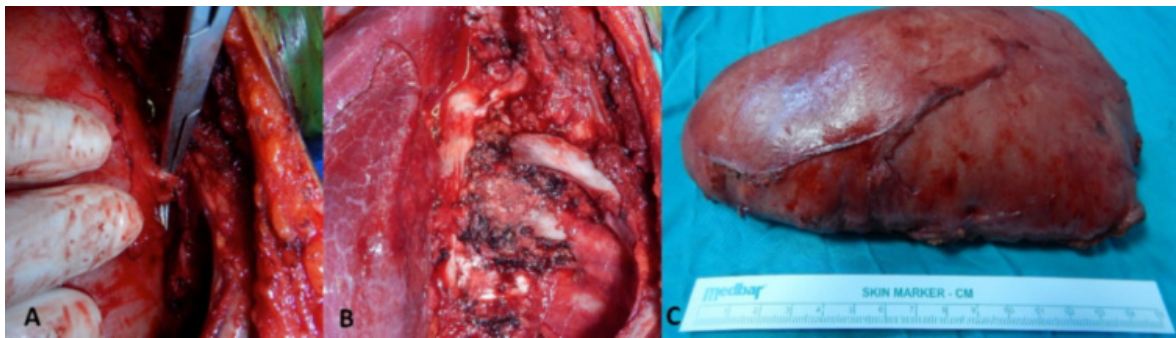


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Original Article

Minimally invasive approach to pleural empyema: uniportal or biportal?

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ABSTRACT

Background: Video-assisted thoracoscopic surgery (VATS) has become the standard treatment method for pleural empyema's surgical treatment. Postoperative pain, length of hospital stay, and cosmetic results are satisfactory, especially with the uniportal approach. In this study, we retrospectively evaluated and compared outcomes of patients treated with uniportal and biportal VATS.

Materials and Methods: A total of 73 patients who underwent VATS for empyema in our clinic between January 2017 and October 2020 were retrospectively evaluated. Patients who underwent uniportal and biportal VATS were compared in terms of age, sex, side and stage of empyema, length of hospital stay, comorbidities, smoking history, American Society of Anesthesiologists (ASA) physical status score, number of chest tubes placed postoperatively, postoperative Heimlich valve placement, postoperative complications, postoperative drainage volume, recurrence, preoperative and 72-h postoperative C-reactive protein (CRP) and white blood cell (WBC) values, and postoperative 24-h and 72-h VAS (visual analog scale) pain scores. Univariate comparisons were done with R-based Jamovi statistical software.

Results: Fifty-two (71.2%) of the patients were male, and the mean age was 57 (IQR 41-67). Empyema was stage 2 in 38 (52.1%) patients and stage 3 in 35 (47.9%) patients. VATS was uniportal in 52 patients (71.2%) and biportal in 21 patients (28.8%). The uniportal VATS group had significantly lower drainage volume ($p = 0.006$) and VAS scores ($p < 0.001$). There were no statistical differences in the other parameters.

Conclusions: Our data indicate that uniportal VATS was superior to biportal VATS for the treatment of empyema in terms of postoperative pain and drainage.

Keywords: minimally invasive surgery, thoracoscopy, video-assisted thoracoscopic surgery, empyema

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Introduction

Pleural empyema is defined as the presence of pus in the pleural space or positive Gram staining of pleural effusion and/or isolation of a microorganism in pleural fluid culture [1]. It usually occurs secondary to pneumonia, and morbidity and mortality rates are between 2% and 30% [2]. Other causes are surgery, thoracic interventions, esophageal diseases, and abdominal sepsis [3].

Parapneumonic effusion develops in 14% to 19% of patients with community-acquired pneumonia, and approximately one-third of these cases occur as complicated parapneumonic effusion or empyema [4,5]. The American Thoracic Society classified pleural empyema in three stages: stage 1 is the exudative stage, stage 2 is the fibrinopurulent stage, and stage 3 is the organization stage [6]. The British Thoracic Society recommends drainage (thoracentesis/tube thoracostomy) and medical treatment for stage 1 empyema. In contrast, for stage 2 and stage 3 cases, removal of debris and purulent tissues to enable re-expansion is recommended, with deloculation and decortication if necessary [7]. Thoracotomy and video-assisted thoracoscopic surgery (VATS) are the preferred approaches when surgical treatment of empyema is required, with VATS yielding satisfactory results [7,8]. Different publications on this popular topic have concluded that the VATS technique is effective and safe for treatment regardless of the stage [9-11]. Early surgical intervention was also shown to significantly reduce the length of hospital stay, costs, and mortality and morbidity rates, especially in patients with stage 2 and stage 3 empyema [12]. A few studies in the literature compare the effectiveness and outcomes of U-VATS and B-VATS in pneumothorax patients [13-15]. There are also reports in the literature of the results and features of lung resection performed with U-VATS [16,17].

Our study is the first to compare U-VATS and B-VATS in the treatment of empyema; our paper retrospectively evaluates the efficacy and clinical outcomes of empyema treated with uniportal VATS (U-VATS) and biportal VATS (B-VATS) techniques.

Material and Methods

This retrospective chart review included 73 patients who underwent VATS for empyema between January 2017 and October 2020. In this period 324 VATS performed for different indications. Five patients with missing data were not included (Figure 1). The patients'

age, sex, side and stage of empyema, length of hospital stay, comorbidities, smoking history, American Society of Anesthesiologists (ASA) physical status score, surgical technique (U-VATS or B-VATS), postoperative Heimlich valve placement, number of chest tubes placed postoperatively, postoperative complications, postoperative drainage volume, recurrence, preoperative and 72-h postoperative C-reactive protein (CRP) and white blood cell (WBC) values, and visual analog score (VAS) at postoperative 24 and 72 hours were analyzed. These parameters were compared between the U-VATS and B-VATS groups.

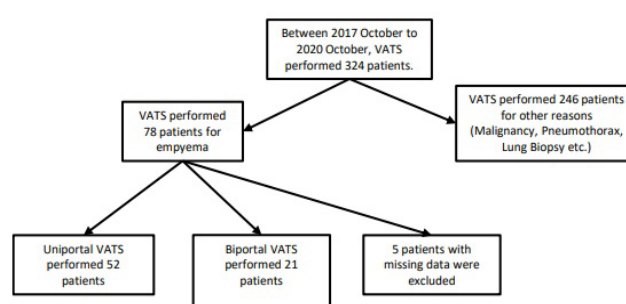


Figure 1. Study flow chart.

ASA scores were obtained from the anesthesiologist's consultation notes, and VAS scores were obtained from the patients' follow-up charts.

All patients were evaluated with chest computed tomography (CT) and ultrasound (US). Pleural fluid and/or sputum culture samples were collected from all patients, and empiric broad-spectrum antibiotic therapy was administered until the results were obtained. Specific antibiotic treatment was administered according to culture results.

During this period, if the amount of free fluid detected in chest CT or US was 1000-1500 mL or more, drainage was performed by tube thoracostomy. VATS was performed in patients who could not be re-expanded and showed no clinical improvement after drainage and those with intense septation and fluid volume less than 1000 mL [18]. All operations were initiated as uniportal, either using the existing drain incision or through an incision made at the site of the most significant free fluid detected on chest US. In necessary cases (e.g., difficult manipulation), a second entry site was made, and the procedure was completed as biportal. After deloculation and complete removal of the tissue debris, patients whose lungs were easily expanded were evaluated as stage 2, while those who had limited ventilation

and incomplete expansion and required decortication were evaluated as stage 3. Patients with impaired postoperative lung expansion (incomplete expansion despite decortication due to intraparenchymal infection severity) and immobile patients (e.g., home care, neurological patients) were discharged with Heimlich valve and followed up. Approval for this retrospective, descriptive, single-center study was obtained from the Clinical Research Ethics Committee of Recep Tayyip Erdogan University Faculty of Medicine (decision no: 2020/193, dated 03.09.2020).

VATS Technique

All surgical procedures were performed under general anesthesia with one-lung ventilation using a selective double-lumen endotracheal tube. The patients were positioned in lateral decubitus with the upper arm suspended in flexion and abduction. The day before the procedure, chest US was performed at the bedside with patients in the lateral decubitus position, and the site of most abundant loculation or fluid was marked, preferably on the midaxillary line. Chest exploration was performed with a 10-mm 30° thoracoscope introduced through an incision at the marked location or through the existing drain incision if present. Adhesions at the entry site were bluntly dissected using fingers and endoscopic instruments to create space. If this space was considered suitable after exploration, the incision was extended to 3 cm, and a tissue-protective retractor (Alexis O wound protect/retractor) was placed. The endoscopic instruments were introduced through this port, and sharp and blunt dissections were performed. If space was not considered adequate for dissection and decortication and manipulation was difficult, a second port incision was made at an appropriate site determined intraoperatively.

The procedures started with rupturing the septa (deloculation), debridement, and evacuation of the effusion. All infected tissues on the pleural surfaces, diaphragm, and sinuses were gently cleared using ovarian forceps. Effusion and tissue samples were obtained for simple culture, tuberculosis culture, cytology, and pathologic examination. Adhesions were separated by sharp and blunt dissection from the diaphragm to the apex. The thoracic cavity was washed several times with warm saline and cleared of infectious tissue and fluid residues by aspiration. After the lung tissue was released within the thoracic cavity, the parietal pleura's thickened sections were decorticated using Kelly forceps and gauze balls. The

affected areas of the visceral pleura were detected by venting the lung. While the lung was ventilated at a low volume, the visceral pleura was separated from the lung surface at a suitable site using Kelly forceps, gauze balls, and ovarian forceps. Starting dissection from this site, all affected pleural surfaces were decorticated. No intervention was performed for minimal leaks from lacerated parenchymal surfaces. Deeply lacerated parenchymal surfaces with massive leaks were repaired by primary suture or reinforced with tissue adhesive.

Local analgesia with 3 mL of 0.5% isobaric bupivacaine (5 mg/mL) was administered by thoracoscope at two intercostal levels above and below the utility incision and at the drain incision. At the end of the procedure, in most cases, a single chest drain (preferably 32 F) was placed after confirming the lung was fully ventilated and expanded. Patients who were expected to have more prolonged postoperative expansion and recovery time received two drains, a 32 F tube at the basal position and 28 F at the apex.

The decision to remove the chest tube was made by evaluating the patient's medical treatment duration, radiological evidence of lung expansion, clinical condition (fever, dyspnea, deterioration of inflammatory markers), decrease of daily drainage below 100 mL and absence of air leak. Clinically stable patients had completed medical treatment and showed normal inflammatory parameters but had lung expansion defect, those with persistent air leak (even minimal), and home care (bed-bound) patients were discharged with a Heimlich valve. These patients' drains were removed upon demonstration of clinical and radiological improvement during follow-up.

The mean postoperative follow-up time of the patients was 141 days. Contrast-enhanced chest CT scans were performed at 3rd and 6th months of follow-up.

Statistical Analysis

R-based Jamovi statistical software (Version 1.6.5 for Mac OS; <https://jamovi.org>) was used for statistical analysis. The distribution characteristics of continuous data were evaluated using the Shapiro-Wilk test, and Q-Q plots. Non-normally distributed data were expressed as the median and interquartile range (25th percentile-75th percentile) and compared between the U-VATS and B-VATS groups using the Mann-Whitney U test. Categorical variables were reported as number (n) and frequency (%) and compared between the groups using

Pearson's chi-squared (χ^2) test. A statistical significance level of $p < 0.05$ was used for all tests.

Results

Fifty-two (71.2%) of the patients were male, and the mean age was 57 (IQR 41-67). Twenty-seven patients (36.9%) were smokers. In terms of comorbidities, 10 patients (13.7%) had chronic obstructive pulmonary disease, 20 (27.4%) had ischemic heart disease, 12 (16.4%) had hypertension, and 9 (12.3%) had diabetes. Empyema was on the right side in 49 patients (67.1%) and was stage 2 in 38 patients (52.1%) and stage 3 in 35 patients (47.9%). Preoperative ASA score was ASA-2 in 25 patients (34.2%), ASA-3 in 31 patients (42.5%), and ASA-4 in 17 patients (23.3%).

The most common symptoms were fever in 46 (63.0%), cough in 25 (34.2%), dyspnea in 9 (12.3%), and chest pain in 7 patients (9.6%). Empyema occurred due to bronchopleural fistula secondary to lung cancer in 1 patient, while all other patients had complicated parapneumonic effusion.

A total of 52 patients (71.2%) underwent U-VATS, and 21 (28.8%) underwent B-VATS. A single chest drain was placed in 54 patients (74.0%), while 19 patients (26.0%) had two chest drains. Nine (12.3%) of the patients were discharged with a Heimlich valve, and 64 (87.7%) were discharged after drain removal. The median postoperative follow-up time was 141 (IQR 105-176) days (Table 1).

The patients were divided into uniportal and biportal groups and compared in terms of age, sex, smoking, empyema side, drainage, recurrence, preoperative and postoperative CRP (mg/L; normal range 0-5 mg/L), preoperative and postoperative WBC ($\times 10^3/\mu\text{L}$), empyema stage (stage 2 or 3), preoperative and postoperative hospital stay, discharge with Heimlich valve, number of chest tubes placed postoperatively, VAS at postoperative 24 and 72 h, and procedure times. Of these findings, U-VATS was only beneficial in terms of drainage volume and VAS scores ($p < 0.001$ and $p = 0.006$, respectively). There were no statistical differences in the other characteristics (Table 2).

The only mortality occurred in the patient with empyema due to bronchopleural fistula secondary to lung cancer. This patient underwent B-VATS and died in the intensive care unit due to poor general condition during postoperative follow-up. Empyema recurred in 3 patients, of whom 2 were treated with re-VATS, and 1

underwent open decortication (OD) in the early period. Two patients with relapse underwent U-VATS, and the other underwent B-VATS. Both patients who underwent re-VATS were in the uniportal group, and a new port incision was not required in the second operation. No late relapse was observed. In terms of minor complications, inadequate expansion persisting longer than 5 days was observed in 2 patients (9.5%), anemia in 2 patients (2.7%), air leak persisting for longer than 5 days in 3 patients (8.1%), and surgical site infection in 1 patient (4.1%) (Table 3).

Table 1. Demographic and clinical characteristics of the patients (n=73).

Characteristic	Median (IQR) or n (%)
Age (years)	56 (41-67)
Sex	
Male	52 (71.2%)
Female	21 (28.8%)
Smoker	27 (36.9%)
Mean follow-up time (days)	141 (105-176)
Comorbidity	
COPD	10 (13.7%)
Ischemic heart disease	20 (27.4%)
Arterial hypertension	12 (16.4%)
Diabetes	9 (12.3%)
Side of empyema	
Right	49 (67.1%)
Left	24 (32.9%)
ASA score	
ASA 2	25 (34.2%)
ASA 3	31 (42.5%)
ASA 4	17 (23.3%)
Empyema stage	
Stage 2	38 (52.1%)
Stage 3	35 (47.9%)
VATS technique	
Uniportal	52 (71.2%)
Biportal	21 (28.8%)
Number of chest tubes	
One	54 (74.0%)
Two	19 (26.0%)
Discharged with Heimlich valve	
Yes	9 (15.1%)
No	62 (84.9%)

Abbrev.: IQR: Interquartile range (25th-75th percentile), COPD: Chronic obstructive pulmonary disease, ASA: American Society of Anesthesiologists, VATS: Video-assisted thoracoscopic surgery.

Table 2. Comparison of demographic and clinical characteristics of empyema patients by VATS port number.

Parameter	Uniportal (n=52) Median (IQR) or n (%)	Biportal (n=21) Median (IQR) or n (%)	p
Age (years)	55.5 (41.5-67)	58 (41-65)	0.98
Sex			0.58
Male	38 (73.1%)	14 (66.7%)	
Female	14 (26.9%)	7 (33.3%)	
Smoking history			0.90
Yes	19 (36.5%)	8 (38.1%)	
No	33 (63.5%)	13 (61.9%)	
Side			0.54
Left	16 (30.8%)	8 (38.1%)	
Right	36 (69.2%)	13 (61.9%)	
Drainage volume (mL)	465 (287-600)	750 (550-1050)	0.006
Relapse	2 (3.9%)	1 (4.8%)	0.86
C-Reactive protein (mg/L)			
Preop	13.9 (7.52-26.25)	13 (8.27-27)	0.76
Postop Day 3	5 (1.99-13.73)	7.38 (2.53-13.4)	0.55
White blood cell count (×103/μL)			0.83
Preop	13.9 (11.3-19.1)	13.4 (9.5-16.9)	0.95
Postop day 3	8.7 (6.9-12.1)	8.7 (6.5-14.7)	
Empyema stage			0.62
Stage 2	28 (53.8%)	10 (47.6%)	
Stage 3	24 (46.2%)	11 (52.4%)	
Hospital length of stay (days)			
Total	15 (13-24)	18 (17-23)	0.32
Preop	7 (6.7-10)	7 (6-10)	0.99
Postop	8 (6-12)	11 (9-12)	0.07
Discharge with Heimlich valve	7 (14%)	2 (9.5%)	0.20
Number of chest tubes			0.36
One	40 (76.9%)	14 (66.7%)	
Two	12 (23.1%)	7 (33.3%)	
VAS Pain Score			
Postop day 1	3 (3-4)	3 (2-3)	<0.001
Postop day 3	5 (4-6)	4 (3-5)	<0.001
Procedure time (min)	143 (132-169.2)	154 (134-169)	0.069

Abbrev.; VATS: Video-assisted thoracoscopic surgery, IQR: Interquartile range (25th-75th percentile), VAS: Visual analog score, Preop: Preoperative, Postop: Postoperative.

Table 3. Minor complications.

Complication (n = 8) (10.8%)	n (%)
Insufficient expansion > 5 days	2 (2.7%)
Anemia	2 (2.7%)
Air leak > 5 days	3 (4.1%)
Surgical site infection	1 (1.3%)

Discussion

Surgical treatment is currently the standard approach for stage 2 and 3 empyema. VATS decortication (VATSD) and OD are the preferred techniques for this purpose. Recent publications have discussed the superiority of VATS in the treatment of pleural

empyema, but these studies generally compared it with thoracotomy. The effectiveness of VATSD in the treatment of pleural empyema has been demonstrated in many recent studies. In 2001, Waller et al concluded that in addition to the benefits of the minimally invasive method (shorter hospital stay, less pain), VATSD was as effective as OD in the treatment of stage 3 empyema [19]. Chan et al retrospectively evaluated 77 patients who underwent surgery for empyema (41 VATSD and 36 OD) and reported that VATSD was as effective as OD and was superior in terms of length of hospital stay and postoperative pain [20]. Cardillo et al compared OD

and VATSD in a retrospective study of 308 patients and concluded that thoracoscopic methods were the gold standard for empyema [21]. In their 2010 meta-analysis of 14 studies, Chambers et al reported that VATSD was superior to OD in the treatment of empyema in terms of postoperative pain, hospital stay, 30-day mortality, and complication and relapse rates [22].

In another recent study, Samancılar et al performed VATSD in 54 patients and intrapleural fibrinolytic treatment in 24 patients and reported satisfactory results in both groups [23]. Pan et al also determined in their recent review that VATS was as good as thoracotomy for the treatment of empyema and was superior in terms of length of hospital stay and postoperative complications. They reported that thoracotomy might be required in a very small number of cases [8].

There are few articles on empyema surgery with U-VATS in the literature. In their 2010 study, Shahin et al evaluated 81 empyema cases, of which 60 underwent VATSD and 21 underwent OD. VATS was performed with 2 or 3 ports, but no comparison was made [24]. Bongiolatti et al evaluated 30 empyema patients who underwent U-VATS and 34 who underwent OD according to preoperative US findings and stated that VATSD was as effective as OD and was associated with a shorter length of hospital stay [25]. In another recent study, Ismail et al performed decortication by U-VATS to a total of 35 patients with stage 2 and 3 empyema. Their results regarding the length of hospital stay, chest tube duration, postoperative pain, and complications were consistent with the literature, and they also reported favorable cosmetic results [26].

In the present study, 3 patients had an early relapse, and 2 of those patients were treated by re-VATS while the other required OD. There was no late relapse, and the rate of minor complications was very low. Our results are consistent with the literature in terms of relapses and minor complications. The effectiveness of U-VATS and B-VATS were found to be consistent with the literature. As in other studies, U-VATS was associated with less postoperative drainage and pain than B-VATS. Although procedure times and postoperative hospital stays were shorter with U-VATS, the differences were not statistically significant.

Limitations of the Study

The retrospective design and inability to conduct multivariate analyses due to the small number of patients

are limitations of this study. The main limitation is that our analysis was conducted in a small sample from a limited area.

In conclusion, VATS decortication is safe and effective for the treatment of stage 2 and 3 empyema in selected patients. Numerous studies already confirmed that VATS is indisputably superior to thoracotomy in terms of less postoperative pain, rapid recovery, and early discharge. The results of our study support the literature. The patients' compliance with postoperative mobilization and respiratory exercises was nearly perfect due to the brief hospital stay and low pain, especially after U-VATS procedures. We believe that as surgeons gain more experience, U-VATS will become the gold standard in the treatment of empyema in the coming years.

Declaration of conflicting interests

The authors declared no conflicts of interest with respect to the authorship and/or publication of this article.

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Ethics approval

Approval for this retrospective, descriptive, single-center study was obtained from the Clinical Research Ethics Committee of Recep Tayyip Erdogan University Faculty of Medicine (decision no: 2020/193, dated 03.09.2020).

Authors' contribution

KT; conceptualized and designed the study, collected and analyzed data. KT,HT; revised the final version of the manuscript and co-wrote the paper. All authors approved the final manuscript as submitted and agree to be accountable for all aspects of the work.

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Original Article

Conservative approach versus invasive approach for pneumothorax secondary to blunt trauma

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ABSTRACT

Background: The management of minimal pneumothorax, which has been detected more frequently with the increasing use of computed tomography, especially after blunt trauma, is controversial. This study aimed to examine the results of patients with pneumothorax secondary to blunt trauma and contribute to the treatment protocol for this group of patients.

Materials and Methods: A retrospective analysis of patients with pneumothorax after blunt trauma (n = 103) presenting to the emergency department during the 1-year period from January 2019 to January 2020 was performed. Oxygen inhalation was performed in patients who were followed up conservatively, and tube thoracostomy was performed in patients who were treated invasively.

Results: Among 766 patients with blunt trauma, 141 (18.4%) patients had a traumatic pneumothorax. Mechanical ventilation was required in 26.2% (n = 27) patients. Further, 73 (70.8%) patients with pneumothorax were followed up conservatively. The remaining 30 (29.1%) patients underwent tube thoracostomy. The median size of the pneumothorax (9 vs. 29 mm; $p < 0.0001$; hazard ratio [HR] = 5.08) was significantly different between the groups. Multivariate analyses revealed that positive pressure ventilation and pneumothorax size did not pose an additional risk of failure in conservative management (HR = 1.49; $p = 0.25$ and HR = 10.2; $p = 0.09$, respectively). Multivariate analyses revealed that the presence of hemothorax and subcutaneous emphysema (HR = 14.4; $p = 0.018$ and HR = 13.1; $p = 0.019$, respectively) led to failure of conservative treatment.

Conclusions: The presence of hemothorax and subcutaneous emphysema was an independent factor leading to failure of conservative treatment. In the presence of these two findings indicating major trauma, invasive treatment is recommended.

Keywords: trauma, pneumothorax, tube thoracostomy, surgery, mechanical ventilation

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Introduction

Thoracic trauma occurs in approximately two-thirds of multiple trauma cases and is the primary cause of mortality in 25% patients with trauma [1]. Traumatic pneumothorax occurs in one-fifth of patients with multiple traumas [2] and is the most significant potentially life-threatening pathology in blunt thoracic trauma [3].

In such cases, the ideal treatment method is tube thoracostomy. The management of minimal pneumothorax that cannot be detected by x-rays, which has been detected more frequently with the increasing use of computed tomography (CT), especially after blunt trauma, is controversial.

Tube thoracostomy is not an uncomplicated procedure. A meta-analysis has reported an average complication rate of 19% [4]. In such cases, it is clinically difficult to perform conservative treatment up to the stage at which an invasive procedure should be performed. Although uncomplicated traumatic pneumothorax can be well-tolerated, conservative approach recommended because tube thoracostomy placement can potentially prevent morbidity and mortality [5].

In the guidelines published by the American College of Surgeons, chest tube insertion is recommended for any traumatic pneumothorax. However, asymptomatic pneumothorax can be managed by observation and aspiration at the discretion of the treating physician. In addition, the need for a chest drain has been reported in patients receiving either general anesthesia or positive pressure ventilation (PPV) to avoid life-threatening pneumothorax [6].

These guidelines are based on the results of cases of large-volume pneumothorax detected by x-ray. Currently, CT scanning is available in almost every trauma unit for patients with multiple traumas, and the frequency of pneumothorax diagnosis at the sub-centimeter level has increased.

Although minimal pneumothorax is encountered incidentally in 2% patients, it can be found in 10% patients after blunt thoracic trauma. This raises the question of whether these small “minimal” pneumothoraxes, which account for 76% of all traumatic pneumothorax cases, can be left untreated, especially when PPV is required [7]. Although an early study has shown a high failure rate in conservatively treated patients receiving

PPV [8], studies have suggested that minimal pneumothoraxes, including those in patients receiving PPV, can be managed conservatively [5,7,9-11].

Case studies have also suggested that it is possible to treat larger pneumothoraxes through observation [12]. Therefore, this study aimed to examine the results of patients with pneumothorax secondary to blunt trauma and contribute to the treatment protocol for this group of patients.

Material and Methods

This single-center, retrospective study was conducted at University of Health Sciences, Bakırköy Dr. Sadi Konuk Training and Research Hospital, Department of Thoracic Surgery between January 2019 and January 2020. In addition to demographic information, data on the type of injury, injury severity score, radiological dimension of pneumothorax, additional pathologies, treatment, need for PPV, length of stay (LOS), complications, and mortality were recorded. Oxygen inhalation (2-4 L/min) was performed in patients followed up conservatively, whereas 28-32F tube was inserted in patients treated with tube thoracostomy. The injury severity score (ISS) was used to determine trauma severity [13]. The ISS is correlated with mortality, morbidity, and hospitalization time after trauma. An ISS of 1-8, 9-15, 16-24, and ≥ 25 indicated minor, moderate, severe, and very severe trauma, respectively [14]. The scoring system described by De-Moya et al was used to measure the level of pneumothorax observed on thorax CT [15]. The score is composed of two parts; (1) the largest diameter of the air pocket and (2) its relationship with the pulmonary hilum. The first step was to measure the largest air collection along a line perpendicular to the chest wall or mediastinum in millimeters. The pulmonary hilum was then identified, and it was determined whether the pneumothorax crossed the trans hilar axial plane. If the pneumothorax line did not cross this plane, 10 was added to the millimeter scale. If the pneumothorax line crossed this plane, 20 was added to the largest dimension measured. The sum of the two numbers was the final score. The study protocol was approved by the University of Health Sciences, Bakırköy Dr. Sadi Konuk Training and Research Hospital Ethics Committee (date/no: 05.04.2021/2021/193).

Statistical Analysis

Non-parametric methods were used to compare the two

independent groups because of the small number of patients recruited in the study. Fisher's exact test was used to compare categorical variables, whereas the Mann–Whitney U test was used to compare continuous or ordinal variables.

Receiver operating characteristic (ROC) analysis was used to successfully estimate the pneumothorax size and De Moya score for optimal cutoff points in conservative follow-up. Univariate analysis was used to compare patients undergoing and not undergoing chest tube insertion after CT. Factors with $p < 0.1$ were then entered into a backward stepwise likelihood regression model. In the model, statistical significance was set at $p \leq 0.05$. Data were analyzed using IBM SPSS Statistics version 20 (IBM Corp., Armonk, NY, USA).

Results

Between January 2019 and January 2020, among 766 patients with blunt trauma who underwent emergency surgery, 142 patients had hemopneumothorax secondary to blunt trauma. After excluding patients who did not meet the study criteria, 103 patients were analyzed (Figure 1).

Table 1 summarizes the demographic characteristics, type of injury, ISS, pneumothorax characteristics, man-

agement, and treatment results of the non-conservatively and conservatively managed patients. Among 766 patients with blunt trauma, 141 (18.4%) patients had a traumatic pneumothorax.

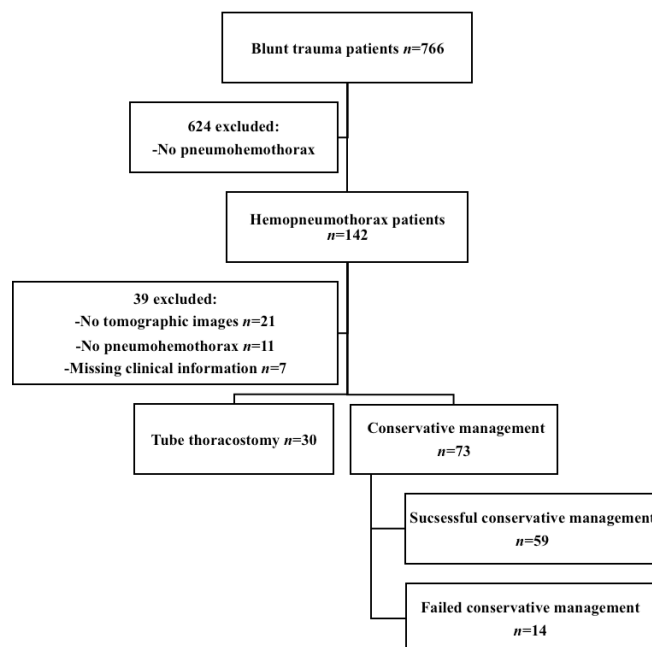


Figure 1. Study group and excluded patients.

Table 1. Characteristics of patients treated with conservative and non-conservative management (n=103).

Patients' characteristics	Conservative (n=73)	Non-conservative (n=30)	p
Age, SD, y	44.3 (18.6)	41.8 (21.1)	0.55
Male sex, %	52 (71.2)	22 (73.3)	0.83
Cause of injury, %			
In-car traffic accident	9 (12.3)	9 (30)	0.03
Non-vehicle traffic accident	10 (13.6)	3 (10)	0.61
Fall	34 (46.5)	12 (40)	0.54
Blows	12 (16.4)	4 (13.3)	0.69
Motorcycle accident	8 (10.9)	2 (6.7)	0.69
Mean ISS, SD	7.2 (5.4)	12.4 (7.6)	0.0002
Mean pneumothorax size (mm), SD	10.7 (6.6)	29.8 (15.2)	<0.0001
Mean pneumothorax size (mm), SD (De Moya Method)	21 (7.9)	45.9 (18.2)	<0.0001
Received positive pressure ventilation, %	15 (20.5)	12 (40)	0.04
Hemothorax, %	19 (26)	16 (53.3)	0.008
Contusion, %	41 (56.1)	25 (83.3)	0.009
Subcutaneous emphysema, %	24 (32.8)	14 (46.6)	0.18
Rib fracture, %	45 (61.6)	26 (86.6)	0.013
Average length of stay, SD	4.2 (4.3)	6 (4.4)	0.58
Average ICU length of stay, SD	0.6 (1.3)	0.9 (1.7)	0.96
Mortality, %	2 (2.7)	6 (20)	0.003

Abbrev.: SD=standard deviation, ISS=Injury severity score, ICU=intensive care unit. ($p < 0.05$ was considered statistically significant and marked in bold).

The mean age of the patients was 43.5 (standard deviation [SD] = 19.3), and most patients were men (n = 74, 71.8%). The most common type of trauma was falling (n = 46, 44.6%). The mean ISS was calculated as 8.7 (SD = 6.5), which was defined as non-major traumas [14]. Mechanical ventilation and intensive care were required in 26.2% (n = 27) patients. In a series of 103 patients, mortality occurred in eight (7.8%) patients.

Table 2 summarizes the characteristics and outcomes of successful and failed observational management.

In total, 73 (70.8%) patients with pneumothorax were followed up with conservative methods and observational treatments. The remaining 30 (29.1%) patients underwent tube thoracostomy. Regarding the comparison of the pneumothorax size, patients followed up conservatively had a significantly smaller pneumothorax than those who underwent tube thoracostomy (median, 9 vs. 29 mm; $p < 0.0001$). This difference was also observed in the De-Moya score (median, 19 vs. 45.5; $p < 0.0001$).

The distribution of patients treated conservatively and invasively according to the pneumothorax size is shown in Figure 2. The pneumothorax size was < 10 mm in most patients who were followed up conservatively.

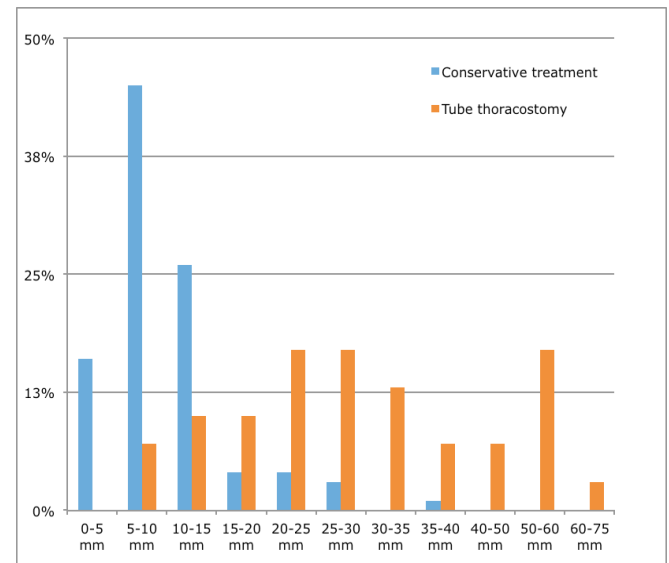


Figure 2. Scatter diagram of conservatively and invasively treated cases according to pneumothorax size.

Table 2. Characteristics and outcomes of conservatively managed patients.

Patients' characteristics	Successful conservative management (n=59)	Failed conservative management (n=14)	p
Age, SD, y	45.1 (19.8)	40.6 (12.6)	0.42
Male sex, %	43 (72.8)	29 (64.2)	0.52
Cause of injury, %			
In-car traffic accident	6 (10.1)	3 (21.4)	0.24
Non-vehicle traffic accident	9 (15.2)	1 (7.1)	0.43
Fall	30 (50.8)	5 (35.7)	0.13
Blows	7 (11.8)	5 (35.7)	0.03
Motorcycle accident	7 (11.8)	1 (7.1)	0.61
Mean ISS, SD	6.7 (5.2)	9.3 (5.7)	0.10
Mean pneumothorax size (mm), SD	9.1 (4.6)	17.8 (8.8)	<0.0001
Mean pneumothorax size (mm), SD (De Moya method)	19.2 (5.9)	28.5 (10.9)	<0.0001
Received positive pressure ventilation, %	8 (13.5)	7 (50)	0.002
Hemothorax, %	11 (18.6)	8 (57.1)	0.003
Contusion, %	34 (57.6)	7 (50)	0.6
Subcutaneous emphysema, %	14 (33.7)	10 (71.4)	0.0007
Rib fracture, %	35 (59.3)	9 (64.2)	0.73
Average length of stay, SD	3.5 (3.9)	7.3 (4.8)	0.05
Average ICU length of stay, SD	0.45 (1.1)	1.5 (1.6)	0.004
Mortality, %	1 (1.6)	1 (7.1)	0.003

Abbrev.: SD=standard deviation, ISS=Injury severity score, ICU=intensive care unit.

The invasively treated group had more severe trauma, a higher ISS (mean 12.4 vs 7.2; $p = 0.0002$), more frequent need for intensive care (40% vs. 20.5%; $p = 0.04$), more frequent hemothorax (53.3% vs. 26%; $p = 0.008$), more frequent lung contusion (83.3% vs. 56.1%; $p = 0.009$), and more frequent rib fracture (86.6% vs. 61.6%; $p = 0.013$) than the conservatively treated group.

There were no significant differences between the two groups in terms of age, sex, type of trauma, and length of stay

The mortality rate was higher in the invasively treated group than in the conservatively treated group (20% vs. 2.7%; $p = 0.003$).

Among 73 patients treated conservatively, 59 (80.8%) patients did not require additional interventions. Mechanical ventilation was performed in 15 (20.3%) patients treated conservatively. Among 14 patients requiring additional intervention, seven (50%) patients were treated with mechanical ventilation, and this rate was significantly higher than that in the successful conservative treatment group (50% vs. 13.5%; $p = 0.002$).

Based on the univariate analysis results, age, sex, ISS, presence of lung contusion, and rib fracture were not significantly related to failure of conservative treatment (Table 3).

Table 3. Hazard ratios for failed conservative management.

Variable	Hazard Ratio	p	95% CI
Age (≥ 65 vs < 65)	1.17	0.74	0.59-3.45
Male sex	1.06	0.91	0.35-3.22
ISS (≥ 15 vs < 15)			
(Severe vs. moderate and minor)	1.09	0.88	0.33-3.59
ISS (≥ 9 vs < 9)			
(Minor vs. moderate and severe)	0.87	0.65	0.43-3.25
Size of pneumothorax (≥ 11 mm vs. < 11 mm)	5.08	0.01	1.41-18.2
De-Moya score (≥ 22 vs < 22)	4.21	0.01	1.34-12.4
Received positive pressure ventilation	1.73	0.31	0.59-5.06
Hemothorax	2.53	0.08	0.87-7.33
Lung contusion	0.43	0.12	0.15-1.25
Subcutaneous emphysema	4.33	0.02	1.2-15.5
Rib fracture	1.15	0.80	0.36-3.71

Abbrev.: ISS=Injury severity score, confidence interval.

$p < 0.05$ was considered statistically significant and marked in bold.

The median pneumothorax size (9 vs. 29 mm; $p < 0.0001$; hazard ratio [HR] = 5.08) and median De-Moya score (19 vs. 45.5; $p < 0.0001$; HR = 3.7) were significantly different between the groups.

To indicate when to discontinue conservative pneumothorax treatment, the cutoff size of the pneumothorax was determined to be 11 mm using the ROC curve. The optimal cutoff value for the De-Moya scoring system was 22 (Figure 3).

However, when the pneumothorax size data were evaluated using multivariate analysis, its effect on failure of conservative treatment was not found to be statistically significant (Table 4). Multivariate analyses revealed that PPV did not pose an additional risk of failure in conservative management (HR = 1.49; $p = 0.25$). In contrast, multivariate Cox regression analysis revealed that the presence of hemothorax and subcutaneous emphysema (HR = 14.4; $p = 0.018$ and HR = 13.1; $p = 0.019$, respectively) led to failure of conservative treatment (Table 4).

The mean ISS of the eight patients who died was 26.4 (> 25 representing serious or critical injuries). In five of eight cases, the patients died due to cranial traumatic pathologies, and in the remaining three cases, the patients died due to uncontrollable abdominal bleeding (the liver, spleen, and inferior vena cava).

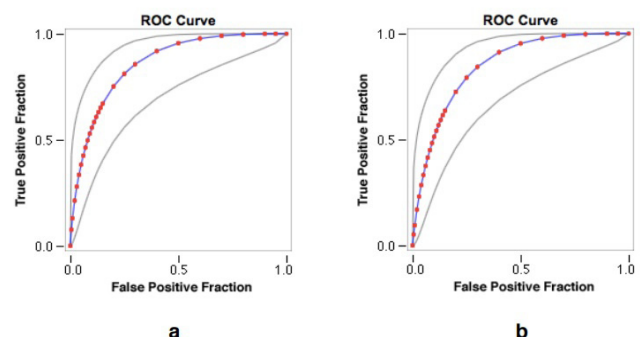


Figure 3. Receiver operating characteristic (ROC) analysis of pneumothorax size and moya score affecting success in conservative treatment. Area under curve (AUC) = 0.86, sensitivity = 80.9%, specificity = 76.2% for pneumothorax size cutoff value of 11mm (a), AUC = 0.849, sensitivity = 79.1%, specificity = 75.3% for De-moya score cutoff value of 22 mm (b).

Table 4. Multivariable Cox regression analysis for failure of conservative management.

Variable	Hazard ratio	p	95% CI
Size of pneumothorax (≥ 11 mm vs. < 11 mm)	10.2	0.09	0.82-138
De-Moya score (≥ 22 vs < 22)	9.7	0.14	0.64-125
Hemothorax	14.4	0.018	1.69-137
Subcutaneous emphysema	13.1	0.019	1.86-110

$p < 0.05$ was considered statistically significant and marked in bold.

Discussion

Tube thoracostomy is a procedure with a complication risk ranging from 15% to 30% [11,16,17]. The current guidelines recommend chest tube insertion for traumatic pneumothorax, especially in patients receiving ventilation support, even if they are asymptomatic [6]. Asymptomatic patients who do not require a ventilator can be managed by observation or aspiration at the physician's discretion.

Some studies have reported that conservative treatment can be used in selected patients to determine whether minimal pneumothorax can be managed conservatively [5,11].

Studies on scoring systems that determine whether chest tube intervention is necessary for occult pneumothorax and those with larger patient groups are needed [15].

This study aimed to determine whether pneumothorax secondary to blunt trauma could be treated conservatively and examine situations requiring chest tube insertion.

In this study, positive pressure ventilation (PPV) did not pose a risk in terms of conservative treatment.

Studies have been focused on the role of conservative treatment in occult pneumothorax not initially visible on radiography [5,11].

Positive findings, including those in patients receiving PPV, have been included in the clinical guidelines [18]. Although these studies are useful for establishing treatment methods for traumatic pneumothorax, they have limitations.

Currently, CT scanning has become routine in emergency departments, especially for patients with multiple traumas. However, guidelines mainly based on radiographic findings are lacking.

Whether a pneumothorax is visible on radiography not only is related to its size but can also be affected by other factors. For example, supine chest radiography has been shown to reduce the sensitivity [7].

The increase in CT rates causes more limited pneumothorax cases and constitutes a common decision problem in clinical practice. Hence, in this study, we examined the consequences of traumatic pneumothorax. Most patients (71%) were treated conservatively,

and most of them (59 of 73 [81%]) did not require additional intervention.

Among 14 patients requiring additional intervention, seven (50%) patients were treated with mechanical ventilation, and this rate was significantly higher than that in patients successfully treated conservatively (50% vs. 13.5%; $p = 0.002$). Multivariate analyses revealed that PPV did not pose an additional risk of failure in conservative management (HR = 1.34; $p = 0.21$).

This finding is consistent with that the current literature on minimal pneumothorax. Moore et al [11] reported a 14% failure rate when PPV was performed in patients managed via observation. The rate was 4.5% in patients not receiving PPV, but it was identified as an independent predictor of failed management in multivariate analysis. In addition, Brasel et al [10] and Enderson et al [8] reported a high failure rate (22% and 53%, respectively) during conservative follow-up in patients receiving PPV. Although Brasel et al [10] concluded that observation is reliable, Enderson et al [8] recommended tube thoracostomy for all patients requiring PPV. In light of this information, the discussions on the management of occult pneumothorax in patients receiving ventilation support are ongoing. In another study, Hoff et al [19] reported that minimal pneumothorax was observed in stable patients, independent of PPV. The OPTICC trial (ClinicalTrials.gov: NCT00530725) with randomization between chest tube insertion and observation in patients receiving PPV was completed in 2021, and the results are awaited.

Hefny et al found that the need for a tube thoracostomy in pneumothorax secondary to blunt trauma was associated with a pneumothorax volume of more than 30 ml. In this study, pneumothorax volume was calculated using software available in CT [21].

Moore et al. accepted the occult pneumothorax margin as 7mm in their prospective observational study. They found that this cut-off value did not affect success in conservative treatment [11].

Pneumothorax size > 11 mm was found to have a risk of failure in conservative treatment (HR = 5.08; $p = 0.01$) in univariate analysis. However, when the pneumothorax size data were evaluated in multivariate analysis, the effect on failure of conservative treatment was not found to be statistically significant (HR = 10.2; $p = 0.09$).

Pneumothorax size was previously thought to be a predictor of failure of conservative treatment [20]. To guide treatment, De Moya et al [15] proposed a scoring system that uses the size of occult pneumothorax and its relationship to the hilum. However, Moore et al [11] showed that pneumothorax size was not an independent predictor of failed observations.

Moore et al accepted the occult pneumothorax margin as 7mm in their prospective observational study. They found that this cut-off value did not affect success in conservative treatment[11].

Pneumothorax size is seen as the most important decision-making criterion for tube thoracostomy. However, in cases with minimal pneumothorax secondary to blunt trauma, a decision should be made together with additional pathologies.

In the present study, presence of hemothorax was determined as an independent variable (HR = 14.4; $p = 0.018$). Walker et al [22] showed that presence of hemothorax was the only independent factor predicting failure of conservative treatment for pneumothorax. This is also considered to be beneficial for patients in clinical practice. Even if draining the blood does not cause a life-threatening problem, it reduces the risk of complications such as infection and fibrothorax in the long term.

In the current study, the presence of subcutaneous emphysema was also found to be an independent variable (HR = 13.1; $p = 0.019$). Subcutaneous emphysema was interpreted as a sign of a complicated pneumothorax and significant damage to the visceral and parietal pleura. Conservative treatment failure was more common in patients with hemothorax and subcutaneous emphysema. In the presence of these two findings, closer follow-up is required, even if the pneumothorax is small.

Chest ultrasonography is superior to radiography for the diagnosis of pneumothorax in patients with trauma [23,24]. Due to the difficulty of mobilization and close follow-up with x-ray or CT, especially in multi-traumatized cases, it is a candidate to enter ultrasound trauma protocols in which the amount of pneumothorax and hemothorax can be followed together in the management of these cases. Ultrasound may play a useful role in the routine management of thoracic trauma.

The type of injury, ISS, and pneumothorax size were

not strong indicators of the need for intervention.

Although the need for ventilation support is controversial, both the current findings and previous study findings [5,11] have shown that pneumothorax can be managed conservatively. Most conservatively treated patients, including those requiring ventilation support and who do not pose an additional risk, were successfully treated without the need for chest drains.

In the presence of a high-volume ventilated lung, tube thoracostomy may be more complicated. Despite the classical guidelines recommending tube thoracostomy for pneumothorax detected in ventilator-dependent patients, the current study suggests that patients with minimal pneumothorax secondary to blunt trauma can be managed conservatively with close monitoring. The presence of hemothorax and subcutaneous emphysema was found to be an independent factor leading to failure of conservative treatment. In the presence of these two findings, indicating major trauma, invasive treatment is recommended.

Declaration of conflicting interests

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Ethics approval

Approval for this retrospective, descriptive, single-center study was obtained from the Clinical Research Ethics Committee of Recep Tayyip Erdogan University Faculty of Medicine (decision no: 2020/193, dated 03.09.2020).

Authors' contribution

Oİ; conceptualized and designed the study, collected and analyzed data, revised the final version of the manuscript and wrote the paper.

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Original Article

We need a common definition and treatment algorithm for displaced rib fracture

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ABSTRACT

Background: Displaced rib fracture (DRF) definition is frequently used to draw attention to severity and importance of fracture in daily practice. DRF is associated with increased morbidity and mortality in addition patients with DRF should be followed more carefully. Despite these characteristics, we do not have a clear definition for DRF concept and big differences of opinion among physicians could be monitored. In this study, we tried to reveal these perceptual differences and emphasized the importance of creating a common language for DRF.

Materials and Methods: We used a special and inventive survey form which contains visual section, true-false section and case reports. In the visual section, real tomography images and schematic drawings were presented to participants and asked which were compatible with DRF. In the true-false section, propositions about the definition of DRF were presented. Finally, imaginary trauma cases were presented and the minimum follow-up period was questioned.

Results: 156 physicians from 23 different center were included in the study. Of the participants, 56 (35.9%) were emergency physicians, 54 (34.6%) were thoracic surgeons and 46 (29.5%) were radiologists. The answers were statistically different for 3 tomography image ($p = 0.056$, $p < 0.001$, $p = 0.001$) and for 1 schematic drawings ($p = 0.001$). Again in 4 of the 7 true-false questions, there were significant differences between answers ($p = 0.001$, $p = 0.001$, $p = 0.005$, $p < 0.001$). The minimum follow-up period for a patient with DRF was also different between physicians, and have been recommended as 15.9 ± 2.2 (2-72 hours) by emergency physicians, 27.3 ± 5.5 (2-120) by radiologist and 31.5 ± 3.1 (2-120) by thoracic surgeons.

Conclusions: Our study clearly demonstrates a big conflict about DRF definition and treatment among physicians. There is also no consensus on the minimum follow-up time. We believe that our study will be a guide for multidisciplinary clinical studies on this subject.

Keywords: fractures, injuries, radiology, ribs, questionnaires, emergencies

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Introduction

Rib fracture is detected in about 10% of patients with thoracic trauma and the number of fractured ribs is an important indicator for determine the severity of trauma [1-3]. Similarly, displaced rib fractures (DRF) are known to be associated with severe thoracic organ injury and also with increased morbidity-mortality [1,4-7]. Additionally, late complications such as delayed hemothorax may develop, so longer hospital stays are recommended for patients with DRF. [1,4,8]. Despite these important features, the concept of “DRF” in daily practice seems to be interpreted differently by physicians. The same tomography image can be interpreted differently by different clinical physicians like radiologist/thoracic surgeons and sometimes same clinical physicians also can not reach to a consensus. Similarly there are different definitions for DRF in the literature [1,3,9]. In this study, we aimed to reveal this disagreement concretely. In this respect, we used a unique and detailed survey form which consist of 3 stages (visual section, true/false section and case report). Participants were selected from, the emergency department physicians, thoracic surgeons and radiologist who were interested in thoracic trauma and the rib fractures. By revealing this disagreement and relativity about defination, we tried to explain why a “gold standard” definition is needed for DRF. We hope that, similar studies with clinical correlation about DRF is required for a correct definition and our study will be a pioneer for this ideas.

Material and Methods

Our study was organized as a prospective survey study. Ethic approval was obtained from the Health Sciences University, Izmir Bozyaka Training and Research Hospital Ethics Committee. Participants were chosen from faculty members, lecturers, specialist doctors and assistant doctors (who completed at least one year) who were employed in the thoracic surgery, emergency department and radiology clinics. All questionnaires were surveyed by a thoracic surgeon and all interviews were conducted face to face. Informed volunteer consent was obtained from all participants. We reached physicians working in different cities, different hospitals across the country and as a result, a total of 23 different clinics were included in the survey.

The questionnaire form was composed of three sec-

tions as: visual section, true-false section and case report section. The visual section consists two sub-section; in first part real tomography images used and in second part, a schematic rib drawing used. Tomography images were selected from patient’s radiological exams who were followed-up in our clinic before (A written informed consent was also obtained from all patients for using tomography images). A total of 9 high resolution images were selected from patient’s tomographies. These images were aligned from the minimally fracture to the severe displaced rib fracture (Figure 1). Participants were asked to choose the ones of these images which they think compatible with DRF. In second visual section (containing schematic drawings) the participants were asked to choose the options that were compatible with DRF (Figure 2). In this schematic drawing part, a descriptive drawing was also included to be used in the next part of survey (Figure 2). After these sections, the participants were asked about the definition of DRF and adjacent lung injury findings, and the participants were asked to mark propositions as “true or false” (Figure 3).

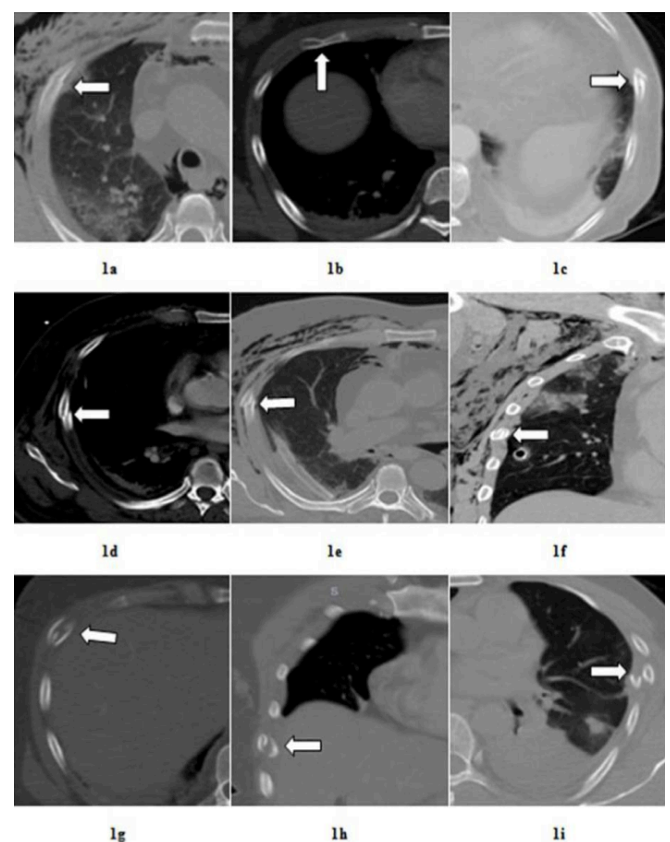


Figure 1. First visual section of the questionnaire generated from the actual tomography sections. The participants were asked to mark the tomography images that they found compatible with displaced rib fractures.

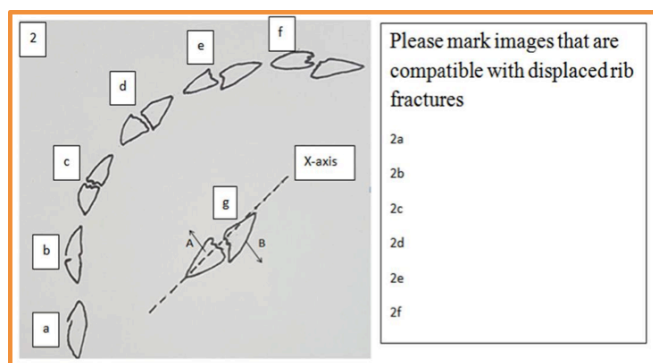


Figure 2. Questionnaire form, 2th question.

Please mark the following statements with "True" or "False"

3. If **pulmonary contusion** accompanied near the fractured rib, this evidence indicates a DRF

4. If **pulmonary laceration** accompanied near the fractured rib, this evidence indicates a DRF

5. If **traumatic bullae and blebs** accompanied near the fractured rib, this evidence indicates a DRF

Please mark the following statements with "True" or "False" by using schematic drawing 2g

6. If there is any angulation on fractured rib, this evidence is sufficient for the diagnosis of DRF

7. If the fracture line is limited on the single surface of the ribs (only A or B only surface), this fracture can not be diagnosed as DRF.

8. If the fracture line is extending from surface A to Surface B, this evidence is enough for DRF diagnosis.

9. For the diagnosis of DRF it is absolutely necessary that the fractured rib was moved on the X-axis.

10. In order to diagnose the DRF the fractured rib should be displaced at least 50% of the rib diameter on the X-axis.

Please indicate the minimum follow-up period in hours / days for the case presented below

11. A 25-year-old male patient without any additional disease / drug use was admitted with the complaint of falling. No pathological examination findings were observed except right side pain. In thoracic computed tomography, no pathological finding was found in the right 5th rib except for non-displaced rib fracture. What is the minimum follow-up period for this patient?

12. If the computed tomography of the same patient shows a displaced rib fracture, what should be the minimum follow-up period

Figure 3. Questionnaire form, 3-12 questions.

In the last part of survey, we tried to learn the minimal needed follow-up period which participants thought to be ideal for DRF and non-DRF patients. For this purpose, two imaginary cases were presented (Figure 3). In first case we presented a 25-year-old male patient with single and non-DRF in 11th question, then we presented the same case in 12th question but this time with DRF. Firstly we asked whether the patient needed be hospitalized, if they recommended, we asked the minimum requirement length of hospitalization.

For each tomography image, the opinion of each physician was recorded as DRF/non-DRF. Answers were grouped as emergency physician's, thoracic surgeon's and radiologist's answers. The same process was applied for schematic drawing questions and "true-false" section too.

The post-hoc power value was also calculated. At 5% Type 1 error level for a total of 156 patients in 3 groups; the power achieved for the effect size ($f = 0.3352$) of the question 11 score was 96.82% and for the effect size ($f = 0.3984$) of the question 12 score was 99.56%.

Statistical Analysis

Descriptive statistics are summarized as counts and percentages for categorical variables; mean and standard deviations and median (minimum and maximum) for others. Shapiro-Wilk test was used to test normality of the continuous variables. The differences between two groups in terms of categorical variables were compared by using Chi-Square test. Differences among three or more groups for non-normally distributed continuous variables were evaluated by Kruskal-Wallis variance analysis. When the p-value from the Kruskal-Wallis test statistics is statistically significant, Dunn test was used to know which group differs from which others. P value less than 0.05 was considered significant. After comparing the 11th and 12th questions between the groups with the Kruskal-Wallis test, the Post-hoc power value was calculated using the GPower 3.1.9.2 software.

Results

Of the 156 physicians who participated in the study, 56 (35.9%) were from emergency department, 54 (34.6%) from thoracic surgery, and 46 (29.5%) from radiology clinics. For the first image, 48 (85.7%) emergency physicians evaluated the rib fracture as a "non-DRF" this ratio was 51 (94.4%) for thoracic surgeons and 45 (97.8%) for radiologists. The result of the statistical analysis showed that; majority of the participants evaluated this fracture as non-DRF, they were convinced on this issue and there was no statistically significant difference between the 3 group's answers for first question ($p = 0.056$). The same analysis were repeated for each tomography image. The differences between answers were significant especially in 1c, 1g, and 1h images (Table 1). For 1c image; 47 of the emergency physicians (83.9%) were defined fracture as non-DRF, this rate was 43 (79.6%) in thoracic surgeons and only 19 (41.3%) in radiologists. The statistical analysis of the three groups proved that the responses were significantly different ($p < 0.001$). The statistical analysis for 1g and 1h images were also similar ($p = 0.001$) and ($p = 0.013$), there were not a consensus among clinics. At the end of the tomographic images, it was seen that radiologists tend to evaluate rib fractures as "displaced" between all groups.

For schematic rib fracture drawings; differences in opinions were concentrated especially in 2d. For this image, 32 (57.1%) of emergency physicians, 35 (64.8%) of chest surgeons and 42 (91.3%) of radiologists reported their opinions in favor of DRF ($p = 0.001$) (Table 1).

Table 1. Answers to questions 1 and 2, and statistical analysis results.

Q. No	Emergency physicians				Thoracic surgeons				Radiologist				p
	DRF		Non-DRF		DRF		Non-DRF		DRF		Non-DRF		
	n	%	n	%	n	%	n	%	n	%	n	%	
1a	8	14.3	48	85.7	3	5.6	51	94.4	1	2.2	45	97.8	0.056
1b	16	28.6	40	71.4	11	20.4	43	79.6	11	23.9	35	76.1	0.603
1c	9	16.1	47	83.9	11	20.4	43	79.6	27	58.7	19	41.3	<0.001
1d	51	91.1	5	8.9	49	90.7	5	9.3	43	93.5	3	6.5	0.868
1e	51	91.1	5	8.9	48	88.9	6	11.1	45	97.8	1	2.2	0.225
1f	23	41.1	33	58.9	20	37.0	34	63.0	24	52.2	22	47.8	0.294
1g	12	21.4	44	78.6	16	29.6	38	70.4	26	56.5	20	43.5	0.001
1h	46	82.1	10	17.9	42	77.8	12	22.2	45	97.8	1	2.2	0.013
1i	56	100	0	0	53	98.1	1	1.9	45	97.8	1	2.2	0.561
2a	0	0	56	100	1	1.9	53	98.1	2	4.3	44	95.7	0.282
2b	1	1.8	55	98.2	2	3.7	52	96.3	4	8.7	42	91.3	0.231
2c	6	10.7	50	89.3	11	20.4	43	79.6	9	19.6	37	80.4	0.326
2d	32	57.1	24	42.9	35	64.8	19	35.2	42	91.3	4	8.7	0.001
2e	54	96.4	2	3.6	53	98.1	1	1.9	46	100	0	0	0.425
2f	56	100	0	0	54	100	0	0	46	100	0	0	-

Abbrev.: DRF: Displaced rib fracture. Non-DRF: Non-displaced rib fracture.

For 3th question (“true-false” section) similar answers were given between both clinics ($p = 0.063$) but there was a significant difference between the 4th question’s answers. 45 (80.4%) of emergency department physicians described lung injury in the neighborhood of rib fracture as a finding in favor of DRF, this ratio was 27 (50.0%) among thoracic surgeons and 23 (50.0%) among radiologists ($p = 0.001$).

7th-10th questions were related to the definition of DRF and we obtained disagreement again. The statistical analysis result was ($p = 0.047$) for the 7th question, ($p = 0.005$) for 8th question, and ($p < 0.001$) for the 10th question (Table 2).

For patients who presented in 11th question; 31

of the emergency physicians (56.4%), 45 of thoracic surgeons (83.3%) and 18 of the radiologists (42.9%) thought that the patient should be hospitalized; other doctors declared that it was not necessary to hospitalize this patient. The results were statistically significant ($p < 0.001$). The suggested follow-up periods were also different. Median follow up period recommended by the emergency physicians was 6 (1-72) hours (95% CI: 2.71-12.13). These values were 12 (1-48) hours (95% CI: 7.46-19.20) for radiologists and 12 (2-48) hours (95% CI: 11.25-17.95) for thoracic surgeons. Differences among these groups were statistically significant ($p < 0.001$). In multi-group comparison, the answers of the emergency physicians for this question were found

Table 2. Answers to questions 3-10 and statistical analysis results.

Q. No	Emergency physicians				Thoracic surgeons				Radiologists				p
	True		False		True		False		True		False		
	n	%	n	%	n	%	n	%	n	%	n	%	
3	16	28.6	40	71.4	6	11.1	48	88.9	8	17.4	38	82.6	0.063
4	45	80.4	11	19.6	27	50	27	50	23	50	23	50	0.001
5	10	17.9	46	82.1	2	3.7	52	96.3	7	15.2	39	84.8	0.058
6	23	41.1	33	58.9	11	20.4	43	79.6	26	56.5	20	43.5	0.001
7	19	33.9	37	66.1	31	57.4	23	42.6	21	45.7	25	54.3	0.047
8	15	26.8	41	73.2	17	31.5	37	68.5	26	56.5	20	43.5	0.005
9	47	83.9	9	16.1	37	68.5	17	31.5	34	73.9	12	26.1	0.161
10	45	80.4	11	19.6	34	63.0	20	37.0	19	41.3	27	58.7	<0.001

to be significantly different from those of radiologists and thoracic surgeons (respectively $p = 0.030$ and $p < 0.001$). The answers of radiologists and thoracic surgeons did not make a significant difference ($p > 0.05$).

In the 12th question, most of the physicians [(92.7%) of emergency department physicians, (94.4%) of thoracic surgeons and (61.9%) of radiologists] declared that the patient should be hospitalized but results were different between clinics ($p = 0.001$) (Figure 4). Suggested hospitalization time were also different. Median suggested hospitalization time of emergency physicians was 6 (2-72) hours (95% CI: 11.63-20.33). These values were 24 (2-120) hours (95% CI: 15.91-38.63) for radiologists and 24 (2-120) hours (95% CI: 25.38-37.60) for thoracic surgeons. Differences among these groups were statistically significant ($p < 0.001$). In multi-group comparison, the answers of the radiologists 12th question were not found to be significantly different from those of emergency physicians and thoracic surgeons (respectively $p = 0.094$ and $p < 0.225$). The answers of emergency physicians and thoracic surgeons made a significant difference ($p < 0.01$). Average follow-up times were longer for all branches compared to the 11th question (Figure 5).

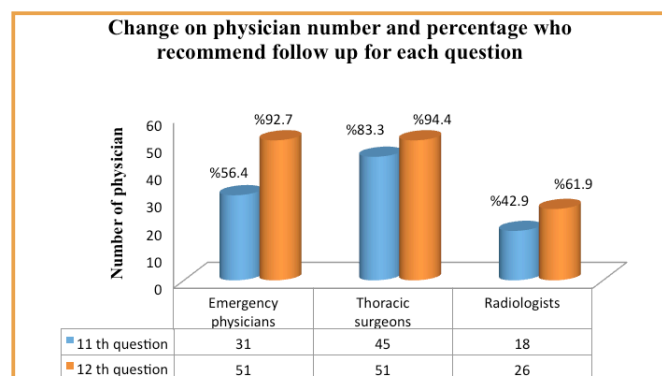


Figure 4. Change on physician number and percentage who recommend follow up for each question.

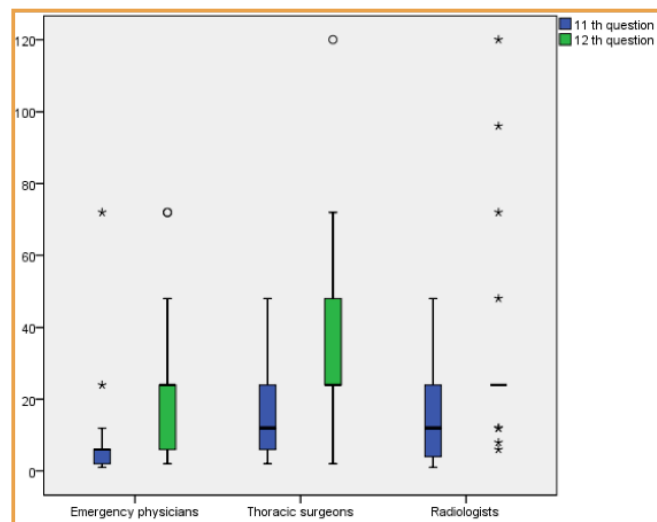


Figure 5. Recommended average follow-up times by each clinic physicians.

Discussion

Thoracic trauma is observed in 10-25% of all trauma cases and rib fracture is detected in 10% of cases with chest trauma [1-3]. As the number of fractured ribs increases, mortality and morbidity also increase. [9-12]. Similar to the number of fractured ribs, location of fracture (unilateral-bilateral), presence of flail chest, presence of displacement are also closely related with the severity of the trauma and the prognosis of the patient [1,3,13,14]. Due to these characteristics, rib fractures have started to be used in trauma scoring systems [1-3,9,13,14].

Displaced rib fractures can affect patients prognosis alone and may even cause mortality [4,5]. In patients with DRF, complications such as delayed hemothorax-pneumothorax could develop, so it is known that patients should be follow up longer than patients with non-DRF [1,4,8]. Despite these characteristics, DRF concept is perceived differently among physicians in both daily practice and also literature studies [1,3,9]. Chien et al [1] described DRF as; a displaced rib fracture was defined as a displacement distance at least half of the rib width fracture. Chapman et al [3] described severe DRF as; a severely displaced fracture was defined as displacement greater than the diameter of the rib with a total loss of contact between the proximal and the distal segment. Talbot et al [9] used these expressions for DRF definition as, when cortical disruption and a substantial abnormality in alignment are evident, a rib fracture is classified as displaced. Displacement may be minimal or obvious. Injury to the surrounding tissues and structures can occur, and several lethal complications have been documented in the literature. The starting point of our study originated from this complexity, disagreement and paradox. These different definitions in literature are frequently encountered in daily life too. The concept of DRF is often used by radiologists to point out a severe rib fracture and to alert the clinician. Again, DRF is perceived as a pathology by emergency physicians which calls for a consultation by thoracic surgeons. However, thoracic surgeons may determine same fractures as non-DRF. Similarly, while some thoracic surgeons may describe a fracture as DRF, others may describe as non-DRF. This definition confusion between clinics may cause serious problems and malpractice legal cases.

Our study reveals different opinions among the different branches and even in the same branch physicians. Question-1f is only one of these examples. While (41.1%) of emergency physicians defined fracture as DRF, (58.9%) rated as non-DRF. As same as, while (52.2%) of radiologists defined their opinions as DRF, (47.8%) rated as non-DRF. As it is seen, for the same tomography image, there is a big disagreement up to (50.0%) among the same branch physicians. Fourth question is another prominent question which exhibited great difference in terms of views among the same-clinic physicians. Participants were asked whether parenchymal laceration could be a finding in favor of DRF or not? It is known that parenchymal laceration can develop without a rib fracture, however, DRF is often accompanied by parenchymal damage. [15-17]. Nevertheless (50.0%) of thoracic surgeons stated that they found it “true” and (50.0%) found it “false”. For the same question, (50.0% true - 50.0% false) rate was observed among the radiologists too. It is a surprising result that, although no statistically significant difference was found among the different clinic doctors ($p = 0.294$), up to (50.0%) of the differences were observed among the same clinics physicians.

Our study clearly shows that there is consensus in the extreme fracture images like 1a, 1b, 1i, 2a. But there are differences of opinion in intermediate forms (1c, 1g, 1h, 2d etc.). The features of the tomography images become important at this stage. In our study, we used a series of images extending from simple linear fractures to severe displacement fractures. It was already an expected finding that participants would reached consensus on the first and last images. If more difficult images (difficult to distinguish) were used, more surprising results could have been observed.

One of the most crucial points of our study is the section of true-false questions in which the definition of displaced rib fracture is questioned. Here, the participants were asked what features must be present in a DRF. A large proportion of the participants reported that the displacement of more than (50.0%) of the thickness of the rib was alone sufficient for DRF. This finding was promising for a common diagnosis and consensus for DRF definition. This definition also corresponds with the Chien et al's [1] DRF definition and seems practical

and acceptable. At this point, the direction of the displacement is also important. In our study, mostly transverse plane sections of tomography were presented to the participants. While making a decision, displacement on sagittal or coronal plane also may be considered.

In the final stage of our study, the participants reported different opinions for the patient with a single non-DRF. There were statistically significant difference about hospitalization indication ($p = 0.002$). Average follow-up times were also different (7.4 versus 14.6 hours). The most surprising result was that for the same patient, the same clinic physicians suggested different follow-up periods ranging from 1 hour to 72 hours. Finally, our survey also revealed that; physicians do not recommend a follow-up period of more than 72 hours for a patient with single non-DRF.

When the same case was presented with DRF, recommended follow-up period increased significantly in all clinics answers. This finding is an expected result considering that important complications like delayed hemothorax can develop in patients with DRF. Therefore, they should be treated for a longer time and more carefully compared to the patients with non-DRF and this finding is also consistent with the literature [1,4,8]. For same question, the mean follow-up period of thoracic surgeons was approximately 2-fold longer than in the emergency department physicians (15.9 hours versus 31.5 hours). In additionally, the participants concluded that, 120 hours is enough for following a patient with single DRF. Although it is an assertive conclusion, it can be concluded that, if any delayed complication develop after 120 hours, doctors should not be blamed for an incomplete or inadequate treatment about follow up duration. This is one of the important points of our study. Serious mortal late complications may develop secondary to rib fractures. At this stage, if a patient dies due to late complication, it can be questioned whether the follow-up period is enough or has enough care been given to the displaced rib. It is also clear that, if a malpractice lawsuit is filed on this issue, different follow-up times will be suggested by the experts and an emergency physician or a thoracic surgeon could face an unjust charge. Our study has already demonstrated that there will be such a difference of opinion. Our study has already demonstrated these differences of opinion

which we may encounter in the future. In this context, establishing a common treatment time algorithm will be protective for all branch physicians.

Our study was multi-centered and had a good participatory population from the different clinics, but there was no international participation. Selected visual survey models are also open to criticism. Tomography images showing transition from simple fractures to severe fractures. Different surveys can be prepared by selecting more compelling or difficult images. Thus, differences of opinion may become more pronounced. We tried to overcome this problem by using an additional schematic drawing.

Another deficiency in our study is seen in the “true-false” section. Since there was no similar questionnaire study before, a new method was used in our study and the questionnaire form was created by ourselves. For this reason, some important points could not be addressed. For example the subject of flail chest, which is a strong indicator of DRF, was not questioned in our survey. We hope that our study will lead similar studies and researchers will develop their own forms. The fact that the answers were not grouped in terms of professional experience in our study is another subject open to criticism. This may be one of the reasons why there are different opinions among the same branch. While there are serious differences of opinion among assistants with less professional experience, this distinction may become more obscure among faculty or experts. However, there were not enough data collected that would demonstrate this issue in a concrete way or prove the contrary in our study. Inclusion of assistant physicians in the study is another subject open to criticism. It should not be forgotten that rib fractures that do not cause complications such as hemothorax / pneumothorax can also be followed up by general practitioners. In this respect, all assistant physicians are also general practitioners and can follow the rib fracture.

We would like to emphasize that we did not aim to investigate which clinic detected DRF more accurately in our study. Since there is not an acceptable definition for DRF, we could not make a comparison between clinic's answers as “true or false” and we think that such a comparison is not ethically appropriate. Likewise, we avoid to express opinion about which clinic suggest the

most correct and appropriate answer for follow-up time. Emergency physicians and thoracic surgeons can treat patients by different algorithms. However, it should be kept in mind that the treatment of rib fractures is basically the subject of thoracic surgery. Similarly, we do not pretend that we have a clear definition for DRF. We have not conducted a clinical study to establish which fractures should be classified as DRF. We did not get any information about which rib fractures had a worse prognosis, were associated with late complications, or had high mortality. However, we hope that, our study will inspire new studies which have clinical correlations about this subject.

In conclusion, DRF always refers to a serious rib fracture and requires additional attention however, there is no clear definition yet. There are serious differences of opinion among clinicians. It has become imperative to make a definitive definition for the benefit of all physicians dealing with trauma. Our manuscript is the first study which reveal this confusion clearly.

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Ethics approval

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Authors' contribution

ÖK; project administration, conceptualization, developed the methodology, co-wrote the paper, SA; conceived and designed the analysis, collected the data, BG; collected the data, contributed data analysis, SK; collected the data, performed the analytic calculations, TİA; validation, manuscript reviewing and editing, AÖ; formal analysis, contributed data/analysis tools, AGE; collected the data, formulated and evaluated research goals and aims, OA, BY; co-wrote the paper, collected the data, AAT; visualization, data presentation, AK; collected data, visualization, co-wrote the paper EZ; collected and processed the data, co-wrote the paper.

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Original Article

Elastofibroma dorsi prevalence in patients with primary complaint of dorsalgia

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ABSTRACT

Background: The aim of this study is to evaluate the prevalence of elastofibroma dorsi (EFD) in computed tomography (CT) imaging of the thorax in patients with dorsalgia.

Materials and Methods: Patients who are greater than or equal to 50 years old and presented to our institution with chief complaint of dorsalgia and received a thoracic CT were identified and recorded in a database together with demographic information. Control group was chosen from consecutive trauma patients in the same age group and presented to emergency room and received thoracic CT as standard of care.

Results: Out of 83 patients with common complaint dorsalgia, 9 (9/83, 10.8%) patients had no lesions and EFD was observed in 74 of the patients (74/83, 89.2%). 140 EFD including 106 (106/140, 75.7%) with a subscapular localization and 34 (34/140, 24.3%) with an infrascapular extension were identified. The maximum diameter average was 4.1 cm (range = 1-11.5 cm). Unilateral lesion was detected in 8 (8/74, 10.8%) and bilateral lesions was observed in 66 (66/74, 89.2%) of the patients. Detection of EFD in patients with isolated dorsalgia complaints was significantly higher than patients with multiple complaints (78.4% vs 21.6%, $p = 0.009$).

Conclusions: Middle aged and elderly patients with isolated dorsalgia show an unusually high EFD prevalence. Clinical suspicion and diagnostic work-up can help those cases for timely diagnosis and treatment.

Keywords: Elastofibroma dorsi, thorax CT, dorsalgia, prevalence.

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Introduction

Elastofibroma dorsi (EFD) is a rare soft tissue tumor described by Jarvi and Saxen in 1961 and is frequently observed in middle-aged and older women [1,2]. It is a benign connective tissue tumor characterized by the proliferation of fibrous and adipose tissue. EFD is typically localized in the scapulothoracic region between the thoracic wall, serratus anterior, and latissimus dorsi muscles. Although EFD may be asymptomatic, it may also cause pain, clunking, scapular snapping, palpable stiffness, and may become visible after reaching certain size, causing cosmetic complaints [3-5]. The etiology of EFD is poorly understood but microtraumas caused by friction between the chest wall and the lower pole of the scapula resulting in proliferation of fibroelastic tissue may be the primary mechanism of EFD [6,7]. The presence of EFD in up to 81% of subjects in autopsy studies may point to aging process as an etiology [8]. Genetic factors may also play a role [9]. EFD is diagnosed with physical examination and imaging findings. Magnetic resonance imaging, which has high soft-tissue sensitivity and specificity, is preferred in the diagnosis of EFD [4]. Lipoma, neurofibroma, dermoid tumors, and malignant fibrosis histiocytoma should be considered in the differential diagnosis [3,10]. Surgical treatment is considered in complaints of increasing pain, edema, scapular snapping, and when the lesion size exceeds 5 cm [6].

The symptomatology of EFD can overlap with other causes of dorsalgia which are mostly clinical diagnoses. Since the degree of musculoskeletal degeneration and the severity of back related symptoms are poorly correlated [11,12] and usually conservatively managed, there may be a patient subgroup with overlooked EFD and get suboptimal results from conservative management. Surgical treatment has been associated with good symptomatic outcomes in EFD in well selected cases [13,14].

The aim of this study is to evaluate prevalence and features of EFD in patients who receive radiological work-up due to dorsalgia.

Material and Methods

Patients aged greater than or equal to 50 years who received imaging studies as a part of their clinical work up between January 2017 and January 2019 and had “back pain” or “dorsalgia” as a recorded complaint in hospital

electronic medical records system were identified and recorded in a database. Patient data on age, gender, and presence of dorsalgia and additional complaints were gathered. A control group was formed from trauma patients presenting to emergency room during the study period, with consecutive cases in the same age group. Approval from the Institutional Review Board of Istanbul Medeniyet University Göztepe Prof. Dr. Süleyman Yalçın City Hospital (Decision number: 2019/0138) was obtained and in keeping with the policies for a retrospective review, informed consent was not required.

Typically localized, semilunar shaped, soft tissue lesions with muscle density were considered as EFD. EFDs were evaluated according to lesion localization (subscapular/infrascapular) and lesion side (unilateral/bilateral, right/left) and size.

The CT images of the patients with/without contrast were retrospectively re-evaluated by a senior radiologist for presence of EFD. GE Optima CT660 (GE Medical Systems) CT scanner was used for all the multislice chest CT imaging with a 128x0.5 mm slice collimation and a tube voltage of 120 kV, 1.5 pitch, 80 mAs. The reconstruction slice thickness was 2.5 mm, and the general reconstruction planes consisted of axial, sagittal, and coronal views. Axial images were used to provide measurements.

Statistical Analyses

Descriptive statistics are summarized as counts and percentages for categorical variables; mean and standard deviations and median (minimum and maximum) for others. The differences between two groups in terms of categorical variables were compared by using Chi-Square test. Differences between groups for normally and non-normally distributed continuous variables were evaluated by Student's t test or Mann Whitney U test, respectively. Degree of association between continuous variables were calculated by Pearson's correlation coefficient. P value less than 0.05 was considered significant.

A post-hoc power analysis was conducted to test the difference between independent groups (dorsalgia and control group) and considering EFD incidence using a total sample size of 166 participants with groups of n = 83 each and alpha of 0.05. The result shows that power was calculated at 1.0 for incidence of EFD.

Results

83 patients (44 male, 39 female) with dorsalgia were identified and included in the study. The mean age was 62.7 ± 8.9 (50-82) years. 22 patients had other complaints (chest pain, cough, epigastric pain, sternal pain, dysphagia, history of smoking, history of known malignancy, known lung nodule, fever) accompanying dorsalgia.

EFD was detected in 74 of 83 patients in our study (89.1%). Gender distribution in EFD patients was comparable (38 male (51.4%), 36 female (48.6%) (F/M: 1.05)) and mean age was 63 ± 9.1 (50-82) (mean male age 65.7 ± 8.8 and mean female age 60.1 ± 8.6 , $p = 0.953$). 49 patients (66.2%) were under 65 years old, 25 patients (25/74, 33.8%) were 65 years and older (Figures 1,2).

66 patients (89.2%) had bilateral lesions, 8 patients had unilateral EFD (5 right, 3 left). No significant difference was found between genders in the distribution of unilateral-bilateral lesions ($p = 0.712$). The mean age of patients with bilateral and unilateral lesions were also comparable (63.1 ± 9.1 (50-82) vs 62 ± 7.6 (53-74), $p = 0.477$) (Table 1).

140 EFDs were detected in 74 patients. 106 EFDs had subscapular localization and the rest ($n = 34$) had infrascapular extension as well. In 9 of 66 patients with bilateral EFD, lesions had differences in localization, while the rest ($n = 57$) were symmetrical (bilateral subscapular in 45 patients, bilateral infrascapular in 12 patients). 71 of the 140 lesions in the study group were

located on the right and 69 on the left. Localizations of right and left sided EFDs were equivalent (16 vs 18 for left and right sided lesions with infrascapular extension, respectively, $p = 0.460$).

The average longest diameter 4.1 ± 2 cm (1-11.5). Males and females had comparable lesion sizes (40.7 ± 21.4 mm vs 52.4 ± 21.7 mm, $p = 0.428$). Right and left sided lesions were of similar size (43.6 ± 21 mm vs 41.3 ± 18.8 mm, $p = 0.305$). Bilateral EFDs were significantly greater than the unilateral lesions (48.1 ± 21.8 mm (10-115 mm) vs 23.9 ± 5.8 mm (15-83 mm) ($p = 0.009$)) (Table 1). No correlation was found between age and maximum diameter, diameter with right-sided lesions, and diameter with left-sided lesions ($p = 0.180$ $r = 0.137$; $p = 0.078$ $r = 0.211$; $p = 0.249$ $r = 0.083$, respectively).

No lesions were detected in 9 patients. Detection of EFD in patients with isolated dorsalgia complaints was significantly higher than patients with other complaints in addition to dorsalgia ($p = 0.009$).

Control group had 83 patients with comparable sex distribution (49 male, 34 female) but was significantly older with a mean age of 70.2 years (± 11.6) ($p < 0.01$). 12 patients were identified as having EFD (significantly lower incidence, $p < 0.0001$) with comparable laterality (9 bilateral, 3 unilateral). The maximum diameters were also comparable (42.1 ± 17.6 mm for right, 49.5 ± 27.9 mm for left). 8 EFDs were subscapular and 13 EFDs had infrascapular extension.



Figure 1. A soft tissue lesion compatible with subscapular elastofibroma dorsi showing a slight extension to the infrascapular area in axial (a), coronal (b), and sagittal (c) thorax CT sections of a 66-year-old female patient with dorsalgia.

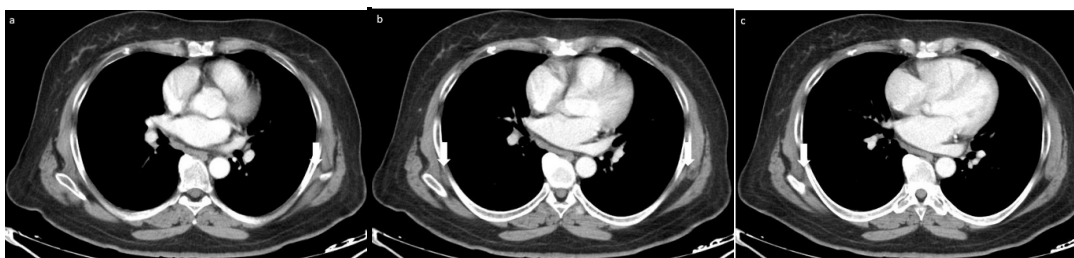


Figure 2. Consecutive axial CT images of bilateral small EFDs of a 50-year-old female patient with dorsalgia.

Table 1. Distribution of patient EFD characteristics according to laterality.

	Unilateral (n=8)	Bilateral (n=66)	p
Age (year $\pm$ SD) (min-max)	62 $\pm$ 7.6 (53-74)	63.1 $\pm$ 9.1 (50-82)	0.477
Sex (F/M)	3/5	33/33	0.712
Localization (subscapular/infrascapular)	7/1	99/33	0.679
Diameter (mm $\pm$ SD)	23.9 $\pm$ 5.8 (15-83)	48.1 $\pm$ 21.8 (10-115)	0.009

Discussion

EFD is relatively rare and under-diagnosed mass due to its slow growth and variable symptomatology [7, 15-17]. Brandser et al reported 2% prevalence in CT imaging of 258 asymptomatic patients over age of 60 [18]. In a more recent study, the prevalence of patients over 18 years of age was 2.73% in the group of asymptomatic patients who underwent CT for another reason; and they showed that the prevalence increased significantly in patients over 65 years old [19]. An autopsy study done on 235 cases, the prevalence of EFD was 16% in patients over 55 years of age with male dominance [16].

In this study, the patient population consisted of adults over the age of 50 with dorsalgia and prevalence of EFD is much higher than articles reporting on asymptomatic populations [18,19]. While dorsalgia may be a common symptom that can be due to a wide variety of causes, the possibility of EFD should be kept in mind, especially if dorsalgia is an isolated symptom at the start of the work up. Study group showed dramatically increased prevalence of EFD when compared to the control group, despite the latter having more advanced mean age.

Many authors believe that the diagnosis of EFD can be made clinically and radiologically [20] and biopsy is generally not necessary for exclusion of malignancy. Minarro et al compared ultrasonography, CT and MRI for the diagnosis of EFD. CT was found to have a better correlation than MRI for evaluation of EFD size [21] but other clinical/radiological features are deemed to be more important. EFD is a tumor with a slow growth course; however, in cases with rapid growth, high contrast enhancement, and atypical localization, lipoma, hemangioma, and soft tissue tumors should be included in the differential diagnosis [3,10]. Therefore, in cases of dorsalgia with high probability of a soft tissue problem, MRI rather than CT may be more preferable, decreasing the radiation exposure. For palpable lesions or follow up of an accessible lesion, ultrasonography can be the study of choice [21].

Many studies report conflicting results regarding laterality in EFD involvement [10,19,22-24]. Also EFD is usually presented as a male predominant disease

[13,16,19,21,25]. Our study population (≥ 50 y/o, with dorsalgia), contrary to the most of the reports showed predominantly bilateral involvement and equivalent gender distribution. This may be due to the fact that most of the studies are done on either asymptomatic populations or populations with multiple complaints [13,16,19].

Surgical excision of small and asymptomatic EFDs is not recommended. Although EFD is often an asymptomatic lesion [26,27] at the beginning, it can slowly reach to large sizes over time and may present with back pain and shoulder pain, edema, stiffness leading to cosmetic problems, and clunking of the scapula [4,7,28]. Significant regression of pain and functional improvement has been noted with complete resection in the literature [13,14,29]. The possibility of EFD should be kept in mind in the clinical work-up of dorsalgia in middle-aged and older population as surgical treatment may be indicated.

While relatively small sample size and retrospective nature of the study are the main limitations, we were able to present a target patient population with relatively high EFD prevalence in CT images. None of the patients included in this study have histopathological verification of EFD via either surgical excision or biopsies. While some patients prefer surgical excision instead of follow up and symptom management, diagnostic accuracy of radiological imaging in EFD seems to be reasonably high for conservative approach.

In conclusion, in this study the high prevalence of EFD (89.2%) in patients with dorsalgia indicates the need for careful evaluation of the scapulothoracic region in terms of EFD by radiologists and clinicians, especially in middle-aged and elderly patients with isolated dorsalgia, aiding timely diagnosis/treatment and eliminating the need for any additional further examination. Patients with isolated dorsalgia not improving on or do not want to follow up with medical treatment might benefit from re-evaluation of previous imaging or usage of MRI to assess this surgically correctable problem.

Declaration of conflicting interests

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Ethics approval

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Authors' contributions

ZNT; design, concept, planning, data collection, data analysis, manuscript writing, supervision, editing, ZB;

manuscript writing, data collection, supervision, editing, OA; design, planning, conduct, data collection, data analysis, editing, FC; design, materials, data analysis, conduct, manuscript writing, EK; design, concept, planning, conduct, supervision, editing.

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Original Article

A multivariate analysis of cigarette smoking in thoracic surgery patients

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ABSTRACT

Background: Cigarette smoke (CS) is a long-term risk factor for several disorders of the respiratory and cardiovascular systems. In this study, we aimed to analyze the current effects of cigarette smoke on patients who underwent thoracic surgery. In line with this purpose, we analyzed patients who were admitted to a thoracic surgery clinic recently and their smoking history.

Materials and Methods: We reviewed patients who underwent thoracic surgery (n = 867) and were admitted to our clinic between January 2018 and December 2018. The patients were categorized into seven groups comprising the most common types of thoracic pathology as, lung carcinoma (n = 338), pneumothorax (n = 219), pleural effusion (n = 131), mediastinal lymphadenopathy (n = 77), pneumomediastinum (n = 22), bronchiectasis (n = 30), and hydatid cyst (n = 10). Cases were reviewed in detail for demographics, smoking history in terms of pack-years, thoracic pathology, and hospitalization duration (days).

Results: Statistical analysis revealed that smoking is still a significant factor contributing to lung cancer. Cigarette smoke did not appear to be a factor in mediastinal lymphadenopathy, bronchiectasis, pleural effusion or hydatid cyst cases. Additionally, hospitalization duration was significantly longer in the smoker group.

Conclusions: The data reviewed are compatible with the current knowledge on the mechanisms of cigarette smoke-related pathologies. Cigarette smoke is still associated with common thoracic surgery pathologies such as lung carcinoma, pneumothorax, and pneumomediastinum. This may be rephrased thus, the absence of cigarette smoke would greatly decrease the prevalence of thoracic surgery cases.

Keywords: cigarette smoking, lung neoplasms, pneumothorax, pleural effusion, lymphadenopathy, mediastinum, COPD, thoracic surgery

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Introduction

Cigarette smoke (CS) is a hazardous mixture of multiple chemical compounds that cause a variety of pulmonary and systemic effects in humans [1]. CS is a long-term risk factor for several disorders of the respiratory and cardiovascular systems. Tobacco is consumed in the form of CS by nearly one-fifth of the world's population [2].

Smoking is the most significant cause of chronic respiratory bronchitis, pulmonary emphysema, and bronchial carcinoma [3]. In addition, studies have revealed that cigarette smoking is the major cause of both obstructive pulmonary diseases and chronic respiratory symptoms such as cough, wheezing, and dyspnea. CS is the main known cause of cancer-related deaths worldwide [4].

Chest radiography studies demonstrated that pulmonary micronodules and lobular opacities were mostly due to changes induced by pathological CS substrates [5]. Moreover, CS-related *in vitro* studies revealed induction of DNA strand breaks, formation of DNA adducts, and mutagenic cytogenetic effects [6].

In this study, we aimed to analyze the current effects of CS on patients who underwent thoracic surgery. In line with this purpose, we analyzed patients who were admitted to a thoracic surgery clinic recently and their smoking history.

Material and Methods

This retrospective study was approved by the local ethics committee (2020-64, December 2020). All participants provided informed consent prior to their inclusion in the study. We reviewed patients who underwent thoracic surgery ($n = 867$) and were admitted to our clinic between January and December 2018. We excluded patients ($n = 40$) who were exposed to toxic substances, or had pectus deformities, traumatic hemothorax, rib fracture, and traumatic pneumothorax. Subsequently, the remaining 827 patients were included in the study.

The patients were categorized into seven groups comprising the most common types of thoracic pathology: lung carcinoma ($n = 338$), pneumothorax ($n = 219$), pleural effusion ($n = 131$), mediastinal lymphadenopathy ($n = 77$), pneumomediastinum ($n = 22$), bronchiectasis ($n = 30$), and hydatid cyst ($n = 10$). Lung cancer patients with mediastinal lymphadenopathy or pleural effusion were included in the lung cancer group. We analyzed the studied cases in two groups depending on their classification as smokers or non-smokers.

Cases were reviewed in detail for demographics, smoking history in terms of pack-years, thoracic pathology, and hospitalization duration (days). Twenty cigarettes smoked every day for one year was referred to as "a pack-year." Lymph node enlargement was defined as a size of more than 7-11 mm on the short axis depending on the regional nodal station [7].

Statistical Analysis

All statistical analyses were performed using SPSS version 21.0 software (SPSS, Inc., Chicago, IL, USA). Descriptive statistics are summarized as counts and percentages for categorical variables; mean and standard deviations and median (minimum and maximum) for others. The differences between two groups in terms of categorical variables were compared by using Chi-Square test or Fisher's exact test, where applicable. Differences between two groups for non-normally distributed continuous variables were evaluated by Mann Whitney U test. P value less than 0.05 was considered significant.

Results

Our retrospective study recruited 827 patients, including 669 men (80.1%) and 158 women (19.1%). The mean age was 48.8 ± 16.7 (range 16-90) years, and the mean hospitalization duration was 3.9 ± 5.8 (range 1-26) days. Of the 827 patients, 554 (67.1%) underwent surgery due to related thoracic pathology. The smoker group comprised 72% ($n = 595$) of the cohort. The mean value of consumption was 24.6 ± 14.3 (range 3-90) pack-years in the smoker group.

Of the 827 patients who were referred to our thoracic surgery clinic, 40.9% had lung cancer, among whom 79% were smokers ($n = 267$); 26.5% had pneumothorax, among whom 75.8% were smokers ($n = 166$); 15.8% had pleural effusion, among whom 67.1% were smokers ($n = 88$); 9.3% had mediastinal lymphadenopathy, among whom 46.7% were smokers ($n = 36$); 2.6% had pneumomediastinum, among whom 86.3% were smokers ($n = 19$); 3.6% had bronchiectasis, among whom 56.7% were smokers ($n = 17$), and 1.2% had hydatid cyst, among whom 20% were smokers ($n = 2$). The characteristics and comparative analysis of the smokers and non-smokers are shown in table 1.

Table 1. Characteristics of the studied groups.

Parameter	Smoker (n=595)	Non-smoker (n=232)	P
Gender			
Male	481 (80.8%)	188 (81.1%)	0.949 ^c
Female	114 (19.2%)	44 (19%)	
Age (years)	49.3±16.9	48.6±18.2	0.56 ^m
Smoking (pack year)	-	24.6±14.3	-
Hospitalization (days)	2.4±5.1	4.6±5.7	0.001 ^m
Pathology			
Lung cancer	267 (44.5%)	71 (30.6%)	<0.001 ^c
Pneumothorax	166 (27.9%)	53 (22.8%)	0.139 ^c
Pleural effusion	88 (14.7%)	43 (18.5%)	0.185 ^c
Mediastinal LAP	36 (6.1%)	41 (17.7%)	<0.001 ^c
Pneumomedi- astinum	19 (3.2%)	3 (1.3%)	0.127 ^c
Bronchiectasis	17 (2.9%)	13 (5.6%)	0.058 ^c
Hydatid cyst	2 (0.3%)	8 (3.4%)	0.001 ^f

c; chi-square test, m; Mann-Whitney U test, f; Fischer's exact test

The parameters were studied to predict the possibility of CS as a factor in related thoracic pathology. Statistical analysis revealed that smoking is a factor contributing to lung cancer, pneumothorax, and pneumomediastinum while it was only significant for lung cancer (Table 1). CS did not appear to be a factor in mediastinal lymphadenopathy, pleural effusion, bronchiectasis, or hydatid cyst cases. Additionally, hospitalization duration was significantly longer in the smoker group (4.6 ± 5.7 , range 1-26 days) than in the non-smoker group (2.4 ± 5.1 , range 1-24 days) ($p = 0.001$).

Discussion

Cigarette smoke causes apoptotic cell death and aging, and damages repair functions performed by lung fibroblasts, which are essential for maintaining the integrity of the alveolar structure by producing extracellular matrix proteins [8]. CS inhibits fibroblast proliferation and migration, and induces DNA damage [9]. CS-induced inhibition of fibroblast activity contributes to the reduction of lung repair following lung injury [10]. In our study, we aimed to demonstrate the current effects of a commonly known toxic substance; CS, in thoracic surgery patients. We mainly analyzed patients with the seven most common groups of thoracic surgery pathologies.

CS is the most important cause of lung cancer [4].

Carcinoma arises from genetic alterations that cause the release of neoplastic cells from homeostatic mechanisms that govern cell proliferation [11]. After the discovery of the significant role of the tumor suppressor gene p53 mutations in human cancer, many studies have revealed a high prevalence of p53 mutations in many types of cancers that were linked to CS exposure. Almost half of all lung cancer cases have been reported to present p53 mutations. Furthermore, the prevalence of mutations in the p53 gene, were higher in lung tumors from smokers as compared to those from non-smokers [12]. In addition, mutations in the KRAS gene occur in approximately one-third of lung cancer patients with adenocarcinoma histology, and these mutations have been found to be associated with smoking [13]. CS has been a known cause of lung carcinoma for decades and continues to be the leading cause of lung carcinoma. In our study, statistical analysis revealed that smoking still had a significant effect on lung carcinoma.

Pneumothorax remains one of the most common pathologies requiring thoracic surgery. As a result of the rupture of subpleural blebs or bullae, primary spontaneous pneumothorax (PSP) typically occurs mostly in tall and young male smokers, with no apparent co-existing lung diseases. The risk of PSP is linked to CS and increases with duration of exposure and daily consumption. CS stimulates bronchiolar remodeling by inducing growth factors in the distal airway wall via increased release of active transforming growth factors [14]. CS induces pathologic changes in the small distal airways, which contributes to the development of local emphysema followed by the consequent formation of bullae. The trapping of air distally due to CS-induced bronchiolar inflammation and respiratory bronchiolitis leading to alveolar overdistension, causes bullae formation and possible rupture [15]. However, the pathophysiologic mechanism of PSP formation due to CS and its clinical correlation are not strictly demonstrated. Controversy still exists around the occurrence of bullae and PSP. In our study, statistical analysis revealed that smoking had an effect on pneumothorax but it was not significant.

CS interferes with macrophage response to infection by decreasing its phagocytic ability [16]. Thus, smokers tend to contract primary tuberculosis and pneumonia. Patients with latent tuberculosis may develop active disease, leading to increased disease burden in the community due to CS. Smoking is associated with a higher incidence of parapneumonic effusion in patients with community-acquired pneumonia and higher pleural in-

flammatory markers [17]. In our study, the most common causes of pleural effusion were tuberculosis and pneumonia-related pleuritis due to the exclusion of malignant and cardiac pleural effusion. We excluded these cases because elaboration of the CS effect on these patients was not possible due to the apparent cause of pleural effusion. Statistical analysis revealed that smoking did not have a significant effect on pleural effusion.

Enlarged lymph nodes are mostly known as a sign of malignancy, although they are also observed in benign conditions such as sarcoidosis, chronic heart failure, obstructive pulmonary disease, and pneumoconiosis. In addition to exposure to various substances, the deposition of CS itself also results in enlargement of the hilar and mediastinal lymph nodes of smokers. Recent studies have demonstrated that benign enlargement of mediastinal lymph nodes is a common finding in heavy smokers and is more often identified in smokers than in non-smokers. Thus, CS itself is hypothesized to be a cause of enlargement of mediastinal lymph nodes [18]. Nevertheless, in our study, statistical analysis revealed that smoking did not have a significant effect on mediastinal lymphadenopathy.

Spontaneous pneumomediastinum (SPM) is mostly a self-limiting pathology that clinically presents as the presence of free air in the mediastinum without any traumatic cause. SPM is usually observed in young adults with the most common clinical symptoms, including shortness of breath, chest and neck pain, difficulty in swallowing, and swelling of the face and neck. The most common precipitating factor of SPM is bronchial asthma, with alveolar rupture being the main etiology. Other causes include barotrauma including the Valsalva maneuver, and inhalation of various substances such as CS, cocaine, and marijuana. Alveolar rupture leads to air leakage to the broncho-vascular sheath, which consequently results in the release of air that reaches the mediastinum [19]. CS has been well reported in the literature as a risk factor for SPM. In our study, statistical analysis revealed that smoking had an effect on SPM but it was not significant.

Bronchiectasis is a pathological, nonreversible enlargement of the bronchial airways. The diagnosis of this condition is made on the basis of visual identification of an airway whose diameter is greater than that of the adjacent artery on computed tomography (CT) scans or lung tissue [20]. Studies suggest that bronchiolitis is the initial lesion in bronchiectasis. The presence and se-

verity of airflow obstruction, correlated with the intensity of mosaic attenuation detected on high-resolution CT scans, cause bronchiectasis. CS-related chronic bronchitis and bronchiectasis share many common pathological findings. There are three well-known causes of chronic bronchitis: cystic fibrosis and primary ciliary dyskinesia, which are genetically determined, and CS [21]. All three conditions compromise the mucociliary transport apparatus and may lead to bronchiectasis. However, in our study, statistical analysis revealed that smoking did not have an effect on bronchiectasis.

Hydatid cyst disease is a parasitic infection caused by *Echinococcus granulosus*, characterized by cystic lesions in the liver and lungs. In the available literature, there is no established relationship between hydatid cyst of the lung and tobacco exposure. Statistical analysis revealed that smoking had no effect on hydatid cysts in our study.

Lung fibroblasts produce extracellular matrix proteins, including collagen, elastin, and fibronectin, which are required for the attachment, structure, and function of alveolar epithelial cells; thus, they are considered mandatory for the maintenance of the integrity of alveolar structure. CS causes a malfunction in the body's repair mechanism by vitiating fibroblast activity [22]. In our study, we found that smokers also had significantly longer hospital stay compared to non-smokers due to longer recovery time.

In our study, nonsmoker patients' ratio to smoker patients was higher compared to literature. In terms of probable factors, subtle exposure to asbestos which is common in our country or living in limestone houses especially in Anatolian region of the country may be agents of thoracic pathology in nonsmoker patients.

Limitations of the Study

Our study was a retrospective, single-center clinical study. It utilized one year's worth of collected data, which consisted of a limited number of thoracic surgery cases. Our study recapitulates the effect of CS on the studied pathologies using up-to-date data; however, it does not contribute additional knowledge to existing literature.

In conclusion, CS initiates damage by causing respiratory bronchiolitis and disrupting the homeostatic mechanisms governing cell proliferation, followed by parenchymal damage. The data reviewed are compatible with the current knowledge on the mechanisms of CS-related pathologies. CS is still associated with common thoracic surgery pathologies such as lung carci-

noma, pneumothorax, and pneumomediastinum. This may be rephrased thus: the absence of CS would greatly decrease the prevalence of thoracic surgery cases.

Declaration of conflicting interests

The authors declared no conflicts of interest with respect to the authorship and/or publication of this article.

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Ethics approval

Approval for this retrospective, descriptive, single-center study was obtained from Istanbul Yedikule Chest Disease and Thoracic Surgery Training and Research Center Ethical Committee (2020-64, December 2020).

Authors' contribution

YS; designed the analysis, wrote the paper, OVY; collected data, performed the analysis, MÖ; collected data, NM; contributed data, LC, MAB; editing.

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Case Report

Giant pediatric intrathoracic ganglioneuroma: a case report and review of the literature

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ABSTRACT

Ganglioneuroma is an extremely rare (incidence 1/1,000,000) well-differentiated neurogenic tumor that mostly originates from neural crest cells. The average age of diagnosis is 7 years old and it is mostly located in the posterior mediastinum (37.5%). Here we present a 10-year-old girl with a giant thoracic ganglioneuroma. The tumor was resected via posterolateral thoracotomy. With our case report included, there are 10 cases of surgically treated pediatric giant intrathoracic ganglioneuroma.

Key words: giant, ganglioneuroma, pediatric, thoracic surgery, posterior mediastinal tumors

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Introduction

Ganglioneuroma is an extremely rare (incidence 1/1,000,000) well-differentiated neurogenic tumor that mostly originates from neural crest cells. It is mostly seen in infants and children and the M/F ratio is 2/3. It can be detected incidentally or it can be diagnosed by symptoms as a result of compression on surrounding tissues or catecholamine release. It is generally located in the posterior mediastinum (37.5%) and retroperitoneally. The general treatment approach is total resection of the tumor, and postoperative local recurrence is rare [1]. In this article, we present the 10th case of surgical resected giant pediatric intrathoracic ganglioneuroma in the literature.

Case Report

A 10-year-old girl was admitted to the hospital with

shortness of breath and chest tightness. A uniform large opacity was detected in the upper zone of the left hemithorax at the PA chest x-ray (Figure 1a). Thorax computed tomography (CT) of the patient revealed a well-circumscribed homogenous hypodense 15x8x7 cm (CCxTXAP) mass with millimetric calcifications located in left posterior mediastinum which causes minimal scoliosis in the T5 and T6 vertebral bodies, and minimal erosion in the left 6th rib (Figure 1b). Thoracic MRI showed a T1W hypointense, T2W hyperintense mass that did not show invasion to the mediastinum, lungs, and adjacent thoracic vertebrae (Figure 1c). In PET-CT SUVmax 2.8 involvement was observed in the mass.

Adamkiewicz artery has been shown to arise from the left T9 level in Thorax CT angiography (Figure 2).

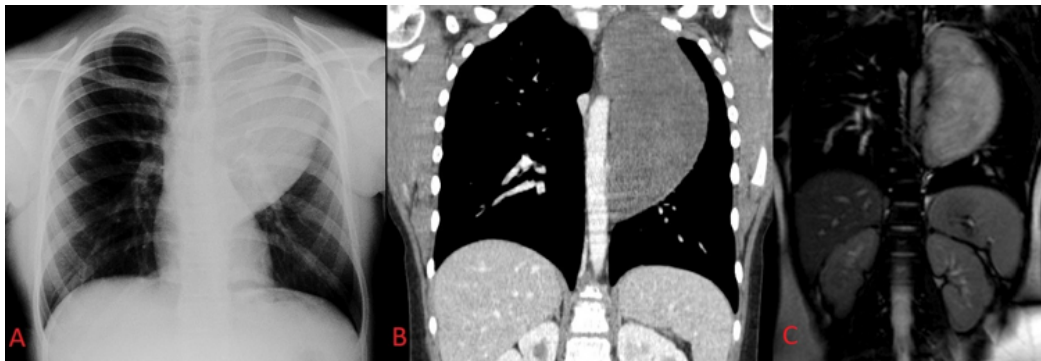


Figure 1. Posteroanterior chest x-ray showing a radioopacity at the upper zone of the left hemithorax (A), coronal reformatted image of enhanced thorax CT demonstrates a well-circumscribed homogeneous hypodense mass lesion (B), coronal T2-weighted MRI reveals the homogenous hyperintense left paravertebral mass lesion (C).

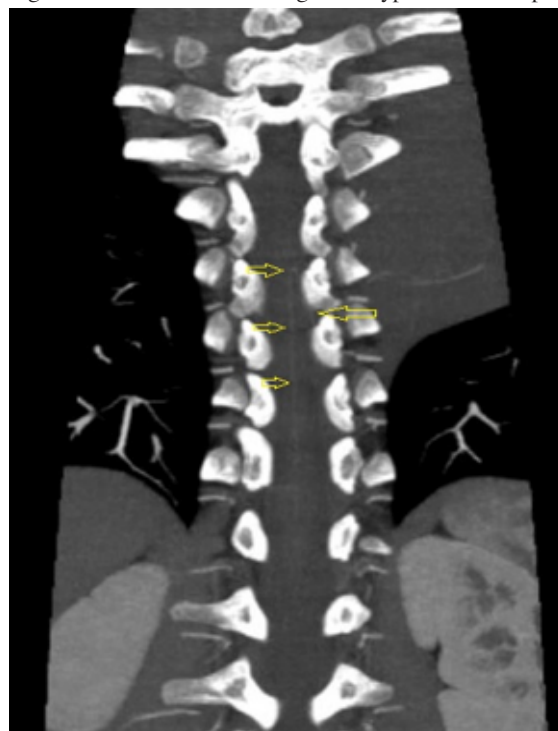


Figure 2. Longitudinal course of the Adamkiewicz artery (arrows) can be seen on maximum intensity projection (MIP) reformatted coronal thorax CT angiography.

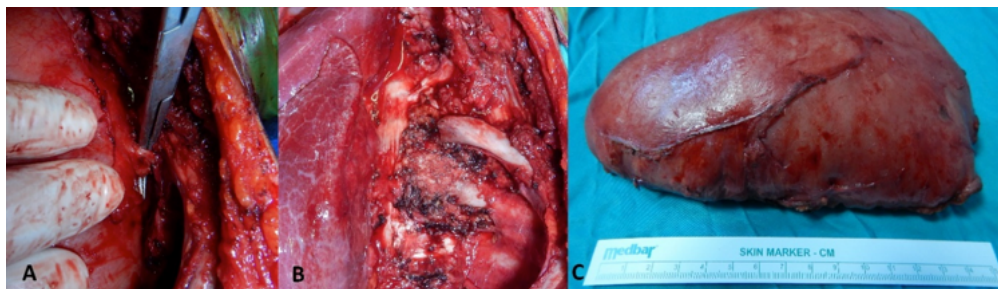


Figure 3. Intraoperative view of the mass and dissection of the intercostal nerve bundles going into the mass (A) , postoperative view of the thoracic cavity (B) and lateral view of the specimen (C).

Normetanephrine and homovanillic acid levels were found to be elevated in the 24-hour urine analysis (1106.7 $\mu\text{g/day}$ and 9.2 $\mu\text{g/day}$, respectively). When the radiological and clinical results were evaluated together, the patient was thought to have a neurogenic tumor, and resection via posterolateral thoracotomy was planned. Making sure that the Adamkiewicz artery was below the dissection site, the posterior mediastinal mass was excised with partial resection of the left 6th rib (Figures 3a-c). The histopathological analysis revealed mature ganglion cells with large eosinophilic cytoplasm dispersed in the schwannian stroma and the pathology result was reported as mature ganglioneuroma with negative surgical margins (Figure 4). She was discharged on postoperative 5th day. No additional treatment was planned for the patient. Patient is asymptomatic during 96-months follow-up period. Written informed consent was obtained from the parents for publication of her data.

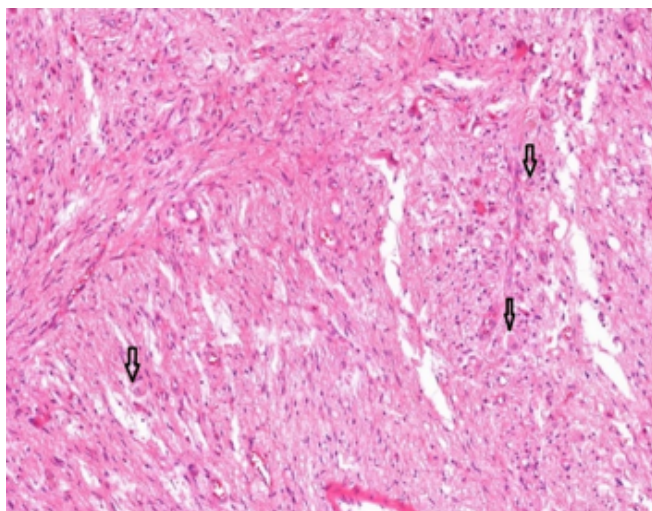


Figure 4. The histopathological analysis revealed mature ganglion cells with large eosinophilic cytoplasm (marked by arrowheads) dispersed in the schwannian stroma (HEx400).

Discussion

Ganglioneuroma is a benign neurogenic tumor that mostly originates from the sympathetic ganglion consisting of ganglion cells and stroma containing Schwann cells [2]. The average age of diagnosis is 7 years old, but it is likely to occur in older ages. Patients are usually asymptomatic at admission and tumoral tissue is detected incidentally [2]. However, cough, chest pain, shortness of breath due to compression of lungs may be present as in our case [3]. Diarrhea, sweating, and hypertension may occur due to the release of catecholamine and metabolites from the tumor. The rate of diagnosing ganglioneuroma under the age of 20 is 60%. It is mostly located in the posterior mediastinum but it can be seen in different parts of the body, especially retroperitoneum, adrenal glands or head and neck region and gastrointestinal tract. Mediastinal ganglioneuroma on imaging should be differentiated from cystic teratoma, neurilemmoma, lymphangioma cysticum or bronchocele [1]. When the tumor location is paraspinal, it can cause long-term compression-related scoliosis [2]. Although the etiology is not fully known, genetic diseases such as neurofibromatosis type 1-2 and multiple endocrine neoplasia (MEN) type 2 may be present in patients who have a family history. There are 10 cases of operated pediatric intrathoracic ganglioneuroma larger than 10 cm with the present case. The mean age of the patients is 9.7 years (5-17 years). Seven (64%) of the patients were female. The average follow-up time was 19.6 months, and none of them had recurrence. Five of all patients had left sided tumors and accompanying scoliosis needed correction surgery [1-6]. Only one patient had subtotal resection and only one patient [2] had serious bleeding as an intraoperative complication [6]. The list of publications operated for pediatric intrathoracic ganglioneuromas larger than 10 cm in diameter are summarized in table 1.

Table 1. List of publications presenting operated pediatric intrathoracic ganglioneuromas larger than 10 cm in diameter.

Year	A	S	Symptoms	Dm (cm)	Site	Treatment	Comp	Relapse	Follow-up (m)
2005 ^[5]	12	F	Scoliosis	10x9, Spinal canal extension	R	R TR and correction surgery (Two stage surgery)	None	None	24
2007 ^[4]	9	M	Scoliosis	10x9	L	L TR and correction surgery (Two stage surgery)	None	None	12
	14	F	Scoliosis	13.5x9, 11x7 Spinal canal extension	R	R TR and correction surgery (Two stage surgery)	None	None	30
2017 ^[2]	12	F	Chest tightness and lower extremity weakness	12x12 Spinal canal extension	L	LM and L TR (Subtotal resection)	None	No progression	12
2017 ^[1]	5	M	Chest and upper abdomen pain	16x14	R	R TR	None	None	1
2017 ^[9]	12	F	Shortness of breath	18x10	R	R TR	None	None	24
2019 ^[8]	8	M	Upper respiratory tract infection	10x8	L	L TR	None	None	1
2020 ^[6]	8	F	Chest and upper abdominal pain	23x20	R	R TR	Bleeding (intraoperative embolisation)	Unknown	10 days
2020 ^[3]	17	F	Scoliosis	20x15, Spinal canal extension	L	Correction surgery and L TR (One stage surgery)	None	None	24
2021 (present case)	10	F	Dyspnea and chest pain	15x8	L	L TR	None	None	48

Abbrev.: A: age, S: sex, Dm: diameter, F: female, M: male, TR: thoracotomy, LM: laminectomy, Comp: complication, m:month, R: right, L: left

Giant ganglioneuroma is observed in cross-sectional images as a mass that compresses the surrounding tissues or surrounds them but does not invade them. It is usually observed as a homogeneous, and more rarely heterogeneous hypodense mass on CT examinations. It may contain calcifications. It shows weak to moderate or no enhancement in the early arterial phase and late venous phases [7]. In MRI, it is predominantly hypointense in T1W sequences, and mostly hyperintense in T2W sequences [1]. Low FDG uptake is expected to be seen in PET-CT [7] and biopsy is usually not required for diagnosis as in our case. Another important role of preoperative

radiological imaging in thoracic ganglioneuroma cases is to reveal the origin and course of the Adamkiewicz artery. The Adamkiewicz artery occurs on the left side in 80% of the cases, between vertebrae T7 and L4. However, the anatomy frequently differs. If this large anterior radiculomedullary artery is damaged during surgery, neurological deficits may occur due to spinal ischemia-infarction [8]. In the present case, the origin of the Adamkiewicz artery was determined by CT angiography and it was reported to be far from the tumor origin.

Ganglioneuroma originates from the neural crest and is known to be hormonally inactive, although it

has neuroendocrine properties. It has been reported that catecholamine and metabolite release rate is 39% in ganglioneuroma, and an elevation of metanephrine, catecholamine, vasoactive intestinal peptide, dopamine, cortisol, homovanilic acid, or vanillylmandelic acid can be detected in urine or blood in those especially with adrenal gland localization [2]. The gold standard for hormonal activity in catecholamine secreting tumors is an increase in the level of metanephrine in the blood. In the present case, the levels of homovanilic acid and metanephrine in the urine were high but the catecholamine and metabolite values in the blood were in a normal range [6]. Thoracotomy or minimally invasive surgical approach can be applied in cases of intrathoracic ganglioneuroma, considering the size and location of the tumor. However, it has been reported in the literature that some ganglioneuroma cases have extension into the spinal canal and laminectomy with a posterior approach was also performed for complete resection. Scoliosis surgery can be performed to fix the thoracic vertebra angle that deteriorates with mass compression [4-9]. In the thoracotomy approach, the relation of the tumor with the surrounding tissues is better explored and intraoperative complications can be managed more safely [1]. Thoracoscopic resection can also be performed in smaller tumors. Malignant transformation of ganglioneuroma is rare. The recurrence rate is low and no additional postoperative treatment is required after complete resection [5].

In conclusion, ganglioneuroma should be considered in the differential diagnosis of giant posterior mediastinal tumors with smooth margins and low SUVmax values in pediatric patients. These tumors can be successfully resected with thoracotomy and can be cured without any additional treatment.

Declaration of conflicting interests

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Authors' contributions

BMKB, GK, MAS, BMY, AGÇ, BAK, AH, SE: conceived and designed the current case report, co-wrote

the paper, collected the clinical data. The authors discussed the case under the literature data together and constituted the final manuscript.

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Case Report

The surgical repair of a trauma-related sternoclavicular joint dislocation using the musculus gracilis tendon

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ABSTRACT

Sternoclavicular joint (SCJ) dislocations are rare, and these dislocations may be traumatic or atraumatic. In this study, we will examine the treatment management of a patient whose SCJ was anteriorly dislocated following a traffic accident. We performed open stabilization for SCJ dislocation using the musculus gracilis tendon. Although the first treatment option in acute cases is closed reduction and stabilization, we recommend surgical repair because the probability of recurrence is high. The gracilis tendon is a good option for joint stabilization.

Keywords: sternoclavicular joint, trauma, accidents, injuries, joints, surgical procedures

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Introduction

Dislocations due to trauma can be anterior or posterior, with the former being approximately nine times more common than the latter. In patients who have conditions known to cause collagen deficiency (such as Ehlers-Danlos syndrome, generalized hypermobility syndrome, infection, or arthritis) atraumatic dislocations may occur [1]. In this study, we will examine and discuss the treatment management of a patient whose SCJ was anteriorly dislocated.

Case Report

A 70-year-old male patient was hospitalized after a traffic accident and presented at hospital with a displaced rib fracture in the right 3rd, 5th, and 6th ribs anteriorly, a non-displaced rib fracture in the left 5th, 6th, 7th, and 8th ribs anteriorly, and a left SCJ dislocation. On physical examination, there was an anterior swelling at the right sternoclavicular junction and tenderness on palpation of the anterior thoracic wall. The patient's chest x-ray is seen in figure 1.

SCJ dislocation was evident in the thorax CT scan (Figure 2).

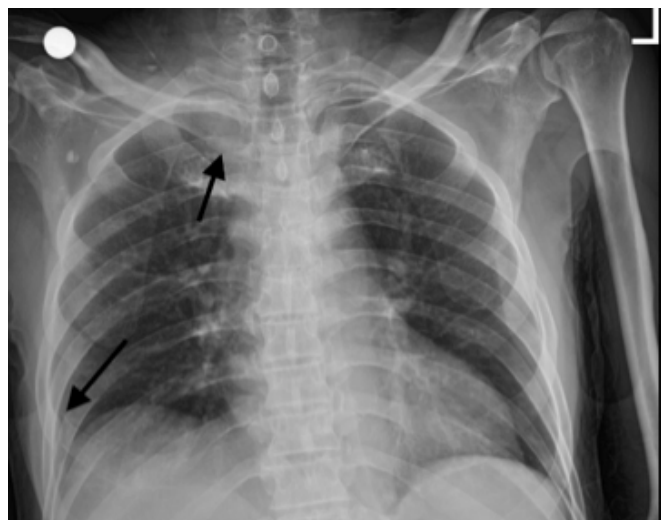


Figure 1. PA x-ray shows rib fracture and sternoclavicular joint dislocation.



Figure 2. Thorax CT scan showing the SCJ dislocation.

The dislocation spontaneously reduced during the first days while the patient was lying down and protruded forward when the patient stood. When offered an operation, the patient declined. Hemothorax and pneumothorax did not develop before the follow-up, and the patient was discharged with a prescription. However, two weeks later, the patient requested surgery because his right arm was very painful and had reduced range of movement, and he felt discomfort due to swelling at the SCJ site. Upon the patient's second admission to hospital, the clavicle head was not reduced, and we operated the patient together with orthopedics. An approximately 8 cm incision was made anterior to the right sternoclavicular joint and it was freed from the surrounding soft tissue. The articular surfaces of the clavicle and sternum were beaten with a rongeur to expose the subchondral bone. An intramedullary canal was drilled on both the clavicular and sternal articular surfaces, two from the sides, and one intramedullary canal. The gracilis tendon graft was then passed through the holes drilled into the clavicle and sternum in the 8 shape, and the joint was reduced and fixed (Figures 3,4). The patient was discharged without any complications on the second post-operative day. Figure 5 shows the technique of passing the gracilis tendon through the holes drilled into the sternum and clavicle. Written informed consent was obtained from the patient for publication of his data.

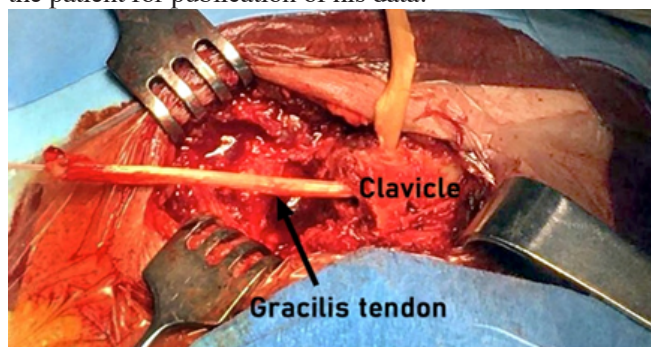


Figure 3. The passage of the gracilis tendon through the holes drilled in the clavicle is shown.

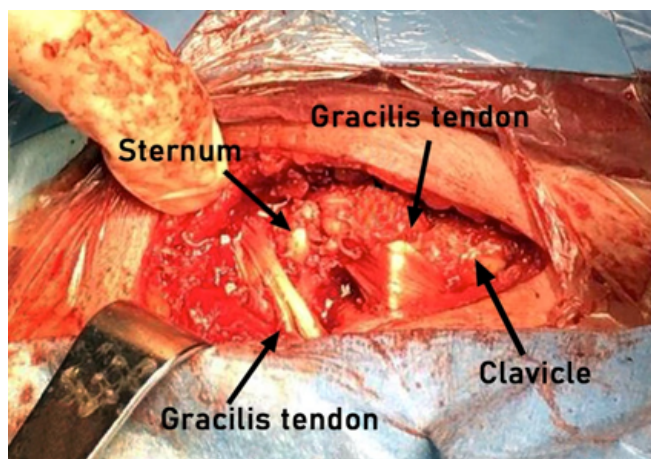


Figure 4. The sternoclavicular joint is fixed with the gracilis tendon.

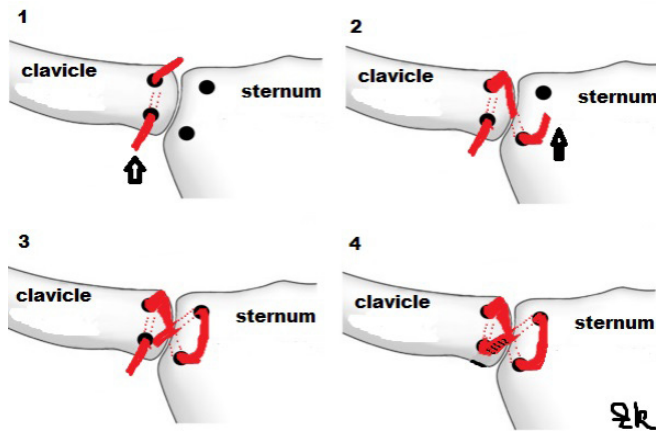


Figure 5. This illustration shows the technique of passing the gracilis tendon through the holes drilled into the sternum and clavicle.

Discussion

SCJ is an inherently unstable diarthrodial synovial joint. Less than 50% of the medial clavicular surface comes into contact with the corresponding articular surface on the manubrium sterni. Therefore, joint stability is achieved through internal and external ligament structures. In addition, the subclavius muscle rises from the first rib just lateral to the costoclavicular ligament and attaches to the lower surface of the clavicle. It is believed that this muscle performs a protective function in terms of SCJ stability by reducing the rate of upward displacement of the clavicle against the pressure from the sides [2]. Anterior dislocation of the SCJ is usually due to the lateral compressive force to which the shoulder is exposed, which results in the preservation of the posterior capsule, although rupture of the anterior capsule and often the costoclavicular ligament can occur [2,3]. Those with anterior dislocation of the SCJ will complain of a painful lump just lateral to the sternum, as with our patient. In patients who present with localized swelling in this area, care should be taken to determine whether they have a dislocation or have a medial clavicle fracture [3]. On examination, patients show swelling at the level of the SCJ and a decrease in the range of movement of the upper extremities. They may also present with new-onset paresthesia of the upper extremity and/or weakened pulses or signs of venous occlusion. In our patient, there was swelling at the level of the SCJ and a decrease in the range of movement of the upper extremity.

Dislocations of the SCJ are very difficult to detect on plain radiographs, but pneumothorax, pneumomediastinum, or hemopneumothorax can sometimes be seen in posterior dislocations [2]. CT

imaging has superior image resolution and allows 3D reconstruction to determine the exact position of the SCJ [4]. MRI is superior to CT in assessing ligament injury and the condition of other soft tissue behind the SCJ. If an intimal tear is suspected in the subclavian artery, CT angiography may be necessary [2].

If the patient presents with acute anterior dislocation in his SCJ (within 7–10 days of onset), a closed reduction can be performed in the operating room with either sedation or under general anesthesia. The patient is placed in the supine position and a pillow is situated between the shoulders. Traction is then applied to the affected upper extremity at 90° of abduction and flexion, and pressure is directly applied to the medial clavicle. After reduction, the patient uses an arm sling for up to four weeks. After this procedure, re-dislocations occur between 21% and 100%, which raises the question of whether simple closed reduction without ligament reconstruction is sufficient [5-7]. We also suspect that closed reduction alone will not suffice due to the arthrodial nature of the joint.

If closed reduction is not possible or if symptomatic instability of the SCJ persists, numerous surgical techniques have been described in literature and there is no evidence that one method is superior to another. Martínez et al described the stabilization technique that was used in our case, using the 8-shaped gracilis tendon and passing it through the holes in the manubrium and clavicle [8]. The use of autologous tendon grafts is preferred because it has a high healing potential and facilitates long-term tissue integration. Different tendon graft options are available for sternoclavicular joint reconstruction. However, the length and diameter of the gracilis tendon are ideal for transverse stable reconstruction. In addition, the morbidity rate in the postoperative knee joint region is low [9].

Conversely, Booth et al and Bae et al used a strip of the sternocleidomastoid muscle to separate it from its sternal attachment, pass it under the first rib and back through a drill hole in the collarbone, and then tie it back on itself. This way, they effectively reconstructed the costoclavicular ligament [10,11]. Abiddin et al performed capsule repair by passing sutures through the holes in the medial clavicle and manubrium [12]. Franck et al used a plate [13], but due to the spongy structure of the sternum and the vital structures behind the joint, we elected not to do this. Kocsis et al used a technique

to stabilize the SCJ by restructuring the costoclavicular ligament using a braided polyester mesh and found that this mesh also locally stimulated fibroblast activation. They achieved good functional results in all patients and found that patient satisfaction was high [14].

In conclusion, SCJ dislocation should be treated both cosmetically and functionally because of symptoms such as pain. Although the first treatment option in acute cases is closed reduction and stabilization, we recommend surgical repair because the probability of recurrence is high. The gracilis tendon is a good option for joint stabilization.

Declaration of conflicting interests

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Authors' contributions

SK, FD, SK; conceived and designed the current case report, co-wrote the paper, collected the clinical data. The authors discussed the case under the literature data together and constituted the final manuscript.

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Case Report

A rare cause of acute airway obstruction: tracheal malignant melanoma

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ABSTRACT

Tracheal tumors are quite rare, with an incidence of 2.6 per 1,000,000 persons. Tracheal malignant melanomas are one of the least common types among these tumors due to the lack of natural epithelial tissue of melanocytes. There is no optimal treatment method for tracheal malignant melanomas because they have an aggressive nature, have limited life expectancy and occur very rarely. This paper discusses the case of and treatment methods for a patient who was initially suspected to have a foreign body aspiration because of presenting to hospital with acute airway obstruction.

Keywords: airway obstruction, tracheal malignant melanoma, tracheal tumor

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Introduction

Tracheal tumors are a very rare form of cancer that occurs at a rate of 2.6 per 1,000,000 persons. Within this group of tumors, which mostly consists of squamous cell carcinomas and adenoid cystic carcinomas, malignant melanomas are rare and the least known type [1].

Malignant melanomas constitute only 1% of the primary tracheal cancers even in the most comprehensive studies on primary tracheal tumors in the literature [1].

Depending on the limited number of studies, there is only limited information, if any, regarding the formation mechanisms of primary tracheal malignant melanomas, their clinical course, optimal treatment methods, and their life expectancy, presented only as case reports for a limited number of patients.

Case Report

A 38-year-old female patient with no known history of disease presented to the emergency department of an external center due to the shortness of breath that had increasingly continued for 1 month. She was referred to our hospital in July 2019 because of suspected foreign body aspiration due to the presence of respiratory acidosis (PaCO₂: 79 mmHg) and prominent stridor in the arterial blood gas examined. The patient was found to experience tachypnea and prominent expiratory stridor in the examination and her blood gas PaCO₂ level was 50.9 (35-45) mmHg.

The thoracic tomography report stated an appearance of a possible foreign body in the proximal trachea that obstructed the lumen almost completely (Figure 1a). The patient was immediately taken for the video rigid bronchoscopy procedure. A solitary lesion was observed in the trachea approximately 4 cm beyond the vocal cords, obstructing the lumen almost completely (Figure 1b). The lesion was dissected and removed from the rigid bronchoscope. During the procedure, it was determined that the lesion extended to the carina and originated from the trachea posterior wall. No massive bleeding was observed in the patient, who underwent debulking. However, cold saline solution was required from time to time. The total volume of the extracted material was 3 cubic centimeters.

In the control process, the PaCO₂ value (38.6

mmHg) of the blood gas of the patient, who was taken to the postoperative intensive care unit, regressed to the normal range. The patient's stridor and complaint of shortness of breath regressed.

PET-CT was requested for the patient, who did not experience hemoptysis during the follow-up. The PET-CT imaging showed a residual lesion in the trachea, located about 2 cm proximal to the carina, and 9x8 mm in size with a SUV max value of 2.7 (Figure 1c). In addition, there were metastatic (SUV max: 4.9) nodules in both lungs, the largest of which was 15x9 mm.

Medical treatment was planned for the patient, who had been diagnosed with tracheal malignant melanoma, as a result of the pathology. It was learned that the patient had been diagnosed with melanoma by skin biopsy 3 years ago but had not received any treatment. The patient began to be treated with Dabrafenib + Trametinib in September 2019.

Five months after the diagnosis, the patient presented to the hospital again due to the shortness of breath and hemoptysis and was intubated and hospitalized in the intensive care unit. The patient underwent video rigid bronchoscopy again. A lesion was detected in the distal part of the trachea, approximately 2 cm from the carina. The lesion narrowing the trachea by 70% was initially cauterized with electrocautery. Then, debulking was carried out with rigid bronchoscopy. The lesion was removed in two parts, the first of which measured 1.7x0.7x0.6 cm and the second of which measured 0.9x0.8x0.7 cm. After debulking, a fiberoptic bronchoscope was passed through a rigid bronchoscope, and cryotherapy was applied under the lesion. The patient, who was intubated postoperatively, was extubated on the third day after the intervention.

Radiotherapy (30x216cGy, totally 6480cGy) was applied to the patient, who was discharged after her clinical course had improved, in March 2020. Upon the patient's epileptic seizure in the 10th month after diagnosis, a cranial MRI was taken, which showed metastasis in the left parietal area (Fig 1d). During the examinations, progression of lung metastases and multiple metastases in the liver were also detected. The patient died 11 months after the diagnosis.

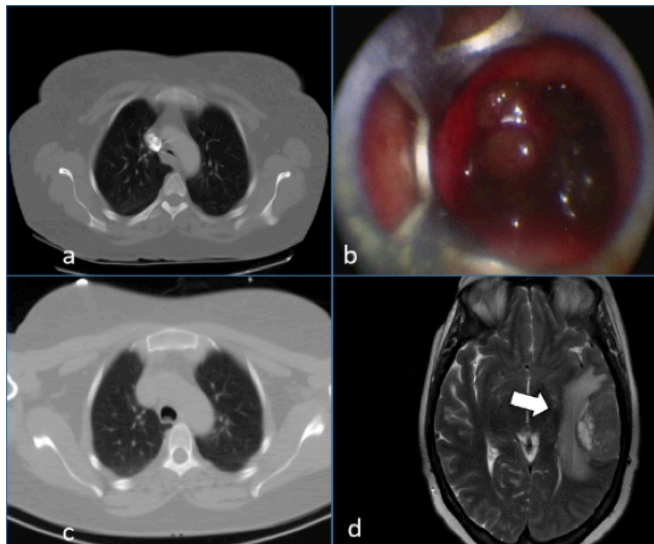


Figure 1. The first evaluation; a photo of a CT scan showing an almost totally obstructed trachea by a solitary lesion (a), rigid bronchoscopy picture showing a solitary lesion approximately 4 cm ahead of the trachea and vocal cords, before the intervention (b), a picture of the remaining residual tumor inside the trachea after the bronchoscopic intervention (c), 10 months after the diagnosis of tracheal malignant melanoma, a cranial MR photo of the patient admitted with epileptic seizure, showing a metastatic mass at the left parietal lobe (d).

Discussion

Tracheal malignant melanomas are cancers that occur sporadically even among primary tracheal cancers [2]. There is no study in the literature involving patient series.

One of the main reasons why they are so rare is that melanocytes are not the natural epithelium of the respiratory tract. Although there is no certain information on how melanocytes reach this area, there are several theories. These involve the migration of melanocytes during embryogenesis, the transformation of respiratory cells into melanocytes and the differentiation of neuroendocrine cells into melanocytes [3].

As in tracheal cancers, the clinical course can include cough, shortness of breath, and hemoptysis [1,2]. Since they are not included in the group of diseases that are suspected first by clinicians during diagnosis as they occur so rarely, patients in this group can also present to the hospital with airway obstruction, as was the case with our patient. Chest radiography and computed tomography can be used in the first step and to detect the tumor localization. PET-CT can be helpful in diagnosis and treatment planning and to indicate the presence of distant metastases [1,2].

Although malignant melanoma can be diagnosed histopathologically by S-100, melan-A, and HBM-45,

it may not be distinguished from primary metastasis. Therefore, diagnostic criteria have been developed to distinguish true primary tracheal malignant melanomas from metastases. These criteria include i) solitary mass presence, ii) invasion from the epithelia to the submucosa iii) absence of a previously detected or excised skin lesions in the patient, iv) absence of a family history of skin disease v) provision of typical histopathological confirmation by electron microscopy or immunohistochemistry, and vi) ‘dropping off’ of melanoma cells together with junctional changes in the mucosa [4]. It was not considered primary because the patient had a previous skin biopsy.

There is no data on the survival time of patients with tracheal malignant melanomas. Considering the studies known in the literature including the one with the largest series, the 5-year survival rate of patients with primary tracheal cancers is reported to be between 15-27% [1,2]. Considering that malignant melanomas have the shortest known doubling time with 48 days, with the exception of anaplastic thyroid cancer and testicular cancer, the survival of patients is expected to be much shorter [5].

Except for one patient who underwent tracheal resection, almost all tracheal malignant melanoma patients in the literature have been reported to die shortly after diagnosis [6].

Since tracheal malignant melanomas are very rare and aggressive in nature, there is no consensus on an optimal treatment method. There are studies reporting that resection and end-to-end anastomosis prolong survival of all patients with no metastasis in primary tracheal cancers regardless of the histopathological type [1,7]. However, a study conducted with the largest series of patients in the literature reported that resection could be applied to only 14 of 308 patients and that surgery might not be effective upon life expectancy [2]. Tracheal reconstruction techniques, and debulking surgery, as we have done with our patient, have a place in the treatment because of the necessity of providing airway patency [1-2].

A recent study showed that bronchoscopic interventions of secondary pulmonary cancer may increase the average life expectancy of the patients. The life expectancy of this group of patients after the diagnosis was 10 ± 13.1 months. However, after the bronchoscopic interventions positive effects on life expectancy were seen. Also, in the same study, the mortality rate due to complications of the bronchoscopic interventions was found

to be 0%. In our case, we performed two bronchoscopic interventions with no major complications to be mentioned. We are convinced that in these types of patients, bronchoscopic interventions can be applied safely [8].

In addition, although radiotherapy has a place in local treatment, its effectiveness on life expectancy has not been demonstrated. Chemotherapy and immunotherapy are used in the treatment of malignant melanoma. Dacarbazine-based treatment protocols are preferred in chemotherapy whereas Vemurafenib (2011), Dabrafenib (2013) and Trametinib (2013) are used in immunotherapy. Programmed death-1 checkpoint (PD-1) inhibitors such as Pembrolizumab are also used in the treatment of malignant melanoma. Furthermore, in the literature we found information about using Pembrolizumab as a medical treatment choice which may be used during pregnancy [9].

When we examined the literature, we found a case similar to ours. In that case, after the patient was diagnosed with malignant melanoma he was treated with interferon. Seven years later the patient developed lung metastasis. At the follow-up stage, in a 3 weeks period, a malignant melanoma causing 90% obstruction of the patient's airway was detected. Similar to our case the patient was treated with rigid bronchoscopy [10].

Especially after the first bronchoscopic intervention, and as a different approach, a stent or a course of radiotherapy could be applied. In our case, the short life expectancy of the patient and the low complication risk of the rigid bronchoscopic intervention were the reasons behind choosing this approach.

As a result, although there are no definitive data on this extremely rare type of tracheal cancer, resection and end-to-end anastomosis seem to be good options, if applicable. In order to provide airway patency in inoperable patients, debulking surgery can be performed safely without causing massive bleeding, as we applied to our patient twice.

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Case Report

Surgical treatment of an intrathoracic rib causing cough in a young woman

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ABSTRACT

Intrathoracic rib is mostly asymptomatic and one of the rare congenital anomalies of thoracic wall. Keeping in mind its appearance and types on chest x-rays can prevent unnecessary examinations and invasive procedures. Locations and relationship to adjacent tissues of intrathoracic ribs determine its clinical course and surgical decision.

Keywords: accessory rib, anatomical variations, intrathoracic rib, rib anomalies, supernumerary rib

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Introduction

The term intrathoracic rib refers to the abnormal position of a rib within the thoracic cavity. It was first described by Lutz in 1947. Later, it was also used with the definition of accessory rib or supernumerary rib [1]. Although the etiology is not clear, genetic factors and chemical effects in the fetal period have been held responsible [2]. This rare congenital abnormality mostly has no effect on respiratory functions and usually detected incidentally by imaging studies. We report a case of supernumerary intrathoracic rib (SIR) located posterior to the abnormally developed T6 vertebrae in an 18-year old female who presented with dyspnea and back pain.

Case Report

An eighteen year-old female patient admitted to our clinic with the complaint of back pain, cough and dyspnea for several years. It was understood that her complaints were unrelated to effort and intensified especially in the evening. She has celiac disease. Vital signs were normal and stable. Oxygen saturation was %98 in room air. There was no history of operation, chest trauma or thoracic disease. There were no pathological findings in the physical examination findings and auscultation of respiratory sounds. Laboratory blood test results are within the normal range. On the conventional chest radiography, an opacity extending from the thoracic vertebra to the diaphragm was observed behind the cardiac shadow. For further examination, CT scan of thorax was performed, 3D and multiplanar reconstruction images were created (Figure 1a). These images revealed an abnormal osseous structure originating from the left lateral corpus of the T6 vertebrae which was protruded infero-laterally into the thoracic cavity (Figures 1b-1d). There was no intraparenchymal lesion. No other skeletal alteration was observed. We performed a postero-lateral muscle sparing thoracotomy incision. The accessory rib had no articulation with vertebrae, covered by the parietal pleura and extended towards the diaphragmatic crus (Figure 2). There were no adhesions with the lung parenchyma. The accessory rib was resected completely by cutting from the vertebral corpus using a rib cutter. It was observed that SIR did not have its own intercostal muscle, artery, vein or nerve. The symptoms regressed after the operation, there was only pain at the incision site. There was no air leakage in the chest tube after the operation. The patient did not describe dyspnea, cough and back pain during the hospitalization period and was discharged on the postoperative day 2. There were no complications during the one year follow up. Written informed consent was obtained from the patient for publication of her data.

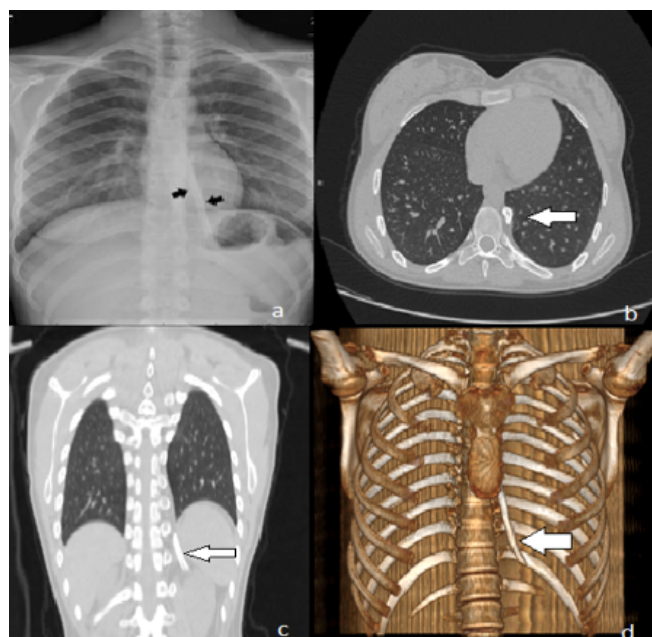


Figure 1. Posteroanterior chest graph shows an accessory rib (arrows) located on the left lateral side of corpus of T6 vertebrae (a), lung window CT image shows an abnormal thoracic rib (arrow) (b), coronal window CT image shows an abnormal thoracic rib (arrow) (c), three-dimensional CT reconstruction image of the bone shows an abnormal supernumerary intrathoracic rib (arrow) (d).

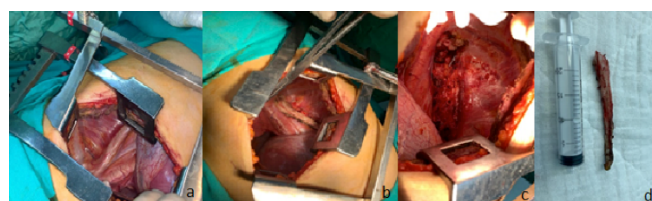


Figure 2. Intraoperative view of the accessory rib (a), releasing the accessory rib from surrounding tissues by stripping the overlying pleura (b), view of the thoracic wall after separation of the accessory rib from the vertebral body with a rib cutter (c), image of the accessory rib that has excised (d).

Discussion

Intrathoracic rib is a congenital anomaly that occurs with abnormal fusion of sclerotomes in embryological life [1,2]. When more than fifty cases in the literature are analyzed, it has been determined that it is more common on the right hemithorax without gender dominance. Most of the cases were diagnosed in pediatric age. SIR is mostly single and unilateral. Its location and relationship to adjacent tissues determines its clinical course, but it is usually asymptomatic. SIR is usually detected incidentally by imaging studies or in autopsy series. The most common symptoms are chest pain, hemoptysis, and dyspnea [1,2]. Etiological factors of accessory rib is

not clear. It is suggested that it is due to the incomplete sclerotome fusion that caused by the change in intrathoracic pressure during embryologic development. Alterations of Myf5/Myf6 expression, the Pax-1 gene, and the Hox gene family have been implicated [3]. The intrathoracic rib is important in differential diagnosis. In the differential diagnosis intrathoracic rib, accessory lobe, asbestos plaques and calcified sequestration should be kept in mind. This unusual opacity detected on chest X-ray can cause concern and unnecessary further examination. Therefore, the benign course in asymptomatic cases prevents over-diagnose and over-treatment. Being aware of its benign course prevents unnecessary examinations and operations. In 2006, Kamaro et al classified accessory ribs according to their origin [1]. Accordingly, Type 1 refers to the supernumerary intrathoracic rib that is articulated from the vertebral corpus or its proximal rib. Type 2 bifid intrathoracic rib. In type 3, the rib is locally compressed into the cavity. In 2021, Xue et al proposed a new classification with five distinct categories that include all anomalies not included in the previous classification (Table 1) [2].

Table 1. The classification of intrathoracic rib [2].

Type 1	Separated from the vertebral corpus by a joint
1A	Originating from the lateral side of the vertebral corpus (right or left)
2B	Originating from the anterior of the vertebral corpus (unilateral or bilateral)
Type 2	Originating from the proximal rib
Type 3	Bifid or forked intrathoracic rib
Type 4	Ribs locally compressed into the cavity
Type 5	Floating intrathoracic rib (without any rib or osseous connection to the vertebrae)

According to this current and useful classification, our case is in category type 1-A. Sometimes this intrathoracic rib can be attached to the diaphragm with a fibrotic band or fatty tissue. Intrathoracic rib is mostly common with scoliosis [2]. Intrathoracic rib can be easily overlooked, especially if the image is not taken under optimal conditions in the chest X-ray. It can be hidden in cardiac shadow or hidden behind the clavicle, especially at the upper levels. Barreiro et al. analyzed the tomography images of 650 patients and found the incidence of accessory thoracic ribs to be 0.15% [4]. CT scan is a steady method to

show intrathoracic ribs. Also, with CT, parenchymal diseases and diaphragmatic extensions can be observed and relations with neighboring organs can be determined. In our case, three-dimensional reconstructions and multiplanar CT were very supportive for a successful operation. The newly defined contrast-enhanced ultrasound (CEUS) also provides reliable information (in) for the identification of masses in the subpleural area [5]. Chronic cough is an indication for bronchoscopy. However, we did not perform bronchoscopy because tracheobronchial system pathologies was not considered in our patient. The cough complaint vanished in the patient who completed the recovery period after the surgery. This may suggest that the supernumerary rib is the cause of the irritation. It is necessary to evaluate thoroughly before deciding on surgery. All possible complications of the intrathoracic rib according to localization and the patient's symptoms and possible complications of surgery should be well evaluated in the decision of surgery. The doctor's decision should be shared with the patient and the final decision should be made with the patient's participation. It has been suggested in the past that surgery does not improve respiratory functions and may have additional secondary negative effects [5]. However, in the study of Cuyan et al the symptoms disappeared after surgery, as in our case [6]. We think that asymptomatic patients may become symptomatic or the existing mild symptoms may be exacerbated, especially in physiological conditions such as pregnancy that may cause elevation of the diaphragm.

In conclusion, the intrathoracic rib should be kept in mind in the differential diagnosis of opacities in chest X-rays. Although it is a benign anomaly, localization and the symptoms of SIR determines decision of the rib resection.

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Authors' contributions

DK,MŞİ; conceived and designed the study, HI,MŞİ; reviewed and recorded the data, DK,MŞİ; wrote the paper, SG,ES; reviewed and edited the manuscript. All authors read and approved the manuscript.

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while the process above is carried out. The investigation described above will be completed even if the authors withdraw their paper, and the responses below will still be considered. In the case of allegations against reviewers or editors, they will be replaced in the review process while the matter is investigated. For further information visit "Useful links".

Data Sharing Policy

Current Thoracic Surgery requires authors to submit a data-sharing statement form and register a data-sharing plan when registering a clinical trial. The ICMJE's data sharing statement policy asks authors to submit a data-sharing statement for randomized clinical trials. The ICMJE's policy regarding trial registration is explained at (<http://www.icmje.org/icmje-recommendations.pdf>).

Accordingly, the data sharing statements must indicate the following:

whether individual deidentified participant data (including data dictionaries) will be shared,

what data in particular will be shared,

whether additional, related documents will be available (e.g., study protocol, statistical analysis plan, etc.),

when the data will become available and for how long,

by what access criteria data will be shared (including with whom, for what types of analyses, and by what mechanism).

Authors must provide a data-sharing statement at submission to indicate whether data will be shared according to the statements written above.

Please visit <http://www.icmje.org/icmje-recommendations.pdf> that provides illustrative examples of data sharing statements that would meet these requirements.

Authors of secondary analyses using shared data must attest that their use was in accordance with the terms (if any) agreed to upon their receipt. They must also reference the source of the data using its unique, persistent identifier to provide appropriate credit to those who generated it and allow searching for the studies it has supported. Authors of secondary analyses must explain completely how theirs differ from previous analyses. In addition, those who generate and then share clinical trial data sets deserve substantial credit for their efforts. Those using data collected by others should seek collaboration with those who collected the data. As collaboration will not always be possible, practical, or desired, the efforts of those who generated the data must be recognized.

Statistical Knowledge

The statistical method used should be reported in a way that a reader whoever could reach the original data could understand the results. The terms used, abbreviations, symbols, the pc programme used and the statistical method should all be defined.

When reporting the results, especially when giving means and percentages, two digits should be given after the comma (112.20 or 112.21 instead of 112.2). P, t, Z values are exceptional and three digits should be given after the comma ($p = 0.001$, instead of $p < 0.05$).

Submission Process

Briefly, except whole numbers, two digits after the comma should be given, but for statistical values like p,t,z,F,Chi-square, three digits after the comma should be given. When presenting p values, the exact p value together with the test statistics should be given, except for the case when this value is smaller than one percent or one per thousand (this time $p < 0.01$ or $p < 0.001$, respectively should be given).

Preparation of Manuscript

Cover Letter

The Current Thoracic Surgery requires authors to disclose in the “cover letter” any commercial association (eg, employment, direct payments, stock holdings, retainers, consultantship, patent licensing arrangements, or honoraria) that might pose a conflict of interest issue concerning the manuscript. All funding sources supporting the work should be acknowledged in a footnote. Also authors should mention that none of the material in the manuscript has been published previously, and that none of this material is currently under consideration for publication elsewhere. This includes symposia, transactions, books, articles published by invitation, and preliminary publications of any kind except an abstract of 400 words or fewer.

Style and Format

Manuscripts should be double-spaced with 3-cm margins on all sides of the page, in Times New Roman font. Every page of the manuscript, including the title page, references, tables, etc., should be numbered. All copies of the manuscript should also have line numbers starting with 1 on each consecutive page.

Symbols, Units and Abbreviations

In general, the journal follows the conventions of Scientific Style and Format, The CSE Manual for Authors, Editors, and Publishers, Council of Science Editors, Reston, VA, USA (7th ed.). If symbols such as $\times$, μ , η , or v are used, they should be added using the Symbols menu of Word. Degree symbols ($^{\circ}$) must be used from the Symbol menu, not superscripted letter o or number 0. Multiplication symbols must be used ($\times$), not the letter x. Spaces must be inserted between numbers and units (e.g., 3 kg) and between numbers and mathematical symbols (+, -, $\times$, =, <, >), but not between numbers and percent symbols (e.g., 45%). Please use SI units. All abbreviations and acronyms should be defined at first mention. Latin terms such as *et al.*, *in vitro*, or *in situ* should not be italicized.

Manuscript Content

Research articles should be divided into the following sections. Principal sections should be Introduction, Materials and Methods, Results, Discussion, Acknowledgements and References sections. Please see below for information about other types of manuscripts.

Title and Contact Information

The first page should contain the full title in sentence case, the full names (last names fully capitalized) and affiliations (in English) of all authors (University, Faculty, Department, City, Country), and the contact address and e-mail for the clearly identified corresponding author.

Abstract

An informative abstract of not more than 250 words must accompany each manuscript. The abstract must be structured to include the study's background, materials and methods, results, and conclusions and key words under 5 separate headings. Abstracts of review articles should be a brief overview of the main points from the review.

Keywords

Please provide at least 3 key words or phrases to enable retrieval and indexing. Acronyms should be avoided.

Always try to use terms from the Medical Subjects Headings (MeSH) list from PubMed <https://www.ncbi.nlm.nih.gov/pubmed>.

Introduction

This should argue the case for your study, outlining only essential background, and should not include the findings or the conclusions. It should not be a review of the subject area, but should finish with a clear statement of the question being addressed.

Materials and Methods

Explain clearly but concisely your clinical, technical, or experimental procedures. Previously published papers related to the methods should be cited with appropriate references. Brand names and company locations should be supplied for all mentioned equipment, instruments, chemicals, etc.

Results

Findings must be described without comment. The same data or information given in a Table must not be repeated in a Figure and vice versa. It is not acceptable to repeat extensively the numbers from Tables in the text or to give lengthy explanations of Tables or Figures.

Discussion

Statements from the Introduction and Results sections should not be repeated here. The final paragraph should highlight the main conclusions of the study. Names of funding organizations should be written in full.

References

In the text, references should be identified using numbers in square brackets in which they appear consecutively. The titles of journals should be abbreviated according to the style used in Index Medicus. A general guide limit the number of references to 25 for original articles, to 6 for case reports and to 85 for reviews. For articles with 1 to 6 authors, list all authors. For articles more than 6 authors, list the first 6 authors followed by “et al.”. Do not use individual sets of parentheses for citation numbers that appear together, e.g., [2,3,5-9], not [2],[3],[5]–[9]. For other styles of publication or Internet articles, see [http:// www. nlm.nih.gov/ bsd/ uniform_ requirements.html](http://www.nlm.nih.gov/bsd/uniform_requirements.html) References should be formatted as follows (please note the punctuation and capitalization)

Journal titles should be abbreviated according to ISI Web of Science abbreviations. Authors are solely responsible for the accuracy and completeness of references.

Articles in Journals

Özpolat B, Gürpınar ÖA, Ayva EŞ, Gazyağcı S, Niyaz M. The effect of Basic Fibroblast Growth Factor and adipose tissue derived mesenchymal stem cells on wound healing, epithelization and angiogenesis in a tracheal resection and end to end anastomosis rat model. *Türk Gogus Kalp Dama* 2013; 21: 1010-9.

For other styles of publication or Internet articles, see [http:// www.nlm.nih.gov/bsd/uniform_requirements.html](http://www.nlm.nih.gov/bsd/uniform_requirements.html)

Tables and Figures

All illustrations (photographs, drawings, graphs, etc.), not including tables, must be labeled “Figure.” Figures must be submitted both in the manuscript and as separate files.

All tables and figures must have a caption and/or legend and be numbered (e.g., Table 1, Figure 2), unless there is only one table or figure, in which case it should be labeled “Table” or “Figure” with no numbering. Captions must be written in sentence case (e.g., Macroscopic appearance of the samples.). The font used in the figures should be Times New Roman. If symbols such as $\times$, μ , η , or ν are used, they should be added using the Symbols menu of Word.

All tables and figures must be numbered consecutively as they are referred to in the text. Please refer to tables and figures with capitalization and unabbreviated (e.g., “As shown in Figure 2...”, and not “Fig. 2” or “figure 2”). The tables and figures themselves should be given at the end of the text only, after the references, not in the running text.

The resolution of images should not be less than 118 pixels/cm when width is set to 16 cm. Images must be scanned at 1200 dpi resolution and submitted in jpeg or tiff format. Graphs and diagrams must be drawn with a line weight between 0.5 and 1 point.

Graphs and diagrams with a line weight of less than 0.5 point or more than 1 point are not accepted. Scanned or photocopied graphs and diagrams are not accepted.

Charts must be prepared in 2 dimensions unless required by the data used. Charts unnecessarily prepared in 3 dimensions are not accepted.

Figures that are charts, diagrams, or drawings must be submitted in a modifiable format, i.e. our graphics personnel should be able to modify them. Therefore, if the program with which the figure is drawn has a “save as” option, it must be saved as *.ai or *.pdf. If the “save as” option does not include these extensions, the figure must be copied and pasted into a blank Microsoft Word document as an editable object. It must not be pasted as an image file (tiff, jpeg, or eps) unless it is a photograph.

Tables and figures, including caption, title, column heads, and footnotes, must not exceed 16×20 cm and should be no smaller than 8 cm in width. For all tables, please use Word’s “Create Table” feature, with no tabbed text or tables created with spaces and drawn lines. Please do not duplicate information that is already presented in the figures.

Tables must be clearly typed, each on a separate sheet, and double-spaced. Tables may be continued on another sheet if necessary, but the dimensions stated above still apply.

Instruction to Reviewers

The Current Thoracic Surgery is a current periodical, peer-reviewed and open access official e- journal of the Turkish Society of Thoracic Surgery which its funded and published three times annually. In April, August and December.

The language of the journal is English.

The Current Thoracic Surgery is dedicated to publishing clinical research, clinical analysis, laboratory and experimental studies, editorials, invited current reviews, case reports, interesting images and “How to Do It” papers.

The journal is based on independent and unbiased double-blinded peer-reviewed principles. Only unpublished papers that are not under review for publication elsewhere can be submitted. Current Thoracic Surgery does not accept multiple submission and duplicate submission even though the previous one was published in a different language. The authors are responsible for the scientific content of the material to be published. The Current Thoracic Surgery reserves the right to request any research materials on which the paper is based.

The Current Thoracic Surgery encourages and enables academicians, researchers, specialists to publish their valuable research in branches of general thoracic surgery, thoracic disease and thoracic surgery anesthesia. The primary aim of the journal is to publish original articles with high scientific and ethical quality and serve as a good example of medical publications in the World.

The Editorial Board of the Current Thoracic Surgery and the Publisher adheres to the principles of the International Council of Medical Journal Editors (ICMJE), the World Association of Medical Editors (WAME), the Council of Science Editors (CSE), the Committee on Publication Ethics (COPE), the US National Library of Medicine (NLM), the World Medical Association (WMA), the US Office of Research Integrity (ORI), the European Association of Science Editors (EASE), and the International Society of Managing and Technical Editors (ISMTE).

All articles are available in PDF format on our website ([www.http://cts.tgcd.org.tr/](http://cts.tgcd.org.tr/)) and can be downloaded free of charge.

All articles submitted for publication are strictly reviewed for their
originality,
methodology,
importance,
quality,
ethical nature and,
suitability for the journal

The Current Thoracic Surgery uses a well-constructed scheme for the evaluation process.

The overall rejection rate of the Current Thoracic Surgery is 29%.

The average time during which the preliminary assessment of manuscripts is conducted is 4 days.

The average time during which the reviews of manuscripts are conducted is 41 days.

The average time in which the article is accepted for publication is 59 days.

The entire submission process for a manuscript is completed online through the self-explanatory online submission system.. The reviewers can reach their personal pages from the same address with their own passwords.

Manuscripts that comply with the main rules of the journal are sent to at least two external reviewers, and the reviewers are asked for their opinion about the suitability of the paper for publication. The reviewed manuscripts are then re-reviewed by the Editor in Chief and the Editorial Board and a decision of rejection or acceptance is shaped.

If the reviewers have any **potential competing interests**, they must notify the editor before agreeing to review a submission.

The Current Thoracic Surgery is committed to the highest standards of **research and publication ethics**. Editors will act in accordance with the relevant international rules of publication ethics (i.e., COPE guidelines, ICMJE Recommendations, CSE White Paper on Publication Ethics, CSE White Paper on Publication Ethics, WAME resources, WMA policies and ORI) if any ethical misconduct is suspected. The Editorial Board of the Current Thoracic Surgery encourages reviewers to comment on possible research or publication misconduct such as unethical research design, duplication, plagiarism, etc.

Plagiarism is a serious problem and the most common ethical issue afflicting medical writing. Current Thoracic Surgery does not allow any form of plagiarism. In accordance with our journal policy, submitted manuscripts are screened with plagiarism software to detect instances of overlapping and similar text (iThenticate and others) at least two times (during the evaluation process and after acceptance). If the reviewers have any suspect, the editors can provide them information obtained by plagiarism screening tools.

An **approval of research protocols by an ethics committee** in accordance with international agreements (“WMA Declaration of Helsinki - Ethical Principles for Medical Research Involving Human Subjects (last updated: October 2013, Fortaleza, Brazil)”, “Guide for the care and use of laboratory ani-

mals (8th edition, 2011)” and/or “International Guiding Principles for Biomedical Research Involving Animals (2012)” is required for all research studies. If the submitted manuscript does not include ethics committee approval, it will be reviewed according to COPE’s guideline (Guidance for Editors: Research, Audit and Service Evaluations). If the study should have ethical approval, authors will be asked to provide ethical approval. If they cannot provide ethical approval, their manuscript will be rejected and also their institutions and when needed, the related bodies in their country will be informed that such studies must have ethics committee approval. If the study does not need ethics committee approval after the editorial board’s review, the authors will be asked to provide an ethics committee approval or a document given by a related independent committee that indicates the study does not need ethics committee approval according to the research integrity rules in their country. If the authors provide either an approval or a document showing that ethics approval is not needed, the review process will continue. If the authors cannot provide either documents, the manuscript will be rejected. For articles concerning experimental research on humans, a statement should be included that shows informed consent of patients and volunteers was obtained following a detailed explanation of the procedures that they may undergo.

Informed consent must also be obtained for case reports.

All recognizable photographs of a patient must be accompanied by written permission from the patient for reproduction. Procedures that were performed to eliminate any pain, harm and distress in subjects/animals should clearly be stated. The authors should clearly state their compliance with internationally accepted guidelines and the guidelines issued by the relevant authority of their country. The journal requests a copy of the Ethics Committee Approval received from the relevant authority.

The Current Thoracic Surgery wants reviewers to treat the manuscripts in confidence. The material of the manuscripts must not be used or shared in any way until they have been published. The Current Thoracic Surgery follows the COPE flowchart in cases of suspected reviewer misconduct. Please refer to COPE ethical guidelines for peer reviewers for “Basic principles to which peer reviewers should adhere” and “Expectations from reviewers”

Our reviewers are expected to check that the articles comply with the SAGER guidelines and encourage authors to do so. Authors should use the terms sex (biological attribute) and gender (shaped by social and cultural circumstances) carefully in order to avoid confusing both terms. Article titles and/

or abstracts should indicate clearly what sex(es) the study applies to. Authors should also describe in the background, whether sex and/or gender differences may be expected; report how sex and/or gender were accounted for in the design of the study; provide disaggregated data by sex and/or gender, where appropriate; and discuss respective results. If a sex and/or gender analysis was not conducted, the rationale should be given in the discussion.

The Current Thoracic Surgery uses bibliographic databases and also accepts authors’ suggestions to find new reviewers. The journal thanks to the reviewers and publishes the reviewer list every year in the last issue.

Besides the routine questions given at the reviewers’ comment page, the reviewers can also use the questions below, when reviewing the manuscripts:

1. Please do state if any conflict(s) of interest is present to review this manuscript.
2. Do you suspect any research or publication misconduct?
3. Does the manuscript contain significant information to justify publication?
4. Is the title of the article appropriate?
5. Does the abstract clearly and accurately describe the content of the article?
6. Is the problem significantly and concisely stated?
7. Are the methods of the study described comprehensively?
8. Is the results section clear and satisfactory?
9. Are the interpretations and conclusions justified by the results?
10. If adequate and current references were given?
11. Is the terminology used in the text appropriate for a medical writing?
12. Are the figure/table legends satisfactory?
13. Is it necessary to shorten/elongate the article sections?

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