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SABER-SHEATH TRACHEA – A RARE OR UNDERDIAGNOSED MORPHOLOGICAL VARIETY OF THE TRACHEA

TRAHEJA OBLIKA KORICA SABLJE – REDAK ILI NEDOVOLJNO DIJAGNOSTIKOVAN MORFOLOŠKI VARIJETET TRAHEJE

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Summary

Introduction. The saber-sheath trachea is characterized by widened anteroposterior and narrowed laterolateral tracheal diameter. It is usually found in patients with chronic obstructive pulmonary disease. The aim of this study was to determine the incidence of this disorder, as well as to gain insight into the basic socio-demographic characteristics of patients and the clinical features of this tracheal deformity. **Material and Methods.** Endoscopic findings of patients undergoing bronchoscopy at the Institute for Pulmonary Diseases of Vojvodina in the period January 1, 2013 – January 1, 2021 were analyzed. Individual socio-demographic data of patients with the diagnosis of saber-sheath trachea were collected and clinical parameters were analyzed. **Results.** The analysis of 15.381 bronchoscopic findings showed an incidence of 0.56%, most often in the elderly, predominantly in men, and those who were active or former smokers. The most common comorbidities were chronic obstructive pulmonary disease, cardiovascular diseases, and diabetes. In all cases, this deformity was found as an incidental finding during bronchoscopy that was mostly done for the diagnosis of primary carcinoma of the bronchus. Accordingly, the most common pre-bronchoscopic symptoms were shortness of breath, cough, and chest pain. No significant narrowing of the tracheal lumen was found in any of the patients, nor did this anatomical variety of the trachea affect the length of survival. **Conclusion.** Although this is a rare disorder, its recognition can guide the clinician to apply additional procedures in order to establish a diagnosis of chronic obstructive pulmonary disease, but also significantly contribute to avoiding potential complications in need of endotracheal intubation.

Key words: Trachea; Cartilage; Congenital Abnormalities; Pulmonary Disease, Chronic Obstructive; Bronchoscopy; Intubation, Intratracheal; Sociodemographic Factors; Signs and Symptoms

Introduction

Saber-sheath trachea is a fixed deformity characterized by an acquired widened anteroposterior and a substantially narrowed laterolateral tracheal

Sažetak

Uvod. Traheju oblika korica sablje karakteriše proširenje anteroposteriornog prečnika, uz istovremeno suženje laterolateralnog prečnika dušnika. Najčešće se javlja kod obolelih od hronične opstruktivne bolesti pluća. Cilj ovog istraživanja bio je da se utvrdi učestalost pomenutog poremećaja, kao i da se stekne uvid u osnovne sociodemografske karakteristike obolelih, kao i kliničke karakteristike ovog deformiteta traheje. **Materijal i metode.** Analizirani su endoskopski nalazi bronhoskopiranih bolesnika u Institutu za plućne bolesti Vojvodine u periodu 1. 1. 2013–1. 1. 2021. godine. Za bolesnike kod kojih je potvrđeno postojanje *saber-sheath* traheje, prikupljeni su pojedini sociodemografski podaci i analizirane kliničke karakteristike obolelih. **Rezultati.** Analizom 15.381 bronhoskopskog nalaza, utvrđeno je da je učestalost traheje oblika korica sablje u ispitivanoj populaciji bila 0,56%; najčešće se javlja kod osoba starijeg životnog doba, i to predominantno kod muškaraca i onih koji su aktivni ili bivši pušači. Najčešće pridružene bolesti bile su hronična opstruktivna bolest pluća, kardiovaskularne i šećerna bolest. U svim slučajevima, deformitet traheje je uočen kao slučajaj nalaz prilikom bronhoskopije, indikovane najčešće zbog primarnog karcinoma bronha. U skladu s tim, najčešći prebronhoskopski simptomi bili su otežano disanje, kašalj i bolovi u grudima. Istovremeno, ni kod jednog bolesnika nije utvrđeno značajnije sužavanje lumena dušnika, niti je ovaj anatomske varijetet traheje uticao na dužinu preživljavanja. **Zaključak.** Iako se radi o retkom poremećaju, njegovo prepoznavanje može usmeriti kliničara na primenu dodatnih procedura u cilju postavljanja dijagnoze hronične opstruktivne bolesti pluća, ali i značajno doprineti izbegavanju potencijalnih komplikacija u slučaju potrebe za izvođenjem endotrahealne intubacije.

Glavne reči: traheja; hrskavica; kongenitalne anomalije; hronična opstruktivna bolest pluća; bronhoskopija; endotrahealna intubacija; sociodemografski faktori; znaci i simptomi

diameter [1]. According to previous research, tracheal narrowing occurs only in the intrathoracic airway, while the extrathoracic part of the trachea remains normal [2]. Unlike tracheomalacia, tracheal rings are thicker and the trachea is not collapsible.

Abbreviations

COPD – chronic obstructive pulmonary disease
 CT – computed tomography
 IPDV – Institute for Pulmonary Diseases of Vojvodina

The exact cause of this anatomical variation of trachea is unknown. It is most frequent in patients with chronic obstructive pulmonary disease (COPD) [3]. Studies show that diagnosing COPD in patients with saber-sheath trachea has a high specificity (92.9%), but a low sensitivity (39.1%) [4]. The diagnosis of saber-sheath trachea is most commonly made as a coincidental finding on chest X-ray or chest computed tomography (CT) [5], or as an incidental finding during bronchoscopy.

In the vast majority of cases, this condition does not result in a significant narrowing of the tracheal lumen and therefore it does not require any specific treatment. However, several cases of difficult endotracheal intubation have been reported in the literature [6, 7]. Therefore, it is important to consider this disorder in the preoperative patient preparation for surgeries requiring general anesthesia.

The aim of the present study was to determine the incidence of saber-sheath trachea based on descriptions of endoscopic findings in patients undergoing bronchoscopy for various reasons, as well as to gain insight into basic socio-demographic characteristics of patients and clinical manifestations of the disorder. We also wanted to provide additional information about this rare, but important condition.

Material and Methods

In this retrospective observational study, 15,381 bronchoscopic findings of patients who underwent bronchoscopy at the Institute for Pulmonary Diseases of Vojvodina (IPDV), Sremska Kamenica, Serbia, from January 1, 2013 to January 1, 2021, were evaluated. The final analysis included findings of 82 patients who met the inclusion criterion of the study: diagnosis of saber-sheath trachea (trachea with a narrowed laterolateral and increased anteroposterior diameter) confirmed by bronchoscopy. Even though all patients who underwent bronchoscopy had previously performed chest X-ray and/or CT scan, radiological confirmation of saber-sheath trachea was not a mandatory criterion for inclusion in this study. Data on gender, age, smoking habits, comorbidities, past surgeries requiring general anesthesia, symptoms reported by patients prior to bronchoscopy, lung function parameters, and disease outcome after a period of follow-up, were extracted from the patients' medical records.

The informed consent of the patients included in the study was not necessary, because the study was based on retrospective analysis of medical data. However, all subjects signed an informed consent for bronchoscopy. Statistical data processing was performed using the software packages IBM Statistical Package for the Social Sciences Statistics v. 23 and Microsoft Office 2013. The central tendency

of numerical features is shown by the arithmetic mean, and the minimum, maximum, and range of values. Attributive features are shown using absolute and relative frequencies.

Results

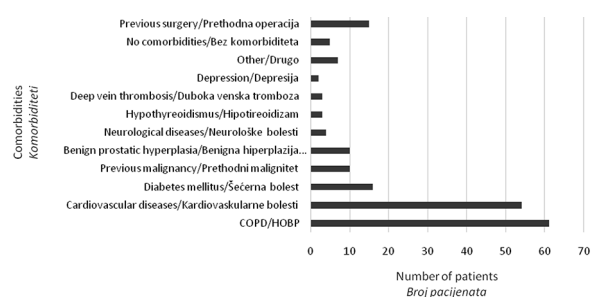
During the eight-year study period, 15,381 bronchoscopies were performed at the IPDV including 14,545 patients undergoing bronchoscopy (some patients underwent bronchoscopy two or more times). Saber-sheath trachea was confirmed by endoscopy in 82 patients, accounting for 0.56% of the overall sample.

Males dominated in our sample (78 respondents, i.e. 95.1%). The average age of patients was 68.3 years, ranging from 53 to 87 years, indicating that this condition was found primarily in elderly.

Every patient in this study had previously smoked cigarettes. A total of 52 patients (63.4%) were current smokers, consuming 65.4 packs per year (ranging from 30 to 140) on average, while 30 patients (36.6%) had previously smoked 50.8 packs per year (ranging from 30 to 100) on average, but were presently non-smokers. **Table 1** shows the main socio-demographic characteristics of our sample.

The COPD was the most common comorbidity in the sample, having been previously identified in 49 patients (59.7%). Spirometry results were available for another 12 of the remaining 33 individuals who had not been previously diagnosed with COPD, and all 12 patients had lung volumes and capacities, as well as bronchodilator reversibility test values that indicated COPD. Thus, COPD was found to be practically present in 61 patients in the study population (74.4%). However, it must be noted that 21 patients (25.6%) were not diagnosed with COPD prior to bronchoscopy and had not undergone spirometry, which is the reason why this part of the research group could not be assessed for the presence of COPD. Of the other comorbidities, the most common were cardiovascular diseases – arteriosclerosis, cardiomyopathy, and ischemic heart disease (54 patients, i.e. 65.8%), followed by diabetes mellitus (16 patients, i.e. 19.5%), history of previously treated malignant diseases, and benign prostatic hyperplasia (10 patients in both cases, i.e. 12.2%). Only five participants (6%) were found to have no comorbidities. A total of 15 patients (18.3%) had previously undergone surgery under general anesthesia, but no data on possible difficulties during endotracheal intubation were found in any of the subjects' medical records. **Graph 1** shows findings on patients' comorbidities.

The most prevalent complaints prior to bronchoscopy were (in descending order): shortness of breath (54 patients, i.e. 65.8%), cough (52 patients, i.e. 63.4%), chest pain (22 patients, i.e. 26.8%), expectoration of blood (10 patients, i.e. 12.2%), fever (8 patients, i.e. 8.5%), while only 3 patients had no symptoms (3.6%). The majority of patients underwent bronchoscopy due to lung carcinoma (66 patients, i.e. 80.5%), whereas 11 patients (13.4%) had



Graph 1. The incidence of comorbidities

Grafikon 1. Učestalost komorbiditeta pacijenata

inflammatory processes in the lungs. Bronchoscopy did not reveal the cause of alterations in the lung parenchyma in the remaining 5 patients (6.1%).

Disease outcome data were available for a total of 65 patients. The average follow-up period after bronchoscopy was 10.7 months (ranging from 1 to 60), and the follow-up shows that most patients with bronchial cancer died with an average survival time of 8.2 months (38 patients; data were unavailable for the remaining 28 patients). In those with inflammatory changes in the lung parenchyma, the symptoms completely resolved after the application of an adequate therapeutic protocol, and patients were asymptomatic during the follow-up period. Considering this, it can be concluded that this anatomical variation did not result in a substantial degree of tracheal constriction and subsequent clinically significant problems.

Discussion

Our research shows that the prevalence of saber-sheath trachea in the general population is low (0.56%), that it mostly affects the elderly, predominantly men, and those who are active or former smokers. The most prevalent comorbidity is COPD, followed by cardiovascular diseases and diabetes mellitus. In most cases, saber-sheath trachea is observed as an accidental finding during bronchoscopy, which is performed most usually due to bronchial cancer. Accordingly, the most common symptoms prior to bronchoscopy are shortness of breath, cough, and chest pain. Patients with this tracheal deformity present with no significant narrowing of

the tracheal lumen, nor does this anatomical variety affect the survival time of these patients.

The data in the literature vary greatly when it comes to the prevalence of this acquired anatomical variety of the trachea. However, everyday clinical practice shows that this variety often remains unrecognized, which indicates that its prevalence is probably significantly higher than reported in the literature. Considering the general population, one study reported the prevalence of saber-sheath trachea of 5% [8], while in our study it was significantly lower – 0.56%. When it comes to studies including only patients with COPD, Green et al. [9] found that 95% of patients with saber-sheath trachea also had COPD, while in Gupta's study [10] the prevalence of this anatomical variety of the trachea in patients with COPD was 35%. Previous research indicated that the tracheal index was negatively related to the duration of COPD, length of cigarette smoking, dyspnea scale scores, and spirometry scores [11–13]. Therefore, it is possible that this tracheal variety occurs in the early stages of COPD, as well as that the laterolateral diameter of the trachea narrows more and more over time. It is possible that this disorder is a consequence of changes in the value of intrathoracic pressure and air trapped in the lungs, with hitherto unrelated concomitant action of associated factors, such as environmental exposure or genetics [1, 12]. Other mechanisms are likely to play a role in the development of this disorder, since not all patients with COPD have saber-sheath trachea, nor all patients with saber-sheath trachea have COPD. It has been hypothesized that this tracheal disorder may also occur due to degenerative changes and ossification of tracheal rings, which may be caused by chronic extensive cough [7].

Several research, including our own, found that this disorder is more common in men [1, 9] and those over the age of 50 [6], the latter supporting the thesis of acquisition of the disorder. However, other investigations have found no variations in prevalence depending on gender or age [12].

The diagnosis of saber-sheath trachea is usually made based on the results of radiological imaging methods (chest radiography or CT scan) or endoscopy with bronchoscopy. The tracheal index, which shows the ratio between the transverse and anteroposterior diameters of the trachea, can be assessed on chest X-ray and is usually measured 1 cm above the aor-

Table 1. Socio-demographic characteristics of the analyzed sample

Tabela 1. Socio-demografske karakteristike analiziranog uzorka

	No/Br	%
Prevalence/Prevalencija	82	0.56
Gender/Pol		
Male/Muški	78	95.1
Female/Ženski	4	4.9
Smoking status/Pušački status		
Smokers/Pušači	52	63.4
Ex-smokers/Bivši pušači	30	36.6
Non-smokers/Nepušači	0	0

tic arch [13]. In this tracheal anatomical variant, the tracheal index is smaller than 0.67 (2 : 3) [12]. Chest CT scans provide substantially more reliable information because the trachea's rotation does not impact the thoracic index measurement, which is not the case with chest X-ray. Despite the fact that saber-sheath trachea is usually easily detected on a chest X-ray, literature data indicate that this condition is frequently overlooked in everyday clinical practice [1]. The diagnosis is made most commonly as an accidental endoscopic finding during bronchoscopy performed for various reasons. During bronchoscopy, the degree of tracheal collapse during expiration can be also assessed [15].

When diagnosing this condition, it is important to rule out other disorders where the trachea can take on the appearance of the saber-sheath. These include mediastinal mass (malignant or benign tumor, metastasis), iatrogenic or post-inflammatory tracheal stenosis, ankylosing spondylitis, and granulomatous disorders such as amyloidosis, sarcoidosis, and granulomatosis with polyangiitis [16].

Shortness of breath, cough, chest pain, hemoptysis, and fever were among the most common symptoms experienced by the patients in our study prior to bronchoscopy. All of these symptoms, however, could be related to the underlying disease (bronchial cancer in the majority of cases, or less frequently, pulmonary inflammation), rather than saber-sheath trachea. It is considered that this anatomical tracheal deformity is usually not associated with any symptoms. Investigations over the last century have shown that it is necessary to reduce the tracheal lumen by 70% to significantly reduce airflow through it, and thus to cause clinically significant symptoms [17]. However, as a result of this malformation, endotracheal intubation can be challenging at times. In such cases, it is recommended to use an endotracheal tube with a smaller diameter than expected [6], or flexible bronchoscopy with the tip of the endotracheal tube introduced to the narrowest part of the trachea, because further advancement of the endotracheal tube may cause tracheal mucosa injuries [12]. Although saber-sheath trachea is normally smaller in diameter than the normal trachea, saber-sheath trachea lumen enlargement has been reported in the literature. Because the circle-shaped tube balloon cannot approach the sagittal elongated tracheal walls in such cases, air leakage during endotracheal insertion is possible [7]. In such circumstances, a laryngeal mask or other su-

praglottic agents provide an alternate option for providing appropriate mechanical ventilation [5, 18].

There is no recommended therapeutic protocol for the treatment of patients with saber-sheath trachea [19]. External tracheal fixation with artificial materials during thoracotomy, as well as suturing an absorbable mesh on the anterior tracheal wall during anterothoracic tracheoplasty, have both been described in the literature, and led to the reduction of airflow obstruction [20]. Temporary stents can also be used to expand the tracheal lumen, though this comes with the risk of injury to the airway mucosa due to stent migration and/or fracture. However, such procedures have yet to be standardized, and the literature only contains descriptions of specific cases [21]. The advantage of this study is that the cohort of patients with saber-sheath trachea from the territory of the Republic of Serbia was analyzed for the first time and that the obtained data increase the total amount of knowledge about this rare and understudied disorder. The limitation of the study is that for a large number of patients data were not available during the follow-up period (survival time, above all), since bronchoscopy was performed on an outpatient basis in the IPDV and patients were further treated in regional health facilities. In addition, in the last ten years a limited number of literature sources describing any aspect of the diagnosis and/or treatment of this malformation is available, which significantly limits the interpretation of the obtained results.

Conclusion

In the general population, saber-sheath trachea is a rather uncommon acquired morphological tracheal abnormality, but it is a relatively common radiological and endoscopic finding in chronic obstructive pulmonary disease patients. As a result, in those who have not previously been diagnosed with chronic obstructive pulmonary disease, it may be considered as a major radiological sign suggesting the necessity for additional diagnostic procedures to diagnose chronic obstructive pulmonary disease. Although narrowing of the tracheal lumen often does not result in significant airflow obstruction or clinical manifestations, physicians of various specialties (primarily pulmonologists, radiologists, and anesthesiologists) should consider this anatomical malformation of the trachea during the preoperative assessment of patients who are planned for general anesthesia or mechanical ventilation for any indication, since this disorder may cause significant difficulties during endotracheal intubation.

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