



Acute supraglottic laryngitis complicated by vocal fold immobility: prognosis and management

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Received: 1 April 2019 / Accepted: 5 June 2019 / Published online: 13 June 2019
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Abstract

Purpose Acute supraglottic laryngitis (ASL) is manifested by supraglottic inflammation that has the potential for rapid and fatal airway obstruction. Complete/incomplete vocal fold immobility (VFIm) in the setting of ASL may contribute to airway obstruction. The rate of VFIm complicating ASL is not known, and it is not clear whether its occurrence alters the course and the management of ASL, particularly the need to secure the airway (by endotracheal intubation/tracheostomy). This study seeks to describe the natural history of VFIm associated with ASL (ASLIm) and to determine the added effect of VFIm on ASL severity, management, and the need for intervention to secure the airway.

Methods This is a retrospective cohort study in a tertiary referral center. The medical records of all patients hospitalized due to ASL between January 2007 and December 2016 were reviewed.

Results Of the 214 patients admitted due to ASL, VFIm was identified in 9 (4.2%). The VFIm resolved within 1–90 days in all 8 patients with available follow-up of 1–3 months. One patient required endotracheal intubation. The 9 ASLIm patients had significantly higher rates of hoarseness and a history of diabetes mellitus. There was no group difference in the need to secure the airway.

Conclusions VFIm is an apparently uncommon finding among patients with ASL. It appears to be usually transient, short-lasting, and full recovery can be expected. VFIm did not alter the ASL course, nor did it put our patients at increased risk for the need for intervention to secure the airway.

Level of evidence 4.

Keywords Acute supraglottic laryngitis · Larynx · Vocal fold immobility

Introduction

Acute supraglottic laryngitis (ASL) is manifested by supraglottic structures inflammation, typically from an infectious etiology, that has the potential for rapid and fatal airway obstruction [1–5].

Trends of acute airway intervention in the setting of ASL have changed toward a more conservative approach [6–11]. Various staging systems have been suggested to evaluate the severity of airway compromise and the need for surgical

intervention (either via endotracheal intubation or tracheotomy), based on edema of different laryngeal structures [12, 13]. None of the existing staging systems refers to vocal fold (VF) mobility as part of disease presentation. Furthermore, our literature search failed to yield any publications on the prevalence of complete/incomplete vocal fold immobility (VFIm) in ASL.

The aims of this study were: to find the rate of VFIm associated with ASL; to describe the natural history of VFIm associated with ASL; to study its impact on disease prognosis and management, and specifically to determine whether the presence of VFIm in the setting of ASL would entail a more severe disease course and make the patient more susceptible to upper airway obstruction and need for surgical airway establishment.

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Materials and methods

Study design

This is a retrospective cohort study conducted at the Tel-Aviv Sourasky Medical Center, a tertiary referral center after receiving an institutional review board approval (0653-18-TLV). The medical records of all adult (age ≥ 18 years) patients diagnosed with their first episode of ASL (International Classification of Diseases, Ninth Revision, Clinical Modification codes 464.30, 464.31, 464.50, 464.51) between January 2007 and December 2016 were retrieved and reviewed. Patients with a history of laryngeal malignancy, foreign body ingestion, thermal/chemical trauma, angioedema, granulomatosis with polyangiitis disease, < 3 months post-surgery of the pharynx, prior known VF dysfunction, or whose records were incomplete were excluded.

Data on demographics (age, gender), medical history, symptoms (including hoarse voice by patient report on presentation), pre-hospitalization antibiotic treatment, vital signs at presentation, head and neck examination findings (including flexible fiberoptic laryngoscopy), white blood count and c-reactive protein (CRP) levels, imaging findings, hospitalization duration, medical treatment, surgical interventions to secure the upper airway, and post-hospitalization in-clinic follow-up were collected and analyzed.

Patients were examined during hospitalization by an otolaryngologist who established the diagnosis of ASL and recorded VF mobility. All patients were followed daily during hospitalization by flexible fiberoptic laryngoscopy until resolution of ASL. Patients whose VFIm did not resolve during hospitalization were further followed at the outpatients' clinic.

For statistical purposes, patients with both incomplete and complete immobility were regarded as a single group—ASL with VFIm group (ASLIm). Patients were divided into 2 groups: those with ASL and VFIm (ASLIm), and those with ASL and intact VF mobility (ASLIntM).

Statistical analysis

Categorical variables were described by frequency and percentage. Continuous variables were evaluated for normal distribution using histograms and Q–Q plots and expressed as mean and standard deviation or median and interquartile range. Categorical variables were compared between categories using the Chi-square test or Fisher's exact test, and continuous variables were compared using

the *T* test or the Mann–Whitney test. All statistical tests were two-tailed, and a *P* value < 0.05 was considered as statistically significant. All statistical analyses were performed using SPSS (IBM Corp. Released 2014. IBM SPSS Statistics for Windows, Version 22.0. Armonk, NY: IBM Corp.).

Results

Two-hundred and thirty-three admissions for isolated ASL were identified in the study period. Twelve patients had more than one admission (nine had two, one had three, one had four, and one had six). In total, the first admission of 214 patients (120 males, 94 females, mean age 48.4 years) was included in the analysis. Nine (4.2%, 6 males, 3 females, average age 47.5 years) were diagnosed with ASLIm, and 205 (95.8%, 114 males, 91 females, average age 48.4 years) were diagnosed with ASLIntM. All nine ASLIm patients presented with unilateral VFIm (five right VFIm, four left VFIm, four with VF complete immobility, and five with VF incomplete immobility). There were no significant group differences in gender, age, and habits between the two groups. Patients in the ASLIm group had a significantly higher rate of history of diabetes mellitus (44.4% vs 11.7%, $P = 0.02$) (Table 1).

On presentation, the ASLIm patients reported a significantly higher rate of hoarseness compared to the patients with ASLIntM (20 vs 3, $P \leq 0.049$) and a trend of higher white blood cell count was observed ($16.710 \times 10^3/\mu\text{L}$ vs $12.510 \times 10^3/\mu\text{L}$, $P \leq 0.063$) (Table 2). There were no other group differences in symptoms, physical findings or imaging study findings between patients with and without VFIm.

Patients were admitted to the otolaryngology department or to the intensive care unit (ICU) for airway supervision (at the discretion of the admitting physician) and started on intravenous antibiotics, intravenous steroids, proton pump inhibitors (PPIs), steroid inhalations, or their combinations (Table 3). There were no significant differences between the two groups in medical treatment, in the need for airway supervision in the ICU and in hospitalization duration. Patients with VFIm did not have higher rates of interventions to secure the airway either by intubation or by tracheostomy as compared to patients with intact VF mobility (1 vs 2 and 0 vs 1, respectively, $P = \text{NS}$; Fig. 1). One patient in the ASLIm group who presented with an infected vallecular cyst underwent direct laryngoscopy and cyst resection at the same admission, and 3 patients in the ASLIntM group underwent direct laryngoscopy and vallecular/epiglottic cyst resection (one at the same admission and two on another elective admission), and none of them needed surgical airway intervention.

Table 1 Demographics and patient characteristics

	<i>ASLIm</i> (n=9) n (%)	<i>ASLIntM</i> (n=205) n (%)	<i>P</i> value
Age, year (standard deviation)	47.6 (18.3)	48.4 (15.8)	0.876
Gender (male)	6 (66.7)	114 (55.6)	0.734
Comorbidities			
Smoking	1 (11.1)	19 (9.3)	0.594
Cardiovascular disease	4 (44.4%)	61 (29.8)	0.459
CVA	1 (11.1)	4 (2)	0.195
GERD	2 (22.2)	14 (6.8)	0.139
Diabetes mellitus	4 (44.4)	24 (11.7)	0.019
Immunosuppression	0 (0)	4 (2)	>0.999
s/p neck irradiation	0 (0)	2 (1)	>0.999
s/p neck dissection	1 (11.1)	1 (0.5)	0.083
s/p transplantation	0 (0)	0 (0)	n/a
Other comorbidities	2 (22.2)	57 (27.8)	>0.999
No comorbidities	3 (33.3)	105 (51.2)	0.33
Prolonged systemic steroid treatment	0 (0)	3 (1.5)	>0.999
Prolonged inhaled steroid treatment	0 (0)	2 (1)	>0.999
Prolonged PPI treatment	2 (22.2)	15 (7.3)	0.154

Italic indicates significance

ASLIm acute supraglottitis with vocal fold complete/incomplete immobility, *ASLIntM* acute supraglottitis with intact vocal fold mobility, *CVA* cerebral vascular accident, *GERD* gastroesophageal reflux disease, *PPI* proton pump inhibitors, *n/a* non-applicable

VFIm resolved within the hospitalization period in six of the nine patients (1–7 days from initial diagnosis). Two of the nine *ASLIm* patients had complete resolution of the VFIm up to 3 months following admission. One patient had VFIm recorded 1-month post discharge, but was then lost to follow-up.

Discussion

VFIm appears to be an uncommon finding in the setting of ASL. The incidence of VFIm in this study was 4.2%. Since VFIm completely resolved in all eight patients with adequate follow-up data, we assume that these patients developed VFIm as a result of ASL rather than had a preliminary undiagnosed VFIm. To the best of our knowledge, this is the first publication regarding VFIm in ASL patients. The exact pathophysiology leading to VFIm in ASL patients is beyond the scope of this study. Possible mechanisms leading to VFIm in ASL may include: mass effect of the edematous epiglottis, false VF or arytenoids on the VFs leading to VFIm, or involvement of the recurrent laryngeal nerve in the inflammatory process leading to VFIm. Since we demonstrated that VFIm in ASL is usually short-lasting and reversible, we assume that it is sound to conclude that VFIm, at least in most cases reported herein, was secondary to a mass effect of the inflamed supralarynx on the VF and not due to

neural dysfunction caused by inflammation of the recurrent laryngeal nerve itself. However, we did not perform objective neurophysiological studies such as EMG to confirm our theory, and therefore refrain from further discussing the pathophysiological realm leading to VFIm.

In their recent series on ASL, Shapira Galitz et al. reported that patients with dyspnea and stridor, edema of the epiglottis, elevated CRP, hyperglycemia, and a history of recurrent episodes of ASL were at a higher risk of airway compromise [14]. We observed that the patients in the *ASLIm* group had higher rates of history of diabetes mellitus, perhaps signifying a more severe inflammatory state of the supraglottis in patients with diabetes mellitus; however, CRP levels were comparable between the *ASLIm* and *ASLIntM* groups. Furthermore, the *ASLIm* patients were not in an increased risk for airway compromise or need of surgical intervention. We also observed that patients in the *ASLIm* group also reported a higher rate of hoarseness and had a trend of higher WBC levels at presentation. These parameters (history of diabetes mellitus, hoarseness and elevated WBC count) might be used to determine patients at risk of VFIm. With that being said, we had very low rates of airway intervention in this study; so the fact that diabetes mellitus was not a predictor for airway intervention might be a selection bias.

While few studies addressed the question of the accurate timing of surgical airway intervention in ASL, none

Table 2 Disease presentation

	ASLIm (<i>n</i> =9) <i>n</i> (%)	ASLIntM (<i>n</i> =205) <i>n</i> (%)	<i>P</i> value
Symptoms			
Duration, days (IQR)	3 (2.5, 4)	3 (2, 4)	0.342
Sore throat	8 (88.9)	198 (96.6)	0.295
Dysphagia	6 (66.7)	179 (87.3)	0.107
Hoarseness	44 (44.4)	33 (16.1)	0.05
Drooling	0 (0)	15 (7.3)	> 0.999
Dyspnea	2 (22.2)	51 (24.9)	> 0.999
Globus sensation	0 (0)	9 (4.4)	> 0.999
Cough	0 (0)	5 (2.4)	> 0.999
Other symptoms (cyanosis, ear pain, blood in saliva, urinary symptoms)	0 (0)	13 (6.3)	> 0.999
Physical examination			
Fever > 37.8 °C	6 (75)	96 (47.1)	0.158
Stridor	0 (0)	2 (1)	> 0.999
Tripod position	0 (0)	2 (1)	> 0.999
Edema of the arytenoids/aryepiglottic fold/epiglottis/FVC	9 (100)	196 (95.6)	> 0.999
Epiglottic/arytenoids exudate	1 (11.1)	21 (10.2)	> 0.999
Partially visible glottis/moderate incompetent airway	0 (0)	13 (6.3)	> 0.999
Nonvisible glottis/severe incompetent airway	1 (11.1)	4 (2)	0.195
Subglottic edema	0 (0)	1 (0.5)	> 0.999
Pyramidal sinus edema	0 (0)	7 (3.4)	> 0.999
Saliva stasis	1 (11.1)	28 (13.7)	> 0.999
Vallecular cyst	1 (11.1)	16 (7.8)	0.629
Epiglottic cyst	0 (0)	7 (3.4)	> 0.999
Trismus	0 (0)	3 (1.5)	> 0.999
Pharyngitis/tonsillitis	1 (11.1)	62 (30.2)	0.288
Lingual tonsillitis	0 (0)	14 (6.8)	> 0.999
Peritonsillar abscess	0 (0)	3 (1.5)	> 0.999
Aphthous epiglottis	0 (0)	9 (4.4)	> 0.999
Vocal fold polyp	0 (0)	2 (1)	> 0.999
Neck mass/tenderness/neck crepitation	4 (44.1)	70 (34.1)	0.5
Blood test results			
WBC (10e3/ μ L) (IQR)	16.7 (12.7, 17.9)	12.5 (10, 15.75)	0.063
Neutrophils % (IQR)	80.4 (72.3, 86.35)	78 (70.3, 83.7)	0.445
Lymphocytes % (IQR)	13.1 (6.6, 18.4)	12.9 (8.6, 18.8)	0.458
CRP (mg/L) (IQR)	56 (42.2, 133.3)	50.5 (13.5, 119.3)	0.549

ASLIm acute supraglottitis with vocal fold complete/incomplete immobility, ASLIntM acute supraglottitis with intact vocal fold mobility, FVC false vocal cords, IQR interquartile range, WBC white blood count, CRP C-reactive protein

of them focused on the role of VFIm and its natural history. Despite our tendency to regard VFIm as an indicator of airway compromise, the present study does not support this notion. Further more, the results of this study demonstrates once again the relatively low rates of airway intervention needed in patients with ASL, even when presenting with VFIm. Moreover, the presence of VFIm was transient, resolved spontaneously, and did not prolong hospitalization.

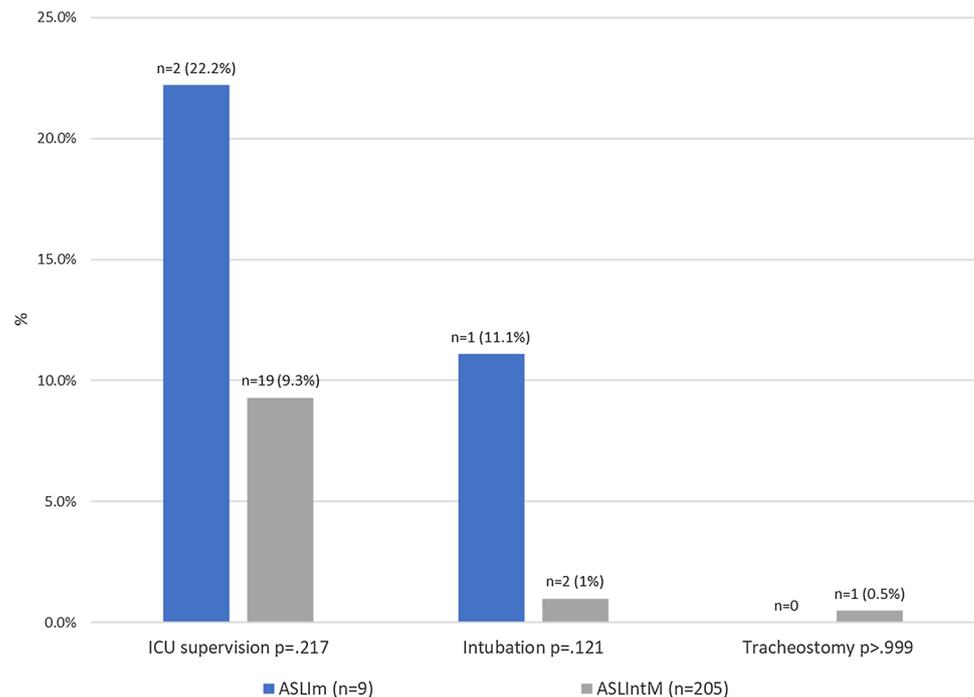
Limitations of our study include the inherent bias of a retrospective analysis, and we might have underestimated the incidence of ASLIm. Another limitation to mention is that the patients were not followed with short interval laryngeal examinations after discharge, so the exact duration of VFIm is inaccurate. As mentioned, since patients presenting with VFIm did not undergo an electromyography, we cannot establish the pathophysiology of the

Table 3 Medical treatment

	ASLIm (<i>n</i> =9) <i>n</i> (%)	ASLIntM (<i>n</i> =205) <i>n</i> (%)	<i>P</i> value
Treatment			
Pre-admission oral antibiotics treatment	3 (33.3)	65 (31.9)	> 0.999
Systemic antibiotic treatment	7 (77.8)	204 (99.5)	0.548
Systemic steroid treatment	7 (77.8)	197 (96.1)	0.632
PPI treatment	5 (55.5)	149 (72.7)	0.183
Inhaled steroids	4 (44.4)	127 (62)	0.314
Hospitalization, days (IQR)	5 (1.5, 7)	5 (3,6)	>0.999

ASLIm acute supraglottitis with vocal fold complete/incomplete immobility, *ASLIntM* acute supraglottitis with intact vocal fold mobility, *PPI* proton pump inhibitors, *IQR* interquartile range

Fig. 1 Airway Intervention. *ASLIm* acute supraglottitis with vocal fold complete/incomplete immobility, *ASLIntM* acute supraglottitis with intact vocal fold mobility, *ICU* intensive care unit



disease. With that being said, this is the first report of VFIm as a complication of ASL and although uncommon, is not negligible.

Conclusions

VFIm occurs in 4.2% of ASL cases. VFIm is part of the inflammatory manifestations of ASL, is usually transient and full recovery may be expected. VFIm does not portend worse outcomes in patients with ASL and does not dictate the need for airway intervention. The findings of this study are important in confirming that no different treatment strategy is required for ASL patients presenting with VFIm.

Acknowledgements Esther Eshkol, MA, the institutional medical and scientific copyeditor (Tel-Aviv Sourasky Medical Center) is thanked for editorial assistance. Tomer Ziv, PhD, is thanked for statistical analysis.

Funding The authors have no funding, financial relationships, or conflicts of interest to disclose.

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