and the margin of error accepted was set to 5%. So the p-value was considered significant if it's  $<0.05$ .

### 3. Results:

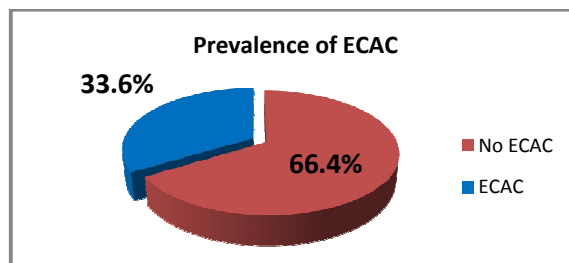


Figure 1. A pie chart showing prevalence of ECAC.

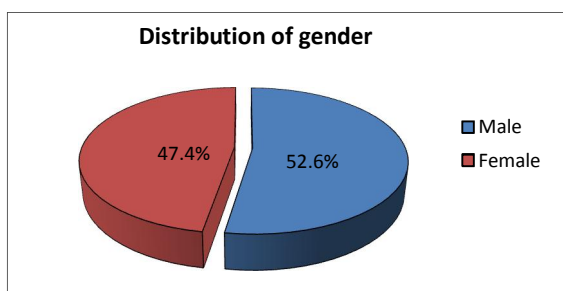
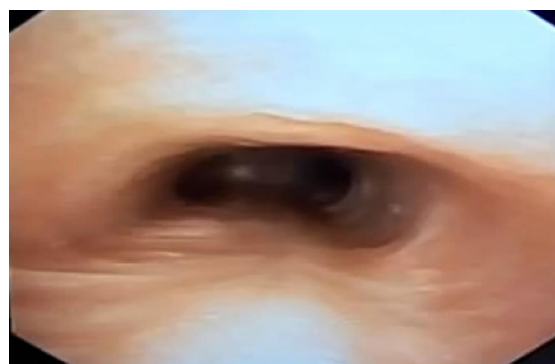


Figure 2. A pie chart showing gender distribution among study population.

#### Flexible Bronchoscopy



Inspiration



Expiration

Figure 3. Bronchoscopic image of trachea during inspiration and expiration of the previous male patient which show moderate degree of airway collapse 75% during expiration.

Out of the 113 patients included in the study who presented with clinical picture suggestive of expiratory central airway collapse, only 38 patients (20 male and 18 female; median age 13 months with range 3-39 months) were diagnosed by flexible bronchoscopy as expiratory central airway collapse, indicating a prevalence of 33.6% of the disease (Figure 1,2).

The age of first symptoms had median age 1 month with range 1 – 4months, this denotes that 50% of study population had their first symptoms during the first month of life and 75% of population had their

first symptoms till before the age of 4 months (Table 1).

The most common presenting symptoms were recurrent chest infection in 14 patients (36.8%), stridor in 13 patients (34.2%), wheeze in 9 patients (23.7%), cough in 9 patients (23.7%), rattling in 6patients (15.8%) and cyanosis in 1 patient (2.6%) (Table 2).

Out of 38 patients 22 (57.9 %) patients had primary (idiopathic) cause of ECAC and 16 (42.1 %) had secondary cause of ECAC (Table 3).

**Table 1. Distribution of demographic data among study population.**

Parameter	Description
<b>Age (months)</b>	13 (3, 39)
<b>Range</b>	1 – 144
<b>Age of first symptoms (months)</b>	1 (1, 4)
<b>Range</b>	1 – 132
<b>Gender (male)</b>	20 (52.6%)
* <b>Categorical variables are presented as proportions (percentages). Continuous variables with non-normal distributions are presented as median and interquartile range.</b>	

**Table 2. Distribution of presenting manifestations among study population.**

Parameter	[No. (%)]
<b>Recurrent chest infection</b>	14 (36.8%)
<b>Stridor</b>	13 (34.2%)
<b>Rattling</b>	6 (15.8%)
<b>Recurrent wheeze</b>	6 (15.8%)
<b>Aspiration</b>	5 (13.2%)
<b>Wet cough</b>	5 (13.2%)
<b>Barking cough</b>	4 (10.5%)
<b>Persistent wheeze</b>	3 (7.9%)
<b>Cyanosis</b>	1 (2.6%)

**Table 3. Distribution of the etiology of ECAC.**

Parameter	[No. (%)]
<b>Primary (dynamic) airway narrowing</b>	<b>22 (57.9%)</b>
1. EDAC	16 (42.1%)
2. Tracheobronchomalacia	6 (15.7%)
<b>Secondary (fixed) airway narrowing</b>	<b>16 (42.1%)</b>
1. Vascular ring	11(28.9%)
2. TEF	2 (5.2%)
3. Bronchogenic cyst	1 (2.6%)
4. Mediastinalteratoma	1 (2.6%)
5. HLH	1 (2.6%)

#### 4. Discussion

The prevalence of ECAC is variable in different studies especially in pediatrics as there are no many studies about this issue. Our study showed that the prevalence of ECAC in pediatric populations who underwent bronchoscopy was 33.6%. In study done by Nygaard et, al. they found that the prevalence of tracheal collapse in adult patients varied from 10.7%

to 19.5% when using an expiratory collapse of 50% as a threshold (8).

Our study was concordant with the study done by Mair and Parsons which was carried out on Thirty-eight infants and toddlers 3 years old and younger. They found that major airway collapse presented in 30% of 129 children who underwent bronchoscopy for respiratory distress (9).

In our study on 113 pediatric patients underwent flexible bronchoscopy because of recurrent chest infection, stridor, barking cough, recurrent wheeze, persistent wheeze, rattling, aspiration and cyanosis there were 38 patients diagnosed with ECAC and the other 75 patients had different diagnosis.

This comes in agreement with Pan et al, who found in their large study on 2749 children presented with chronic cough, recurrent wheeze, recurrent airway infections, atelectasis etc., for performing fiberoptic bronchoscopy, among whom 459 children were diagnosed with airway malacia and they represented 16.70% of all the patients underwent bronchoscopy due to the same symptoms and signs (10).

Regarding the origin of the disease (57.9%) of our patients had idiopathic ECAC while (42.1%) of our patients have secondary ECAC. And this is in disagreement with study done by Murgu and Colt who found that (5.5%) patient had idiopathic origin and (94.4%) patients with secondary origin. The difference may be because their study was on adults patients (11).

Boogaard et al found that airway malacia was diagnosed in 160 children (94 males) at a median age of 4.0 years. The airway malacia was classified as primary in 136 (85%) children and secondary in 24 (15%) children and this concordant with our study regarding the origin of central airway collapse in children. From these studies we can notice that the origin of ECAC in pediatrics differ from that in adults (12).

According to the previous literatures, the secondary causes of ECAC were mainly tracheoesophageal fistula, vascular malformations, congenital cysts, mass lesions causing mechanical compression, malignancy and any process causing chronic irritation/inflammation of the airway (ie, aspiration, reflux, chronic asthma). We found in our study that the most common causes of secondary ECAC was external vascular compression and it represent 28.9% of the secondary causes, while TEF was noticed in 5.2%. The least causes was bronchogenic cyst, mediastinalteratoma and HLH each represented by 2.6% of the secondary causes of ECAC.

In our study on the severity of ECAC, we used the cut off value  $\geq 50\%$  reduction in the cross-sectional area of airway to diagnose ECAC by flexible bronchoscopy. Majid et al 2014 reported in their study that a decrease in tracheal lumen size by 50% or less during exhalation is regarded as nonpathologic, between 51 and 75% as mild obstruction, between 76 and 90% as moderate obstruction, and between 91 and 100% as severe obstruction (13).

Weinstein and coworkers in their study, used a 75% cutoff value, which is a reasonable criterion to avoid false positives (14).

#### **Limitations of our study:**

This prospective study had several limitations. First, bronchoscopy was not performed in controls. However, bronchoscopy is an invasive procedure particularly in children as it is difficult to justify in a control group of young children. Second, another limitation of our study is that the lumen size was subjectively estimated by anteroposterior diameter and not quantitatively measured by cross-sectional area by bronchoscopy.

#### **5. Conclusion**

Flexible bronchoscopy is the “gold standard” for diagnosing and assessing changes in airway luminal size associated with expiratory central airway collapse. The prevalence of TBM and EDAC depends upon the patient population, associated comorbidities and underlying etiologies, diagnostic tools used and criteria used to define the airway collapse.

#### **Declaration of Conflicting Interests**

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#### **Corresponding author:**

Rehab Zaki Elmeazawy, MSc.

Department of Pediatrics, Tanta University, Tanta, Egypt.

Email: [rehab\\_zaki2013@yahoo.com](mailto:rehab_zaki2013@yahoo.com)

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