The Egyptian Journal of Hospital Medicine (October 2024) Vol. 97, Page 3515-3519

Clinical Staging of Combined Laryngoceles: A Novel Concept

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ABSTRACT

Background*:* Virchow described laryngocele for the first time in 1867 as a herniation of the laryngeal ventricle or "laryngocele ventricularis", while Dominique Larrey used the term "goitre aeriennes" to describe the condition in a case for the first time.

Aim: To achieve precise clinical staging of combined laryngoceles based on anatomical internal extension. This aim was achieved by analyzing our case series and by validating this staging system (correlating between patient complaint, endoscopic, and imaging findings).

Patients and methods: This retrospective study included a total of 24 cases with combined laryngoceles admitted to the Otorhinolaryngology Department, Mansoura University hospital, Mansoura, Egypt between 1998 to 2022.

Results: The study found that all cases had an external component detected. There were 16 cases with V1 grade (ventricle), 66.7% with V2 grade (ventricle and vestibule), and 16.7% with V3 grade (ventricle with vestibule and vallecula). There was no significant difference in dysphonia, aerodigestive obstruction, and dysphagia between cases with and without air content. However, cases with air content had a higher incidence of stridor and dyspnea. No significant difference was found in endoscopic grading based on air content within the laryngocele.

Conclusion*:* Clinical staging of combined laryngoceles (V1, V2, V3) is a simple, feasible, and reliable method validated clinically, endoscopically, and radiologically, avoiding bias.

Keywords: Laryngoceles; Endoscopic; Imaging findings.

INTRODUCTION

Virchow described laryngocele for the first time in 1867 as a herniation of the laryngeal ventricle or "laryngocele ventricularis", while Dominique Larrey used the term "goitre aeriennes" to describe the condition in a case for the first time [1,2].

The ventricle is a laryngeal dilatation of a fusiform shape between the true and false vocal cords, extending from the thyroid notch to the arytenoids. It communicates anteriorly with a pouch called the saccule. A membranous sac with ciliated pseudo-stratified cylindrical epithelium lining it and a thin basal membrane containing a variable number of goblet cells make up its histological structure [1].

A space known as a laryngocele arises due to the laryngeal saccule's pathological dilatation. They reach higher inside the false vocal fold and communicate with the larynx. It is possible to categorize these uncommon lesions as internal or combined. While combined laryngoceles are situated both medially and laterally to the thyrohyoid membrane, internal laryngoceles radiate medially towards it [3].

Congenital conditions, elevated laryngeal pressure, and mechanical obstruction are a few of the many theories that explain the laryngocele formation [4].

Because the mechanical obstruction usually occurs due to malignant causes, it has a great importance at the clinical level; men over 50 are typically affected and the majority are of the combined and unilateral types [2,3].

Although laryngoceles normally contain air, if the neck becomes obstructed, they may also contain mucus. Additionally, an infection of the dilated saccule may result in a laryngopyocele, which may cause a substantial supraglottic edema and mechanical obstruction. Sometimes laryngophones can appear as severe airway emergencies [4].

The aim of the current study was to achieve precise clinical staging of combined laryngoceles based on anatomical internal extension. This aim was achieved by analyzing our case series and by validating this staging system (correlating between patient complaint, endoscopic, and imaging findings).

PATIENTS AND METHODS

This retrospective study included a total of 24 cases with combined laryngoceles admitted to the Otorhinolaryngology Department, Mansoura University Hospital, Mansoura, Egypt between 1998 to 2022.

Inclusion criteria: Patients with combined laryngoceles who had complete data of CT and MRI / telescopic assessment and follow up.

Exclusion criteria: Internal laryngoceles and 2ry laryngoceles, patients with other malignant diseases, patients with prior laryngeal surgeries and incomplete data.

Procedure and assessment

The records were reviewed to obtain full history with special focus to the duration and severity of symptoms (dysphonia, stridor, dyspnea, dysphagia, acute upper airway obstruction, external cervical swelling) and any history of previous endoscopic or surgical procedure. Complete upper airway endoscopic examination was conducted by using 70° rigid laryngoscope (4 mm) to assess the internal component. Computed tomography (CT) scanning of the neck with contrast with 2-mm axial images and magnetic resonance imaging (MRI) were done to assess the components extension, content, communication with detailed evaluation of the saccule, para laryngeal space,

Received: 18/04/2024
Accepted: 18/06/2024

and the thyrohyoid membrane clearly demarcated. When building the staging system, we relied on the extension of the internal component as it is responsible for the main clinical presentation of the patients. Where V1 stage has ventricular extension (1st group), V2 has vestibular and ventricular (2nd group), while V3 (3rd group) has vallecular, vestibular, and ventricular extension. In order to validate the staging system proposed, we correlated this to clinical presentation to endoscopic and imaging results.

Ethical consideration

The study was conducted in accordance with Helsinki Standards as revised in 2013 [5]. The whole study design was approved by the local Research Committee, Faculty of Medicine, Mansoura University (Code: MS.22.05.2003). Confidentiality and personal privacy were respected in all levels of the study, collected data were/will not be used for any other purpose.

Statistical analysis

Data analysis packages were used SPSS version 21. Qualitative data were presented by number and percentage; quantitative data were presented by mean, standard deviation. Tests of significant were done by (chi square for qualitative, ANOVA test and Kruskal Wallis test for quantitative Parametric and non-parametric respectively) and level of significance were being set at P equal to or below 0.05.

RESULTS

The current study included 24 cases with combined laryngocele who were recruited from the Otorhinolaryngology Department, Mansoura University Hospital, Mansoura Faculty of Medicine. The mean age of the cases was 47.71 ± 9.80 years. All the included cases were males (Table 1).

Table (1): Demographic data in the cases of the study

Items		**Study subjects N = 24**	
Age (years)	Mean ± SD	47.71 ± 9.80	
	Median (min-max)	44 (35-75)	
		Number	**Percent**
Sex			
Male		24	100
Female		0	0

There was no statistically significant difference between the cases within the three groups of endoscopic grading regarding the age. Dysphonia was detected in all the cases. There was a statistically significant difference between the three groups of endoscopic grading regarding the presence of stridor and dyspnea. Our series showed three critical cases in stage V3 with acute aerodigestive obstruction. One of them died during induction of anesthesia and the other two cases were operated successfully. Dysphagia and aerodigestive obstruction were detected only in the cases with V3 endoscopic grading (Table 2).

Table (2): Comparison of the age and symptoms according to the endoscopic grading.

Variable	**V1 (Ventricle) N= 4**	**V2 (Ventricle and vestibule) N= 16**	**V3 (Ventricle with vestibule and vallecula) N= 4**	**Test of sig.**
Age (Years) [**Mean ± SD]**	48.25 ± 15.88	46.13 ± 7.68	53.50 ± 7.94	KW= 0.700 P = 0.705
Symptoms				
Dysphonia	4 (100%)	16 (100%)	4 (100%)	1
Stridor	2 (50%)	16 (100%)	4 (100%)	MC =10.909 P = 0.004*
Dyspnea	0 (0%)	16 (100%)	4 (100%)	MC = 24 P < 0.001*
Dysphagia	0 (0%)	0 (0%)	3 (75%)	MC = 17.143 P < 0.001*
Aerodigestive obstruction	0 (0%)	0 (0%)	3 (75%)	MC = 17.143 P < 0.001*

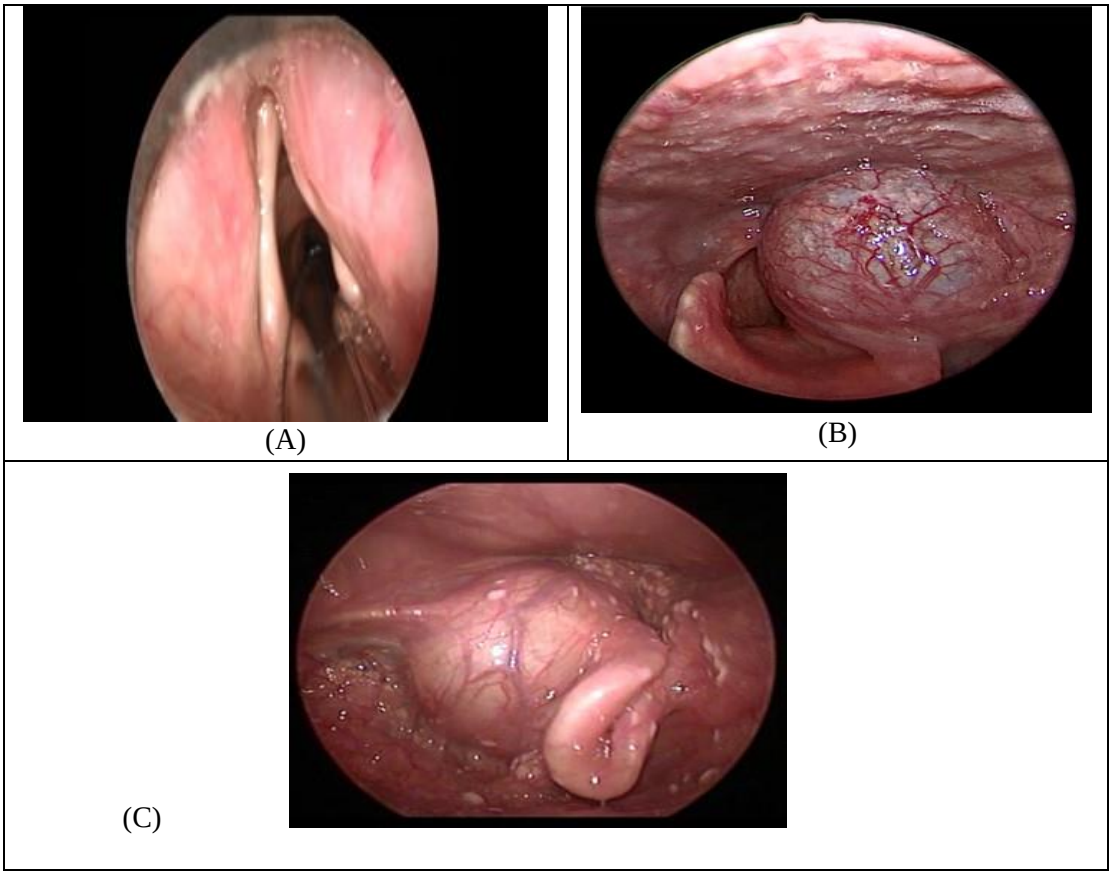
Categorical data expressed as number (%), MC= Monte-Carlo test, KW: Kruskal wallis test, *: statistically significant

This table shows that external component was detected in all the cases. According to the endoscopic and imaging findings, V2 grade (ventricle and vestibule) was the most common finding (Table 3).

Table (3): Clinical findings in the cases of the study.

Items	**Study cases N = 24**	
External component	**Number**	**Percent**
Present	24	100
Absent	0	0
Endoscopic and imaging findings (evaluating the internal component)		
V1 (ventricle)	4	16.7
V2 (ventricle and vestibule)	16	66.7
V3 (ventricle with vestibule and vallecula)	4	16.7
Incidence of site affection		
Ventricle	**24**	100
Vestibule	20	83.3
Vallecula	4	16.7

Figure 1 shows the different endoscopic grading.



(Figure 1): A: Endoscopic grading (V1), B: Endoscopic grading (V2), C: Endoscopic grading (V3).

Most of the cases (58.33%) had both fluid and air content (Table 4).

Table (4): Imaging findings (content) in the cases of the study.

Items	**Study cases N = 24**	
Imaging findings	**Number**	**Percent**
Air only	**3**	**12.5**
Fluid only	7	29.17
Fluid and air	14	58.33

Table 5 shows that there was no statistically significant difference in the presence of dysphonia, stridor, aerodigestive obstruction, and dysphagia between the cases with and without air content. However, there was statistically significantly higher incidence of dyspnea in cases with air content. There was also no statistically significant difference in the endoscopic grading according to the presence or absence of air content within the laryngoceles.
Pre- and post-operative dysphagia score non-significant (p = 0.05).

Table (5): Symptoms and endoscopic grading according to the air content.

Variable	**No air content N= 7**	**Air content N= 17**	**Test of sig.**
Symptoms			
Dysphonia (N%)	7 (100%)	17 (100%)	———
Stridor (N%)	5 (71.4%)	17 (100%)	FET =5.392 P = 0.076
Dyspnea (N%)	4 (57.1%)	16 (94.1%)	FET = 8.524 P = 0.017*
Aerodigestive obstruction (N%)	0 (0%)	3 (17.6%)	FET = 2.241 P = 0.530
Dysphagia (N%)	0 (0%)	3 (17.6%)	FET = 2.241 P = 0.530
Endoscopic grading			
V1 (ventricle) (N%)	3 (42.9%)	1 (5.9%)	MC = 4.941 P = 0.085
V2 (ventricle and vestibule) (N%)	3 (42.9%)	13 (76.5%)	
V3 (ventricle with vestibule and vallecula) (N%)	1 (14.3%)	3 (17.6%)	

Categorical data expressed as Number (%), FET= Fischer's exact test, MC= Monte-Carlo test, *: statistically significant.

DISCUSSION

The occurrence of laryngocele is incredibly uncommon, occurring in only one in every 2.5 million individuals annually [(6)].

The incidence peaks in the fifth and sixth decades of life, and it is five times higher in males than in females. Our results coincided with the previous studies in terms of age and sex predominance [(7)].

The constant symptom was dysphonia followed by stridor with or without dyspnea. Dysphagia and aerodigestive obstruction developed only in advanced cases (V3). The highest proportion of patients seeking medical consultation were presented in stage V2 (16/24) when stridor and dyspnea started to bother them. Progress from V1 to V3 has an intimate relation to clinical presentation and is an alarm to facing acute aerodigestive obstruction if neglected and consequently, early surgical interference is recommended.

Even less common is infection within a laryngocele that results in pus buildup and the development of a laryngophone [(8)].

Vasileiadis *et al.* [(9)] reported 39 cases, only four developed acute airway obstruction, of whom one had a laryngopyocele.

Our series showed three critical cases in stage V3 with acute aerodigestive obstruction. One of them died during induction of anesthesia and the other two cases were operated successfully.

Byard and Gilbert [(10)] discovered that the release of pus into the airway, which can result in aspiration, jugular vein thrombosis, or mediastinal abscess, increased the risk of death from laryngopyocele. Three deaths were documented as a result of this condition. The literature has only reported 64 cases of laryngopyocele.

Air content was pathognomonic to diagnose laryngocele in radiological studies. Imaging also assessed the nature of the content as well as the degree of extension (compartmental involvement) that coincided with endoscopic findings and clinical presentation. **Blickman *et al.*** [(11)]**.** Air content was seen in 17 cases, where three of them had air only, 14 cases had both air and fluid, and fluid alone was present in 7 cases.

We suppose that the nature of the content can reflect the degree of communication between larynx and laryngocele as well as its pathogenesis and staging. Wide patulous communication between larynx and saccule allows free passage of air and dilatation of saccule containing only air that may not progress to stage V3. While narrow communication between larynx and saccule can result in obstruction, fluid retention and air trapping with more rapid increase in size and laryngocele may reach stage V3 with subsequent supraglottic collapse and more distress. Consequently, we can conclude that the opening of the saccule may be a risk factor for pathogenesis of laryngoceles.

We found statistical significance correlating between presence of air with both symptom severity and staging. We encountered a case with stage V3 containing air that was serious leading to acute aerodigestive obstruction, hence we sometimes call it tension laryngocele that may create difficult intubation.

Reviewing the literature revealed multiple staging systems for benign conditions of the larynx **Heyes *et al.*** [12]**.** of whom **DeSanto *et al.*** [13] classification is the most commonly utilized, with cysts classified as either ductal (mucosal retention cysts) or saccular (submucosal). Two other subtypes of saccular cysts are lateral and anterior: The laryngeal airway has tiny anterior cysts that are situated at the saccular orifice. More often than not, lateral cysts cause distortions in supraglottic anatomy and extend into the laryngeal vestibule.

Large saccules, laryngoceles, saccular cysts, and saccular mucoceles, according to **DeSanto** [14], reflect a spectrum of developmental stages.

Our study gave a particular concern to combined laryngoceles and reviewed our tertiary hospital experience with such rare lesions within the last 30 years. There was an intimate relationship between the three studied variables (physical, endoscopic, and imaging) validating our novel staging. We believe that this simple staging system is of much practical, applicable, and feasible benefit for otolaryngologists.

Our study showed that there was no statistically significant difference in the presence of dysphonia, stridor (P = 0.076), aerodigestive obstruction (P = 0.530), and dysphagia (P = 0.530) between the cases with and without air content. However, there was statistically significantly higher incidence of dyspnea (P = 0.017) in cases with air content. There was also no statistically significant difference in the endoscopic grading (P = 0.085) according to the presence or absence of air content within the laryngoceles.

Similarly, A study examined the current evidence on the pathophysiology, impact and management of UVFP and swallowing. They showed that pre-and post-operative dysphagia score was non-significant (p = 0.05) **Ha *et al.*** [15] .

CONCLUSION

Clinical staging of combined laryngoceles as (V1, V2, V3) is a novel staging and is away from bias. It depends on the internal component extension to ventricle (V1), vestibule (V2), or vallecula (V3). This staging represents a simple, feasible, and reliable method and is validated both clinically, endoscopically, and radiologically.

RECOMMENDATIONS

From our study we recommend that: Performing larger scale multicenter studies, avoid risk factors for development of laryngocele, and regular check and examination for cases with symptoms suggestive of laryngocele.

DECLARATIONS

- **Funding:** No fund
- **Availability of data and material:** Available
- **Conflicts of interest:** No conflicts of interest.
- **Competing interests:** None

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