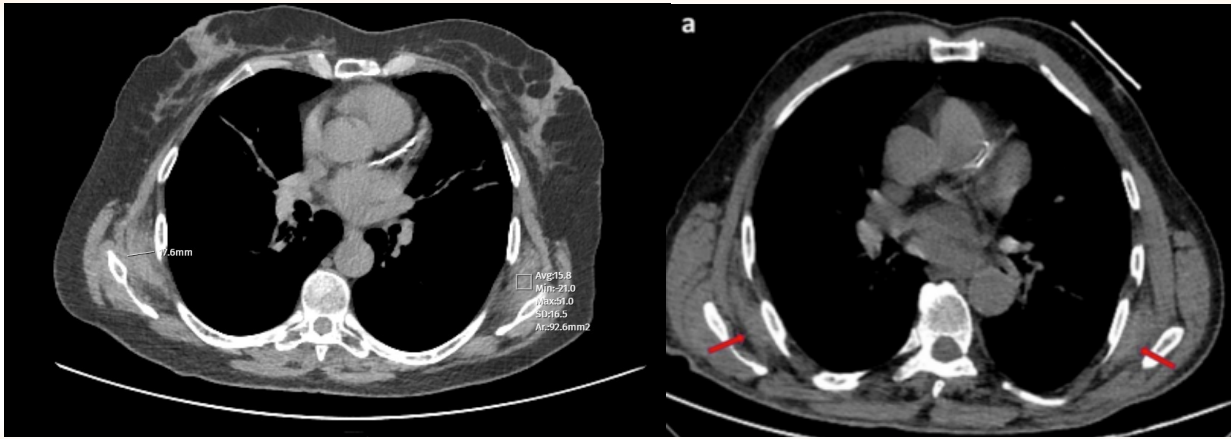


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Original Article

A quantitative method to evaluate rest periods and respiratory functions in the recovery process of chest trauma cases: peak expiratory flow rate measurement

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ABSTRACT

Background: In this study, the factors that had an effect on the recovery of respiratory functions of the patients with chest trauma were investigated, and it is aimed to question the adequacy of the follow-up and rest periods with a different method.

Materials and Methods: One hundred and eighty patients with chest trauma hospitalized in our thoracic surgery department between August 2018 and December 2019 were analyzed. The effective factors on their recovery to normal respiratory functions were investigated using “Peak Expiratory Flow-meter” (PEF-meter). PEF values were recorded on the first three days of hospitalization and on the 10th, 40th and 70th days after their discharges.

Results: We examined when the chest trauma patients were able to reach a PEFR value of 80% (PEF80) as done in bronchial asthmatics. Pneumothorax, lung laceration, presence of pneumomediastinum and tube thoracostomy were found to delay achieving a PEF80. It was found that the location of the fracture, rather than the number of rib fractures, was effective in improving respiratory functions. Non-smokers were seen to have a higher capacity to return to normal respiratory activity after trauma.

Conclusions: In case of fractures occurring on the anterior and lateral of the ribs and in the situation of the presence of pneumothorax, pneumomediastinum, lung laceration, tube thoracostomy and smoking history pain management and follow-up should be handled more detailed. It should be taken into account that these cases may require longer rest period and disability/incapacity reports should be prepared bearing this in mind.

Keywords: peak expiratory flow rate, pneumothorax, pulmonary function test, rib fractures, smoke, thoracic injuries

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Introduction

Today, traumas are increasing due to both the increase in transportation opportunities and higher violence rate. Trauma requires hospitalization at every stage of life and is one of the main causes of disability. 10-15% of trauma patients' admissions are due to chest injuries, and the resulting pathologies are related to approximately 25% of mortalities [1-3]. Pneumothorax, hemothorax, lung contusion and rib fracture are the frequently encountered pathologies in chest trauma cases [4]. Frequently observed rib fractures cause chronic pain-related problems [5].

The treatment of most chest trauma cases do not call for a major surgery and more than 80% of the cases can be treated with pain control, effective respiratory physiotherapy and tube thoracostomy [3]. However, those who leave the hospital following their treatment may experience respiratory function limitation, which affects their quality of life [6,7]. Quantitative data are needed to measure the effect of trauma on respiratory functions and to objectively evaluate the treatment and follow-up approach [7]. "Peak Expiratory Flow" (PEF) measurement is a valuable method used to assess respiratory function. In monitoring the severity, course and response to treatment of the disease, the importance of daily recording of PEF value in bronchial asthmatics has long been known [8,9]. In addition to the factors that prevent the expansion of the thoracic cage and hinder respiratory muscle function, many factors such as any pathology that obstructs the airways and damage to the central nervous system can cause changes in the PEF value [8].

A thoracic surgeon should be able to competently treat a trauma patient, in addition to managing and monitoring the possible complications in the post-discharge period. The hospitalization period of trauma patients is mostly determined with the ending doctor and hospital requirement. Thinking differently from this customary condition, we aimed to categorize the cases with chest trauma using a quantitative method based on the question: "Which patients should be followed up for longer period of time?". In this study, bearing in mind that PEF value is affected by chest wall pathologies, we performed the measurable follow-up of patients with chest trauma using PEF-meter device which measures the peak flow velocity in chest trauma patients. We also aimed to reveal the pathologies that prevented the patients initiated an inpatient treatment and later followed up in the outpatient

clinic from achieving the expected respiratory function value. It was also aimed to investigate whether the data to be obtained would be a determining factor in terms of both patient follow-up and rest periods to be given.

Material and Methods

The study was prospectively carried out with the participation of 180 chest trauma patients. The patients were hospitalized in our thoracic surgery department and also followed up in the outpatient clinic later between the dates of August 2018 and December 2019. All patients were included in the study after reading and confirming the "informed voluntary consent form". The study was initiated with the approval letter obtained from the Ege University School of Medicine Ethics Committee dated 19.06.2018 and decision number 18-6.1/37.

Those excluded from the study were as follows: the patients hospitalized on the second and following days of their trauma; those with mental or auditory disabilities that prevent the use of PEF-meters, those with cranial traumatic injuries with altered consciousness, those with lip, tongue, and facial injuries; those having underwent surgical intervention outside our hospital and referred to us; those who underwent surgery under general anesthesia due to trauma in our hospital; severe multisystem injuries requiring intensive follow-up of other clinics and lastly the cases under 18 years of age.

PEF measurements were made on the first 3 days of hospitalization and on the 10th, 40th and 70th days post discharge in accordance with the method in the outpatient clinic control. The value measured on the first day of the patient admissions was recorded as "clinical value 1st day" (CV1) then 2nd day (CV2) and 3rd day (CV3), and also the value measured in the outpatient follow-up on the 10th day of the discharge was recorded as the "outpatient value 10th day" (OV10), then as 40th day (OV40) and as 70th day (OV70).

Measurements were made using the "MSA-100 Peak Flow Meter" (patented by Beijing M&B Electronics Instruments Co Ltd, Beijing, People's Republic of China) digital PEF-meter. With personalized mouthpieces, the device was prepared for multiple uses. The use of the PEF-meter device with the appropriate technique was explained to all participants.

The measurements were made at the same time of day and in body position (feet hanging off the side of the

bed and in a way to let a comfortable breathing position). Three consecutive PEF measurements were made each time and the highest one was recorded. The applied treatments were standardized with a drug group consisting of analgesics, mucolytics, antibiotics and proton pump inhibitors. The surgical procedure of those who needed tube thoracostomy was performed using the same method, from the same site. In addition to the demographic data of the patients, such data as trauma mechanism, injury site, radiological findings, intervention, smoking habit, length of stay were analyzed. These variables were evaluated according to the PEF levels, which should be appropriate for the age and height of the subjects.

Statistical Analysis

Categorical data were expressed as numbers and percentages. Numerical variables were summarized as mean $\pm$ standard deviation when normally distributed, and as median (min, max) when not normally distributed. Cross-tables were created for group comparison in categorical data and chi-square analysis was performed. Comparisons between the two groups were made using the t-test for those with normal distribution in terms of numerical values, and using the Mann-Whitney U test for those who did not show normal distribution. The $p < 0.05$ was accepted as statistically significance.

Results

Of the 180 patients, 145 (80.6%) were male and 35 (19.4%) were female, with a mean age of 52.67 ± 18.27 (18-93). In the series of 77 (42.7%) active smokers, there were 57 (31.7%) non-smokers and 46 (25.6%) ex-smokers. Comorbidities were observed in 64 of the cases (35.5%) most of which were in the form of hypertension (35 cases), diabetes mellitus (27 cases), coronary artery disease (23 cases), and chronic obstructive pulmonary disease (8 cases).

One hundred fifty-six (86.7%) of the subjects in the study had blunt injuries, while 24 (13.3%) had penetrating injuries. When analyzed according to subgroups; falling down was observed in 72 (40%), in-vehicle traffic accidents (IVTA) in 45 (25%), and sharp object injuries in 20 (11.1%) cases and in addition, motorcycle accidents (8.3%), out-vehicle traffic accident (OVTA) (7.2%), assault (3.9%), crushing (2.2%) and firearm injuries (FAI) (2.2%) were also observed.

Additional injuries requiring consultation of other clinics were also examined. Eight (4.4%) patients with

cranial injuries were not excluded from the study as they did not cause problems in clinical perception and use of PEF-meters. Six of the cases (3.3%) had abdominal injuries not requiring surgery. In 32 patients (17.8%) with additional fractures at any level of the vertebral column, no condition to prevent the use of PEF-meters was observed. The number of cases with thoracic vertebral fracture was 14 (7.8%). While we observed clavicle fractures in 8 (4.4%) cases, scapula fractures were found in 4 (2.2%) cases.

When those with parenchymal and pleural pathologies were evaluated, it was seen that there was pulmonary contusion in 52 (28.9%) cases, atelectasis in 36 (20%) and parenchymal laceration in 17 (9.4%) cases. Pneumothorax was present in 86 (47.8%) patients and hemothorax in 71 (39.4%). Also, 11 (45.8%) had displaced fractures and 24 cases (13.3%) were present with sternum fracture. The number of cases with rib fractures in the right and left hemithorax was close to each other, and 18 (10%) patients had bilateral rib fractures. There were other pathologies without rib fracture in 35 of the cases (19.4%). Several general patient characteristics are given in table 1.

Table 1. Patient characteristics and their traumatic injuries.

		Count	Rate (%)
Gender	Male	145	80.6
	Female	35	19.4
Smoking history	Never smoked	57	31.7
	Ex-smoker	46	25.6
	Smoker	77	42.8
Type	Blunt	156	86.7
	Penetrating	24	13.3
Injury	Cranial injury	8	4.4
	Intra-abdominal injury	6	3.3
	Vertebral fracture	32	17.8
	Clavicle fracture	8	4.4
	Scapula fracture	4	2.2
	Sternal fracture	24	13.3
	Rib fracture	145	80.6
	Lung contusion	52	28.9
	Lung laceration	17	9.4
	Pneumothorax	86	47.8
	Hemothorax	71	39.4
PEF 80	Patients reached PEF 80	94	52.2
	Can't reach PEF 80	86	47.8

The number of patients who achieved the PEF80 value in the 70-day follow-up period was 94 (52.2%), while the number of those achieving the PEF100 value was 55 (30.6%). There was no significant relationship between gender and achieving post trauma normal respiratory functions ($p = 0.780$). The mean age of those who achieved PEF80 in the 70-day-follow-up period was 54.48 ± 18.49 (20-93), while those who could not accounted for 50.61 ± 17.92 (18-93). Although those achieving PEF80 looked older than those who could not, no statistical significance was demonstrated ($p = 0.157$).

While 63.2% of non-smokers achieved the PEF80 by the 70th day; 58.7% of ex-smokers and 42.9% of active smokers were able to achieve this value, which shows that non-smokers had a higher capacity to restore normal respiratory functions in the post trauma period ($p = 0.046$). When active smoking was analyzed separately, 44 (57.1%) of 77 patients in this group did not achieve the PEF80 value. Being an active smoker during the trauma period was the worst factor affecting the post-traumatic recovery of respiratory capacity ($p = 0.015$). No relationship was found between the amount of smoking in pack-years ($p = 0.056$) and the number of years before quitting ($p = 0.241$), and returning to normal respiratory functions in post trauma ($p > 0.05$). Although a statistically significant relationship could not be demonstrated between the presence of comorbidity and failure to achieve the PEF80 value, it was an interesting finding that 60.9% of those with comorbidities achieved the PEF80 value.

In terms of etiology, it was understood that 82.1% of those who could not achieve the PEF80 were present with a blunt trauma. However, the fact that the number of cases with penetrating trauma in our series was very small should be taken into consideration, which might also account for why there was no significant relationship between the etiology of trauma and the ability to return to normal respiratory function ($p = 0.217$). It was found that both bone fractures such as scapula ($p = 0.491$), clavicle ($p = 0.491$), thoracic vertebrae ($p = 0.490$), and an injury outside the rib cage did not affect achieving the PEF80 value ($p > 0.05$).

No statistically significant relationship was found between the pneumothorax side (right/left) ($p = 0.127$) and the ability to return to respiratory functions, and 55.6% of bilateral pneumothorax cases could not achieve PEF80. 62.5% of those with pneumothorax reaching PEF80 were those who had no pneumothorax at their first admission in post-trauma period. Presence of pneumothorax on the first chest tomography at admission was determined to

affect patient's returning to normal respiratory function process ($p = 0.031$). Although similar results were found when examined for hemothorax, statistical significance could not be demonstrated ($p = 0.055$).

There was no relationship between achieving the PEF80 and whether there was a unilateral or bilateral rib fracture ($p = 0.795$), its number ($p = 0.771$), the presence and number of displaced rib fractures ($p = 0.838$), however, it was determined that the localization of the fracture in the rib affected respiratory recovery. In a 70-day follow-up, 66.1% of patients with posterior rib fractures reached PEF80, whereas 59.1% with anterior rib fractures and 54.7% with lateral rib fractures could not. It was determined that those with a posterior rib fracture achieved the PEF80 at a higher rate ($p = 0.032$).

Lung laceration and pneumomediastinum were found to be effective factors in delaying the normalization of respiratory functions ($p = 0.038$) and ($p = 0.009$), respectively. Even though the number of cases with laceration was low, it was statistically significant that 12 cases did not reach PEF80, whereas 5 cases did. Considering the treatments, 61% of those who did not undergo tube thoracostomy and were followed up with medical treatment alone achieved PEF80. Also, it was remarkable to see that 79.4% of those who could not achieve the value had previously undergone tube thoracostomy. In terms of reaching PEF80, those who underwent tube thoracostomy were left behind compared to those who received medical treatment alone ($p < 0.001$). The relationship between PEF 80 value and several patient characteristics is given in the figure 1.

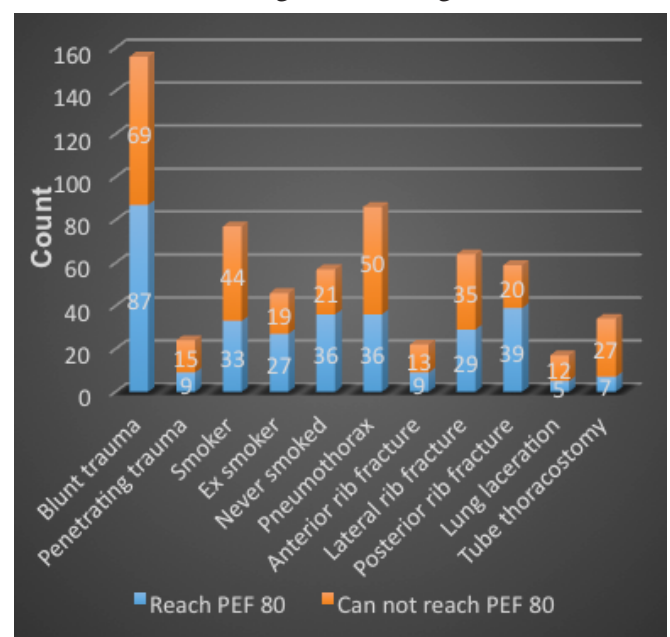


Figure 1. Distribution of patient count who can reach and cannot reach PEF 80 value according to various patient characteristics.

Discussion

In recent years, studies investigating the quality of life in post trauma period have started to be conducted. In a study by Ulvik et al [10] which was started with 325 cases and completed with 210 cases at the end of a seven-year follow-up and in another study by Kaske et al [11] concluded with 129 cases after following 380 cases for two years, it was reported that the quality of life decreased due to post-traumatic health problems in most of the patients, and that they still complained of pain even after years. The authors emphasized that pain control is an important factor in returning to normal health functions.

In a study reporting the five-year follow-up of 205 traumatized cases, it was reported that those with blunt trauma often experienced physical disability, while those suffering from penetrating trauma generally exhibited psychiatric problems [12]. Although there was no statistically significant difference between the two types of injury in terms of restoring normal respiratory functions in our study of 180 cases, the majority of whom had blunt trauma, the low number of cases with penetrating trauma in the series should be considered.

The literature generally focuses on post-traumatic long-term pain complaints, but there is no study designed in this series on the restoration of respiratory functions in the post-traumatic period. In order to question the adequacy of the follow-up period to be applied to trauma patients, we investigated the factors delaying the return to normal respiratory functions after discharge with a quantitative method.

As stated by other authors, we also believe that pain is the main factor preventing the return to adequate respiratory functions in patients with chest trauma. In our study, we tried to make this measurable by using PEF-meter and also to establish a connection with the damaged chest structures. For this purpose, we encountered no statistical relationship between reaching PEF80 in the 70-day follow-up and the number of rib fractures and in which hemithorax the fracture was ($p > 0.05$) while examining the number of post-traumatic rib fractures, the fracture side (right, left, bilateral), the localization of the fracture in the rib (posterior-anterior-lateral) and other bone fractures. However, it was determined that those with posterior rib fractures achieved the PEF80 more rapidly than those who had fractures with anterior and lateral localizations ($p = 0.032$). We attribute this

to the fact that the posterior ribs are supported by more muscle mass and soft tissue, and also to the supportive effect of the scapula in stabilization.

Although it is known that smoking increases respiratory problems, the number of studies investigating its effect on respiratory functions in the post-thoracic trauma period is limited. In a prospective study by Calfee et al in 2011, the respiratory problems related to smoking in the intensive care unit for blunt trauma patients were analyzed and it was reported in this study examining lung injury that the rate of development of acute lung injury increased significantly in both active and passive smokers [13]. Similarly, in our series, active smoking during the trauma period was found to negatively affect the restoration of respiratory functions ($p = 0.015$), though no statistically significant relationship was found between the amount of cigarette smoked and returning to normal respiratory values in post trauma period ($p = 0.056$).

In a study of 1,478 cases investigating the problems caused by the presence of comorbidities, the association of advanced age (over 65 years) and comorbidity with mortality was demonstrated. The authors attributed this to the negative effects of advanced age and comorbidity on fluid-electrolyte and acid-base balances, heart rate-rhythm-contraction strength, and coagulation mechanism [14]. Although there was no statistically significant relationship between age and comorbidity and achieving the PEF80 value in 180 cases of our series, it was an interesting finding that patients with comorbidities and a higher mean age were able to achieve PEF80, which can be explained by the high compliance of the patient group to the hospital, physician and medical treatment.

In a study by Svennevig et al in which the sequela of pulmonary contusion caused by trauma was examined using pulmonary function test and arterial blood gas parameters, with a mean follow-up period of 4.9 years, it was pointed out that the patients with rib fractures tried to live with significant respiratory sequelae for years [15]. In our series in which we analyzed the earlier results, no statistically significant relationship was found between lung contusion and achieving the PEF80 ($p = 0.809$). In order to draw a meaningful conclusion, we are of the opinion that both the number of cases and the follow-up period should be increased. In a study of 34 pediatric cases treated for lung contusion and laceration, it was stated that no respiratory sequelae were en-

countered [16], while in our study including adult cases, lung laceration was found to be a determining factor in preventing the cases from reaching PEF80 in 70-day follow-up period ($p = 0.038$).

Tube thoracostomy is a painful procedure and most patients complain about pain at the tube thoracostomy incision site. It was determined that 79.4% of those who could not achieve the PEF80 were those who underwent tube thoracostomy due to the pathology caused by trauma ($p < 0.001$). It can be concluded from this data that tissue damage is more common and the restoration of-normal respiratory functions will be slower due to pneumothorax and hemothorax, and it can also be considered that tube thoracostomy itself is a painful procedure.

It should be kept in mind that pneumomediastinum may accompany pneumothorax in high-energy chest traumas [17], and eight of nine patients with pneumomediastinum in our series could not achieve the PEF80 during the follow-up period ($p = 0.009$). In the literature, no study has been found that reveals the time to return to normal respiratory functions in patients with pneumothorax and hemothorax. In our series, there was a significant difference between the presence of pneumothorax in the first post-trauma computed tomography imaging and failure to achieve the PEF80 ($p = 0.031$), but there was a moderately statistical relationship ($p = 0.055$) between the presence of hemothorax and failure to achieve the PEF80.

In conclusion, although thoracic surgeons have different follow-up fashions for trauma patients, patients are followed up for an average of one or two months. The vast majority of cases return to work after the first month of trauma. However, our study consisting of 180 cases based on objective measurements revealed that nearly half of the patients could not achieve 80% of the required pulmonary function value even after a 70-day follow-up period.

In our series, we tried to determine what pathologies were present in those who had difficulty in achieving the PEF80 value. Even in the light of the preliminary information we obtained with the limited number of cases, we are of the opinion that a specific evaluation should be carried out based on the trauma type while preparing the treatment and resting reports. We recommend conducting studies of this nature, bearing in mind that series with more cases and longer follow-up peri-

ods are needed to calculate the optimal and specific resting times for each trauma type.

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

The study was approved by the Ege University School of Medicine Ethics Committee dated 19.06.2018 and decision number 18-6.1/37.

Authors' contribution

SA; project administration, conceptualization, developed the methodology, co-wrote the paper, AÇ; validation, manuscript reviewing and editing, SKA; conceived and designed the analysis, collected the data, co-wrote the paper, ÖK; collected the data, formulated and evaluated research goals and aims, AGE; collected and processed the data TİA; conceptualization, developed the methodology, AÖ; collected the data, KT; manuscript reviewing, UÇ; manuscript reviewing

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Original Article

Seven-year retrospective analysis of pleural effusion cytologies and cyto-histopathological correlation

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ABSTRACT

Background: The cytological examination of pleural effusions is a quick and effective diagnostic method used to elucidate the etiology. The aim of this study is to reveal cytopathological diagnosis distributions by retrospectively examining seven-year pleural cytology and analyze cyto-histopathological correlations in cases with biopsy in this study.

Materials and Methods: In this study, 545 pleural fluid aspiration materials that resulted as Bolu Abant İzzet Baysal University İzzet Baysal Training and Research Hospital between 2014 and 2020 were evaluated retrospectively. The evaluation results were divided into four groups as inconclusive, negative for malignancy, suspected malignancy, and malignant cytology. The biopsy diagnoses of 64 cases were grouped as inadequate, benign, and malignant. Sensitivity and specificity values were calculated by looking at the relationship between cytology and biopsy diagnoses.

Results: According to the cytology diagnoses, 435 cases (79.8%) were negative for malignancy, 43 cases (7.9%) were malignant, 32 cases (5.9%) were suspected of malignancy, and 35 cases (6.4%) were inconclusive. When the diagnoses of the cases with both cytology and biopsy analyses were compared, the sensitivity of the cytology was 96.7%, the specificity was 91.2%, the positive predictive value was 90.6%, and the negative predictive value was 96.7%. The biopsy and cytology results were found to be compatible ($p < 0.05$)

Conclusions: Pleural effusion develops due to different etiologies, and the cytological examination of pleural fluid is an important diagnostic method for malignancy. In our region, the most common cause of pleural effusion is benign pathologies which are consistent with data in the literature. The correlations between cytology- and biopsy-based diagnosis results were found high.

Keywords: Pleural effusion, malignancy, pleural effusion cytology, pathology, retrospective

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Introduction

The pleura is the serous membrane that surrounds the lungs and thoracic cavity, consisting of two layers as parietal and visceral layers [1,2]. Pleural fluid is produced by the parietal pleura and reabsorbed by lymphatic channels [3]. A disruption in this homeostasis causes pleural effusion [3].

Pleural effusion occurs with the accumulation of fluid in the pleural cavity in intrathoracic or systemic diseases [1,4]. Fluid may accumulate in the pleural cavity because of benign or malignant causes [5,6]. Biochemical, microbiological, and cytological examinations are performed in pleural fluids taken by thoracentesis in patients with pleural effusion [7]. Pleural cytology examination also provides useful information in terms of diagnosis, tumor staging, and guiding treatment, protecting patients from a more invasive examination and reducing health expenditures [7,8]. When pleural cytology is not diagnostic, pleural biopsy or thoracoscopic biopsy is performed [7].

The sensitivity of pleural cytology varies by 40-87% because of the retrospective nature of studies, differences in the criteria examined and cytopathological methods used in different studies, and the usage of old guidelines [7,8]. It is known that cytology has a high diagnostic value in malignant pleural fluids [9,10]. It has been reported that the most common causes of malignant pleural effusion are lung cancer and breast cancer [1].

The aim of this study is to examine pleural cytology samples sent to the pathology laboratory because of pleural effusion, reveal the characteristics and cytopathological diagnosis distributions of seven-year pleural cytology results retrospectively, and analyze cyto-histopathological correlations in cases with biopsy.

Material and Methods

This study is a retrospective descriptive study. In this study, 545 pleural fluid aspiration materials belonging to cases who underwent thoracentesis because of pleural effusion with results sent to Bolu Abant İzzet Baysal University, İzzet Baysal Training and Research Hospital between 2014 and 2020 were evaluated retrospectively. The clinical parameters, cytology results, and biopsy results of the cases were obtained from archive records. Our study was conducted in accordance with the Declaration of Helsinki and was approved by the Bolu Abant İzzet Baysal University Ethics Committee for Clinical Research (2021/39).

In cases with benign pleural cytology, one of the cytology samples was evaluated, and the other cytology samples were not included in the study. In the case of multiple pleural cytology results, malignant cytology results were included in the study. The cytology samples were stained with the Papanicolaou method and the May Grunwald-Giemsa stain. In cases where a cell block could be obtained, cell block was created and stained with hematoxylin and eosin. The cytopathological evaluation results were divided into four groups as inconclusive, negative for malignancy, suspected malignancy, and malignant cytology [11,12]. Malignant pleural effusion was accepted as the presence of malignant cells in cytology [13].

In 64 cases with biopsy results, the biopsy results were classified as inconclusive, benign, and malignant. The statistical comparisons and cyto-histopathological correlation analyses were carried out in terms of the cytology and biopsy diagnoses.

Statistical Analyses

In this study, frequency analysis and percentage distribution analyses of the data were carried out using the SPSS 21 software (Version 21, Armonk, NY: IBM Corp). The variables did not show normal distribution, and they are presented with median (minimum–maximum) values. The comparative distributions results are given with cross-tables. Sensitivity and specificity values were calculated by looking at the compatibility between the cytology and biopsy diagnoses. $P < 0.05$ accepted as statistically significant.

Results

In this study, the ages of the patients with 545 pleural cytology results ranged from 2 to 105, with a mean age of 66.3. Three hundred and twenty-nine of the cases were male (60.4%), and 216 (39.6%) were female.

According to their cytological diagnoses, 435 cases (79.8%) were negative for malignancy, 43 cases (7.9%) were malignant, 32 cases (5.9%) were suspected of malignancy, and 35 cases (6.4%) were inconclusive. When the suspicion of malignancy in the cytological diagnoses was evaluated together with the malignant group, 75 cases were diagnosed as malignant, and their mean age was 65.2 years. The characteristics and diagnostic distributions of the pleural cytology examinations are given in table 1.

Table 1. Characteristics and diagnostic distribution of pleural cytology.

		Cytological diagnosis									
		Insufficient		Negative for malignancy		Suspicion of malignancy		Malign		Total	
		n	%	n	%	n	%	n	%	n	%
Gender	Female	11	31.4	168	38.6	12	37.5	25	58.1	216	39.6
	Male	24	68.6	267	61.4	20	62.5	18	41.9	329	60.4
	Total	35	100.0	435	100.0	32	100.0	43	100.0	545	100.0
Quantity	Low	29	82.9	337	77.6	20	62.5	27	62.8	413	75.9
	Middle	6	17.1	77	17.7	10	31.3	14	32.6	107	19.7
	High	0	0.0	20	4.6	2	6.3	2	4.7	24	4.4
	Total	35	100.0	434	100.0	32	100.0	43	100.0	544	100.0
The location of cytology	Right pleural space	17	48.6	238	54.7	12	37.5	26	60.5	293	53.8
	Left pleural space	17	48.6	160	36.8	13	40.6	15	34.9	205	37.6
	Bilateral	1	2.9	17	3.9	7	21.9	1	2.3	26	4.8
	Unknown	0	0.0	20	4.6	0	0.0	1	2.3	21	3.9
	Total	35	100.0	435	100.0	32	100.0	43	100.0	545	100.0
Color	Serous	25	71.4	281	64.6	21	65.6	15	34.9	342	62.8
	Serohemorrhagic	6	17.1	36	8.3	2	6.3	10	23.3	54	9.9
	Hemorrhagic	4	11.4	109	25.1	9	28.1	18	41.9	140	25.7
	Pus	0	0.0	9	2.1	0	0.0	0	0.0	9	1.7
	Total	35	100.0	435	100.0	32	100.0	43	100.0	545	100.0
Cell Block	No	27	77.1	301	69.2	26	81.3	17	39.5	371	68.1
	Yes	8	22.9	134	30.8	6	18.8	26	60.5	174	31.9
		35	100.0	435	100.0	32	100.0	43	100.0	545	100.0

A cell block could not be obtained in 371 cases (68.1%), and cell blocks were present in 174 cases (31.9%). Immunohistochemical staining was applied to the cell blocks for diagnosis in 40 cases (7.3%), and the most frequently used immunohistochemical stains were TTF-1, CK7, and Napsin-A. There were 32 cases which showed specific malignant diagnoses cytologically. The most frequently identified diagnoses of these cases were lung adenocarcinoma (14 cases, 43.8%) and breast invasive ductal carcinoma (5 cases, 15.6%) (Figure 1). The distribution of cytological diagnoses by sex is given in table 2.

The biopsies of 64 cases (12.5%) were also available from pleural cytology examinations. The biopsies were taken from the pleura (40 cases, 62.5%), lung (15 cases, 23.4%), peritoneum (3 cases, 4.7%), colon (2 cases, 3.1%), lymph nodes, endometrium, liver, and stomach (1 case in each, 1.6%). In the biopsies, 34 cases (53.1%) were diagnosed as malignant, 22 cases (34.4%) were diagnosed as

benign, and 8 cases (12.5%) were diagnosed as inconclusive. The most common malignant biopsy diagnoses were lung adenocarcinoma cases (13 cases, 20.3%), and the second most common were lung squamous cell carcinoma cases (5 cases, 7.8%). Immunohistochemical staining was applied to 36 of the biopsy materials (56.3%). The most frequently used immunohistochemical stains were TTF-1, which is an adenocarcinoma marker, Napsin-A, and calretinin and WT-1, which are mesothelioma markers. The diagnostic distributions of the biopsies are given in table 3.

Among the cases with malignant biopsy-based diagnoses, 91.2% were diagnosed as malignant cytology results. However, there were 3 cases with malignant biopsy diagnoses and negative cytology results for malignancy. The biopsy diagnoses of these cases were lung adenocarcinoma, lung squamous cell carcinoma, and endometrioid adenocarcinoma.

Of the cases with biopsy results that were benign, 95.5% had benign cytology results. The cytology of 85% of the cases with a biopsy-based diagnosis of chronic pleuritis was negative for malignancy, and one case was diagnosed as malignant in cytology. This case was diagnosed as lung adenocarcinoma in cytology, but chronic pleuritis was diagnosed in pleural biopsy in which a lung biopsy was not taken. The diagnoses of 87.5% of the materials with inconclusive biopsy results were negative in terms of malignancy, and there was no malignant biopsy in this group. The cytology of 92.3% of the cases diagnosed as lung adenocarcinoma was ma-

lignant. Malignant cytology was present in 80% of the cases diagnosed with lung squamous cell carcinoma. The mean age of the patients among the malignant biopsy results was 61.5 years.

When the cytological and biopsy diagnoses were compared, the sensitivity value of the cytology was 96.7%, the specificity value was 91.2%, the positive predictive value was 90.6%, and the negative predictive value was 96.7%. The diagnostic value of the cytological examinations is given in table 4. The biopsy and cytology results were found to be compatible ($p < 0.05$).

Table 2. Distribution of cytological diagnoses by gender.

		Gender					
		Female		Male		Total	
		n	%	n	%	n	%
Cytological Diagnosis	Lung adenocarcinoma	6	31.6	8	61.5	14	43.8
	Lung small cell carcinoma	1	5.3	3	23.1	4	12.5
	Lung squamous cell carcinoma	1	5.3	1	7.7	2	6.2
	Serous carcinoma of the ovary	3	15.8	0	0.0	3	9.4
	Invasive ductal carcinoma	5	26.3	0	0.0	5	15.6
	Peritoneal papillary serous carcinoma	1	5.3	0	0.0	1	3.1
	Malignant lymphoma	1	5.3	1	7.7	2	6.2
	Endometrioid Carcinoma	1	5.3	0	0.0	1	3.1
	Total	19	100.0	13	100.0	32	100.0

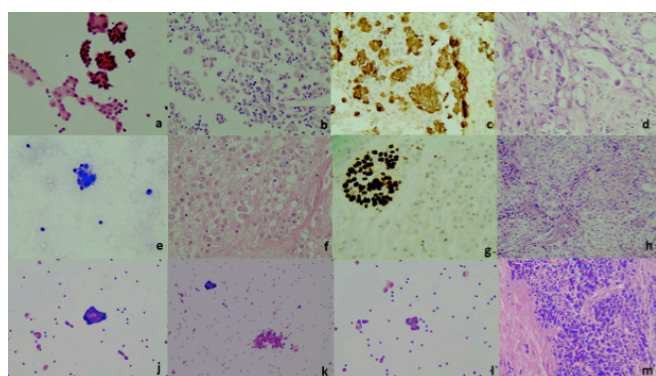


Figure 1. Malignant cytology (lung adenocarcinoma) Hematoxylin-eosin (HE), x400 (a), malignant cytology in cell block, x400 (b), immunohistochemical Napsin-A positivity in cell block, x400 (c), malignant biopsy, lung adenocarcinoma, HE, x400 (d), malignant cytology (invasive ductal carcinoma), May Grunwald-Giemsa (MGG), x400 (e), malignant cytology in cell block, HE, x400 (f), immunohistochemical estrogen receptor positivity in cell block, x400 (g), malignant biopsy, invasive ductal carcinoma, x200 (h), malignant cytology (lung small cell carcinoma), HE, x400 (j-k-l), malignant biopsy, lung small cell carcinoma, HE, x400 (m).

Table 3. Biopsy diagnoses

	n	%
Lung adenocarcinoma	13	20.3
Lung small cell carcinoma	4	6.3
Lung squamous cell carcinoma	5	7.8
Serous carcinoma of the ovary	3	4.7
Invasive ductal carcinoma	2	3.1
Invasive lobular carcinoma	1	1.6
Peritoneal papillary serous carcinoma	2	3.1
Endometrioid carcinoma	1	1.6
Colon adenocarcinoma	1	1.6
Intestinal T cell lymphoma	1	1.6
Other malignancy (Adenocarcinoma)	1	1.6
Insufficient Biopsy	8	12.5
Chronic pleuritis	20	31.3
Reactive mesothelial proliferation	1	1.6
Granulomatous infection	1	1.6
Total	64	100.0

Table 4. Diagnostic value of cytology.

		Biopsy Diagnosis											
		Malign biopsy		Benign biopsy		Total		Sens	Sp	ppv	npv	p	Kappa
		n	%	n	%	n	%	96.7	91.2	90.6	96.9	0.0001	0.875
Cytologic diagnosis	Malign	31	91.2	1	3.3	32	50.0						
	Benign	3	8.8	8.8	29	96.7	50.0						
	Total	34	100.0	30	100.0	64	100.0						

Abbrev.; Sens: Sensitivity, Sp: Spesifity, ppv: positive predictive value npv: negative predictive value

Discussion

The etiologies of pleural effusion may vary according to the age of the patient, geographical region, society, hospital, and clinic [2,14,15,16]. The pleura may be involved with primary and secondary tumors [17]. In malignancy, increased cellular permeability, tumor-associated angiogenesis, increased pleural fluid production, and lymphatic obstruction are the causes of pleural effusion in pleural metastasis [5,6,17]. The local and systemic effects of the tumor, treatment complications, and the direct involvement of the pleura with malignant cells are fluid formation mechanisms [6,17].

In malignant cases, cytopathological evaluation is more valuable than pleural tissue biopsy [1]. Cytological examination has advantages such as the easy acquirement of materials, fast results, fewer complications, and inexpensiveness [11]. The cytological diagnostic value in malignant pleural effusions varies in the range of 9-80%, and the diagnostic value in pleural biopsy varies in the range of 11-70% [2]. Factors such as cancer type, the experience level of the cytologist, and the volume of pleural fluid affect changes in these rates [18].

It was stated that at least 50-75 mL of aspiration material should be sent to increase the efficiency of the analysis in pleural cytology [3]. The cell block method used after the centrifugation of the pleural fluid is a method that supports the diagnosis because it reveals the tissue structure and allows immunohistochemical and molecular analyses [7,10,17]. In our study, the cell block method was performed in 26 (60.5%) of the cases with malignant cytology results.

When a malignant pleural disease is confirmed, two positive mesothelial markers (among calretinin, CK5/6, Wilms tumor 1, D2-40) and at least two negative immunohistochemical lung adenocarcinoma markers (TTF-1, CEA, Ber-EP4) to distinguish malignant pleural mesothelioma from

secondary tumors contribute to the differential diagnosis [3,10]. In our study, immunohistochemical staining was applied to the cell blocks, and the most commonly used markers were TTF-1, Napsin-A, calretinin, and WT-1.

Lung cancer, breast cancer, and lymphoma constitute approximately 75% of malignant pleural effusions [4,5]. In our study, the most common cause of malignant pleural effusion was lung adenocarcinoma, and the second most common cause was breast invasive ductal carcinoma.

Primary cancer foci may not be detected in malignant pleural effusions [1]. If the cytology result is negative, pleural biopsy is necessary [17]. However, it may show lower sensitivity in the case of insufficient tissue sampling and focal metastasis to the pleura [7]. In our study, there was a case with lung adenocarcinoma in cytology and a benign pleural biopsy. Since lung biopsy was not performed in this case, the biopsy result was negative.

The cytology experience of Ozdamar et al with 9043 cases showed that pleural cytology constituted 3% of all cytology results [11]. Cobanoglu found the most common cases as lung squamous cell carcinoma (9 patients, 30.60%) in their study of 49 malignant effusion cases [6].

In the study by Assawasaksakul et al, malignant diagnoses were made in 278 (78.8%) of 353 pleural cytology materials [7]. One hundred and thirty-two patients were diagnosed with lung non-small-cell cancer, 33 patients were diagnosed with breast cancer, and 31 patients were diagnosed with hematological malignancies [7]. In the study of 3077 pleural effusions conducted by Porcel et al, 840 patients (27%) had a malignant etiology [16]. Of the malignant cytology results, 309 (37%) consisted of lung malignancies, 123 (16%) consisted of breast malignancies, and 82 (10%) consisted of unknown primary malignancies [16]. Adenocarcinoma was commonly present in lung the lung malignancies (150 cases) [16].

In the study by Bayrak et al, malignant causes were found to be the largest group with 56 (36.6%) patients in 153 cases with pleural effusion [4]. Among the malignant effusion cases, the most common cause was lung cancer with 30 (54%) cases, and the second most common cause of malignant effusion was breast cancer with 9 (34.6%) patients [4]. In the distributions of the lung cancer cases according to histological types, there were 9 (30%) non-small-cell lung cancers, 10 (34%) squamous cell carcinomas, 5 (16%) adenocarcinomas, and 6 (20%) small-cell lung cancers [4].

Dagli et al reported 3 (1%) inconclusive cases, 8 (2.7%) atypical cases, 246 (82.6%) benign cases, 10 (3.4%) suspicious cases, and 31 (10%, 4) malignant cases [1]. They reported that the cause of malignant pleural effusion was metastatic carcinoma in 24 (8.1%) cases and malignant mesothelioma in 7 (2.3%) cases [1]. In the study by Gonlugur et al with 454 cases, malignant pleural effusion was seen in 32.6% of the cases, and the most common pleural effusions were malignant mesothelioma cases due to environmental asbestos exposure (67 cases, 14.8%) [14]. Uzunlar et al found malignant pleurisy in 54 (33.54%) cases and benign pleurisy in 107 (66.45%) cases [9]. In the malignant pleural effusion cases, the diagnostic value of cytology was found to be 55.56%, and the diagnostic value of pleural biopsy was 59.25% [9].

In the study by Dumanlı et al, the cytological malignant effusion rate was 17.15% (6 patients), the malignant pleural biopsy rate was 25.71% (9 patients), and the most common malignant tumors diagnosed in biopsy were adenocarcinoma metastasis at 8.57% (3 patients) and malignant mesothelioma at 5.71% (2 patients) [5]. Since our study was not conducted in an endemic region, there was no case of mesothelioma.

Thomas et al mentioned right sided pleural effusion as the most common case. The most common cause was reported as tuberculosis (344 cases, 84.5%) with malignant effusion at a rate of 5.2% in 21 patients [19]. The most common malignancies were lung adenocarcinoma (8 patients, 38%), breast carcinoma, and lymphoma (4 patients each, 19%) [19]. Riquet et al sampled pleural fluid during lung resection in 143 patients [12]. Pleural fluid positivity was observed in 10.7% (13/121) of the cases diagnosed as malignant in biopsy [12]. Positive adenocarcinomas were observed more frequently [12]. Mercer et al diagnosed 325 (32.6%) cases as malig-

nant pleural effusion from 998 pleural cytology materials [13]. The most common malignant diagnoses were breast cancers, non-small-cell lung carcinoma, and gynecological cancers, respectively [13].

In our study, the most common cause of pleural effusion was benign etiologies in accordance with the literature data. The mean age in the malignant effusion cases was 65.2. In our study, 2 patients (6.3%) were diagnosed with malignant lymphoma.

In the study by Tokgoz et al with 240 cases, 76 (32%) of the cases were female, 164 (68%) were male, and the mean age of all cases was 58 ± 20 years [15]. Among the effusion cases, 138 (57%) were on the right side, 71 (30%) were in the left hemithorax, and 31 (13%) were bilateral [15]. Malignant effusion constituted 25.4% of the cases [15]. The most common diagnoses were 38 (62%) primary lung cancer cases, 8 (13%) malignant mesothelioma cases, and 4 (7%) lymphoma-induced effusion cases [15]. In their study of 126 unilateral pleural effusion cases, Bintcliffe et al reported a mean age of 75 years, with 83 (66%) male cases and 43 (34%) female cases [20]. Right pleural effusion was seen in 78 cases (62%), and left pleural effusion was seen in 48 cases (38%) [20]. Malignant pleural effusion was diagnosed in 58 patients (46%) [20]. The most common types of malignancies were malignant mesothelioma (18 patients, 31%), lung cancer (17 patients, 29%), and breast cancer (5 patients, 9%) [20]. Similar to these studies, effusion was observed mostly in the male patients and in the right pleura in our study. Our malignant effusion rate in cytology was 14%.

In the study by Arnold et al which is the largest prospective study of 921 patients, a mean age of 70.2 ± 13.8 years was found with male predominance [8]. They performed cell block analyses in cases where 40 mL of cytology samples were obtained from the patients [8]. They found the malignant effusion rate to be 56% in 515 patients. Of these cases, the most common ones were lung cancer cases (32%, 166 cases, adenocarcinoma most common with 100 cases), which were followed by mesothelioma cases (29%, 148 cases) [8]. In their study, the mean sensitivity was 46%, and they stated that cytology was more valuable in detecting adenocarcinomas [8]. In our study, lung adenocarcinoma was the most common tumor, and a high consistency was found with a kappa value of 0.875 in our cytology and biopsy diagnoses.

Since this study is a retrospective study, there were

parts for which sufficient information could not be obtained. The study focused on malignant effusion cases, and benign etiologies could not be typified. In the study, the inability to send a sufficient amount of fluid to form a cell block limited immunohistochemical staining. Additionally, the limitations of the study were the inability to diagnose the cases whose cytological material came to our laboratory with biopsies performed in another center.

In conclusion, pleural effusion develops due to different etiological reasons, and the cytological examination of pleural fluid is an important diagnostic method for malignancy. In our region, the most common cause of pleural effusion is benign pathologies which are consistent with data in the literature. The most common cause of malignant pleural effusion is lung adenocarcinoma. The correlations between the cytology and biopsy diagnoses were found high. Since malignant pleural effusions often develop secondary to lung adenocarcinomas, it is thought that the lung should be investigated first for the primary focus in pleural effusions.

Declaration of conflicting interests

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Ethics approval

The study was approved by Bolu Abant İzzet Baysal University Ethic Committee for Clinical Research (2021/39).

Authors' contributions

SED,SO: involved in conceptualization, design, data analysis, manuscript writing, and review. SO: participated in data collection, data entry, and data analysis. SED,SO: participated in data analysis, manuscript writing, and review. All authors read and approved the final manuscript.

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Original Article

Prevalence and radiologic features of elastofibroma dorsi: a CT study

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ABSTRACT

Background: The study aimed to examine the prevalence and radiological features of Elastofibroma Dorsi (ED), detected incidentally in patients who underwent CT for various reasons.

Materials and Methods: A total of 8378 chest CT examinations were reviewed retrospectively for ED from January 2019 to April 2020. Lesion side, thickness, density, and whether stated in the initial report were examined.

Results: Of the 8378 patients, 87 patients (1.03%) (41 women and 46 men) had a total of 114 ED. There was no significant difference in the mean age of ED patients regarding gender ($p < 0.001$). When the ages of the patients included in the study with ($n = 87, 52.0 \pm 14.7$) and without ED ($n = 8291, 42.0 \pm 15.2$) were compared, there was a statistically significant difference ($p < 0.001$). The rate of bilateral ED was 31%. A positive correlation was determined between mean ED thickness and age ($p = 0.001$ and $p = 0.041$ for right and left, respectively). It was noted that none of the patients were diagnosed in the first radiological report.

Conclusions: ED is a slow-growing non-neoplastic, soft tissue tumor with a typical periscapular location. Being aware of this entity will prevent the need for advanced diagnostic evaluation. The characteristic appearance of these lesions will reduce unnecessary biopsy and surgery with increased radiologist awareness of their recognition.

Keywords: elastofibroma dorsi, prevalence, computed tomography, soft tissue neoplasms

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Introduction

Elastofibroma Dorsi (ED) is a non-neoplastic, slow-growing, benign, rare soft tissue tumor [1]. It is generally observed between the fourth and seventh decades of life, mainly in women [2,3]. 12-73% of cases are bilateral [4-6]. Although the exact etiology remains unclear, friction, trauma, tissue degeneration, and genetic predisposition among co-occurrence hypotheses have been described in the literature [6].

Clinically, 50% of cases are asymptomatic, and when symptoms occur, mild or severe pain accompanied by a clicking sound during abduction and adduction of the shoulder is typical for ED [7,8,9].

In the cross-sectional examination, ED contours may be sharp or ambiguous. The mass contains both fibrous tissue and adipose tissue. CT and MRI are particularly effective in diagnosis as they demonstrate the layered nature of the tumor's fat and fibrous tissue (with imaging properties similar to muscle on CT and MRI). Imaging features and localization are usually typical, and a definitive diagnosis can be made in most cases. CT is less sensitive in demonstrating adipose tissue than MRI, and therefore, it can be determined as a homogeneous mass slightly lower than muscle density on CT [9].

This study aimed to examine the prevalence and radiological features of ED, detected incidentally in a large population of patients who had CT for various reasons.

Materials and Methods

Patient cohort

All patients referred to the Radiology Department for various reasons (e.g., nodule follow-up, infection, lung cancer, staging of other tumors, acute trauma, mediastinal mass) between January 2019 and April 2020 were evaluated retrospectively. A total of 8378 chest CTs constituted the study group. Of these patients, 4584 (55.3%) were male, 3704 (44.7%) were female, and the mean age was 42.1 ± 15.2 years (0-94). Approval from the Institutional Review Board of Haseki Research and Training Hospital was obtained without the requirement for patient informed consent because of the retrospective origin of the study (Decision number: 2020-131).

Imaging

All CT images were acquired using a 128 detector CT alignment (Philips Ingenuity, Netherlands). Images were obtained with the patient at the end of inspiration and su-

pine position with 120 kV tube voltage, tube current-exposure time product, 512x512 matrix, 415 mm FOV, 200-300 mAs, and 1.25 mm section thickness after reconstruction.

Image analysis

After reviewing all patients retrospectively, the two radiologists evaluated the determined ED lesions with consensus regarding lesion density and thickness, and the lesion side was noted. While evaluating the lesion size, the thickness was measured as the greatest diameter in the axial plane (Figure 1). The initial radiology report of the patients with ED was reviewed regarding whether the lesion was mentioned in the first evaluation.

Statistical Analysis

SPSS 15.0 for Windows program was used for statistical analysis. Descriptive statistics were presented as numbers and percentages for categorical variables and mean, standard deviation, minimum, maximum, median, and interquartile range values for numerical variables. Numerical variables in two independent groups were compared using the Student's t-test when the normal distribution condition was met and the Mann-Whitney U test when the normal distribution condition was not met. Chi-Square Test performed the comparisons of the rates in the groups. Correlations were analyzed by Spearman Correlation Analysis when the parametric test condition was not met. The statistical alpha significance level was accepted as $p < 0.05$.

Results

In our study, 87 (1.03%) of 8378 patients had a total of 114 EDs. There were unilateral lesions in 60 (69.0%) patients and bilateral (31.0%) lesions in 27 (Figures 2,3). Of the ED patients, 46 (52.9%) were male, and 41 (47.1%) were female. The mean age was 54.6 ± 14.8 years (range 20-85) for females and 49.7 ± 14.5 years (range 24-81) for males. There was no significant difference in the mean age of men and women with ED ($p < 0.001$) (Table 1). When the ages of the patients included in the study ($n = 87$, 52.0 ± 14.7) with and without ED ($n = 8,291$, 42.0 ± 15.2) were compared, there was a statistically significant difference ($p < 0.001$) (Table 2).

In our study, the rate of bilateral ED was 31%. There was no significant difference between the mean age of patients with bilateral lesions and those with unilateral lesions ($p = 0.101$), and no significant gender difference was determined in both patient groups ($p = 0.737$) (Table 3).



Figure 1. The thickness was measured as the greatest diameter in the axial plane as shown in the right (17.6 mm) and the density was measured as shown in the left of the patient (15.8 ± 16.5 , range (-) 21-51).

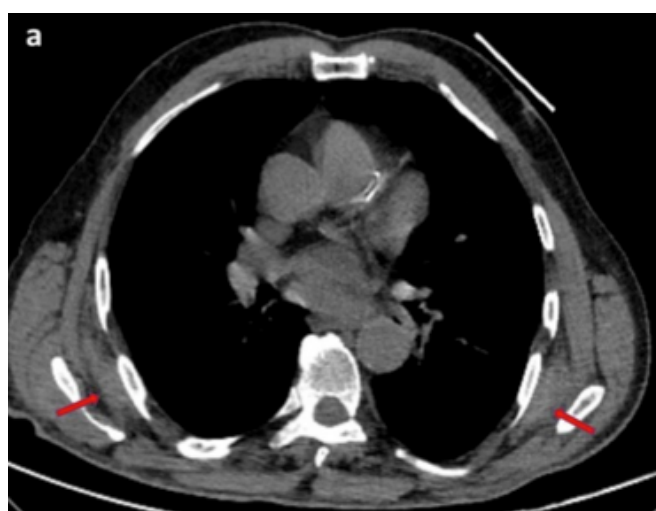


Figure 2a. Non-contrast axial CT image; bilateral ED (red arrows) with a streaky fatty component in an 83-year-old male patient located between the serratus anterior and latissimus dorsi muscles bilaterally.



Figure 2b. Coronal CT image; of the same patient.

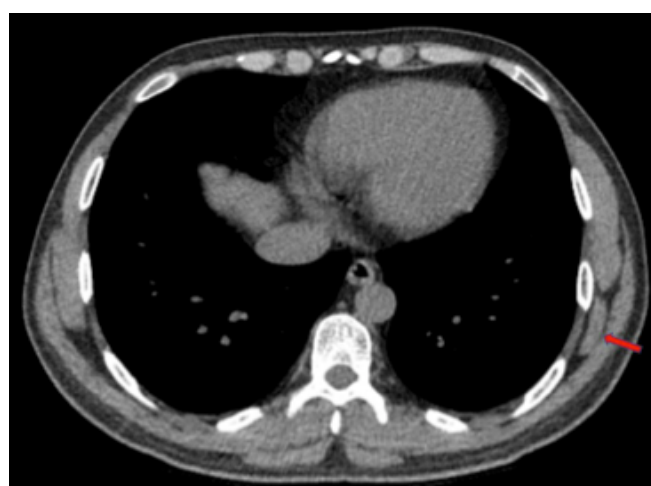


Figure 3. In the axial CT image, a 66-year-old female patient with left unilateral ED (red arrow).

Table 1. The entire patient population; Number of patients with and without ED, age, and gender.

		Age			Gender		
		Mean $\pm$ SD (Min-max)	Median (IQR)	p	Female n (%)	Male n (%)	p
N=8378	Total	42.1 $\pm$ 15.2 (0-94)	41 (30-52)		3746 (44.7)	4632 (55.3)	
Elastofibroma dorsi	No	42.0 $\pm$ 15.2 (0-94)	41 (30-52)	<0.001	3705 (98.9)	4585 (99.0)	<0.001
	Yes	52.0 $\pm$ 14.7 (20-85)	52.5 (40.25-63)		41 (1.1)	47 (1.0)	

Table 2. Average age of patients with and without ED.

Elastofibroma dorsi	No (n=8291)	Mean $\pm$ SD (Min-max)	42.0 $\pm$ 15.2 (0-94)
		Median (IQR)	41 (30-52)
	Yes (n=87)	Mean $\pm$ SD (Min-max)	52.0 $\pm$ 14.7 (20-85)
		Median (IQR)	52.5 (40.25-63)
	p-value		<0.001

Table 3. Age and gender of patients with unilateral and bilateral ED.

		Female/Male		Age
Elastofibroma dorsi	n (%)	n (%)		Mean $\pm$ SD (Min-max)
Unilateral	60 (69.0)	29 (48.3%) / 31 (51.7%)		50.3 $\pm$ 14.3 (20-85)
Bilateral	27 (31.0)	12 (44.4%) / 15 (55.6%)		55.9 $\pm$ 15.3 (24-81)
p-value		0.737		0.101

All lesions were evaluated separately, and the mean ED thickness was measured as 8.2 ± 6.5 mm (range 2.9-54.2). It was measured as 8.95 ± 5.81 mm (3-27) on the right and 7.60 ± 6.89 mm (2-54) on the left side. A positive correlation was determined between mean ED thickness and age ($p = 0.001$ and $p = 0.041$ for right and left, respectively). There was no significant difference in mean ED thickness regarding the side and gender ($p = 0.086$ and $p = 0.621$, respectively).

The mean ED density was 18.4 ± 24.3 (-58-61.5) HU. It was 14.58 ± 27.70 (-58.6-61.5) HU on the right, and 21.28 ± 21.15 (-32-57.7) HU on the left side, with no significant difference between them ($p = 0.243$). When the right and left side densities were compared according to the genders, no significant difference was determined ($p = 0.947$ and $p = 0.792$ for the right and left, respectively). In patients with ED, there was no significant correlation between density and size for both sides ($p = 0.172$ and $p = 0.352$ for right and left, respectively). The left-sided ED frequency was significantly higher than the right-sided ED ($p = 0.048$).

Almost all ED patients had lesions isodense with adjacent muscle structures and contained linear hypodense areas on imaging. When the initial imaging reports of the patients were reviewed, none of the lesions were mentioned in the first radiological report. All patients with ED had non-contrast imaging examinations. Therefore, the enhancement feature could not be evaluated. When evaluated from the clinical point of view by retrospectively reviewing the hospital electronic medical record system, none of the patients with ED had dorsalgia or shoulder pain, which are the main symptoms of this entity.

Discussion

ED is a benign, rare soft tissue tumor with a characteristic appearance and localization on CT and was first defined by Jarvi and Saxen in 1961 [10,11]. The typical location is at the level of the lower end of the scapula between the latissimus dorsi and serratus anterior muscles. Elastofibroma develops in this localization in 99% of cases [4]. However, different localizations, including the olecranon, tricuspid valve, interspinal, ligamentum flavum, foot, greater trochanter, deltoid, axilla, intra-articular, intraspinal and ischial tuberosity, stomach, cornea, and me-

diastinum, have been reported in the literature [12-21]. Nagamine et al diagnosed elastofibroma in the olecranon and ischial tuberosity in addition to the subscapular level, which is classically located in the patient group, and associated this condition with genetic and structural factors [4]. In elastofibromas observed outside the classical localization described in the literature, the diagnosis was made almost entirely pathologically.

A retrospective study by Brandser et al, in which they evaluated 268 patients describing the prevalence and imaging features of ED in 1998, indicated the prevalence of ED as 2% in the group of patients over 60 years of age. Our study determined the prevalence of ED as 1.03%. We associated this result with the lower mean age (42.1 ± 15.2) of our entire study population compared to the aforementioned study [20,22]. In the study of Tepe et al, the prevalence of ED was 2.73%, and the mean age of patients with and without ED was 68.2 and 56.1 years, respectively [12]. In our study, this result was 42.0 and 52.0, respectively, and similarly, the mean age of our patients without ED population was low. In their study involving the Jarvi and Lansimies autopsy series, they reported the prevalence of ED as 16%. This difference with the CT series can be attributed to the small size of the lesions in the second study and the macroscopic definition of some lesions as only a streak in the fascia [23].

In studies with large series in which ED was reported in the literature, the female gender was generally more common [6,12,22-25]. Although there was no statistically significant difference in our study, there were 46 male patients (52.9%) in total with predominance. In the study of Chandrasekar et al, similar to our study, there was male predominance with the rate of 80% [26]. The tendency of ED to be bilateral has been detected in many studies, and it has been reported at a rate of approximately 12-73% [4-6,27-29]. In our study, the rate of bilateral lesions was 31%. There was no significant difference between the mean age of patients with bilateral lesions and those with unilateral lesions, and our result was consistent with the literature [12].

A significant relationship was observed between ED thickness and age on both sides in our study. In the literature, a similar relationship was reported by Tepe et al only for the right side [12]. However, there was no significant

difference in ED thicknesses when compared regarding the side or gender in our study. Moreover, the mean density of ED was measured as 18.4 ± 24.3 HU on the right and 21.28 ± 21.15 HU on the left side. Negative HU values of fat density depend on the fat component in the tumor, and according to our knowledge, such a measurement has not been evaluated before in the literature. None of the patients with ED in our study were diagnosed with ED at the initial report. Although the diagnosis of ED is easily made with CT, it can be overlooked in the first report, similar to other studies in the literature [12,22,31].

Diagnosis of ED is made radiologically by MRI and CT according to the typical location of the tumor and imaging characteristics [7,11,22,30]. Lesions with a fibrous component and linear adipose tissue in typical periscapular localization, especially when bilateral, can be diagnosed as ED accurately without biopsy. Adipose tissue has a hypodense appearance on CT and T1-T2 hyperintense signal on MR. The imaging feature of fibrous tissue is similar to muscle tissue in CT and MRI [12]. The sensitivity of CT in the diagnosis of ED is lower than MRI. After intravenous gadolinium, the mass shows minimal-moderate or significant contrast material uptake [3].

ED excision is not recommended in asymptomatic cases since no malignant transformation has been described before [6,7,31]. When the biopsy is required, open biopsy or at least core needle biopsy is recommended. Fine needle biopsy is not recommended due to the hypocellular nature of the tumor [7]. Tumors that may be located in this region in the differential diagnosis include fibroma (in cases with fibrous predominance), aggressive fibromatosis, metastasis, hemangioma, and atypical lipoma and liposarcoma in patients with lipomatous component [7,33-36]. These tumors can be distinguished from ED radiologically by CT and MRI as they do not demonstrate the laminar pattern of ED.

Microtrauma, elastin degeneration, enzyme defect, genetic factors, and vascular insufficiency have been suggested in the etiology [36-41]. More than 50% of ED cases are asymptomatic. However, some patients may experience swelling, shoulder stiffness, friction noise with shoulder movements, and pain [34,42,43]. Rarely, clinical findings may change, and ED may be mistakenly confused with subacromial bursitis or rotator cuff tear [27].

The treatment of ED is surgical excision. A surgical operation is recommended when the tumor is greater than 5 cm, the patient is symptomatic and radiological findings are compatible with malignancy [44]. A conservative approach can be preferred if the patient is asymptomatic, and the tumor can be followed up clinically. Radiotherapy can be used in high-risk patients for surgery [14].

None of the patients with ED had either dorsalgia or shoulder pain, which are the main symptoms of ED. The condition might be related to the small mean size of ED and only six patients had a tumor size of greater than 2 cm. Patients diagnosed with ED are under follow up by thoracic surgery clinic in our hospital but there are no patients requiring surgery yet.

The main limitation of our study was the lack of histopathological verifications of the patients.

In conclusion, ED is a slow-growing, non-neoplastic soft tissue tumor with a typical periscapular location. Being aware of this entity and its radiologic features will prevent the need for advanced diagnostic evaluation. The characteristic appearance of these lesions, typically bilateral, will reduce unnecessary biopsy and surgery with increased radiologist awareness of their recognition.

Declaration of conflicting interests

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Ethics approval

Approval from the Institutional Review Board of Haseki Research and Training Hospital was obtained without the requirement for patient informed consent because of the retrospective origin of the study (Decision number: 2020-131).

Authors' contribution

BKY,SÖ,Tİ,TSC,NG,BA; material preparation, data collection and analysis. BKY; the first draft of the manuscript was written by. BKY, RT: revised the final version of the manuscript. All authors read and approved the final manuscript. All authors contributed to the study conception and design.

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Original Article

Impact of COVID-19 pandemic on non-small cell lung cancer: a retrospective study

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ABSTRACT

Background: The most important treatment method in non-small cell lung cancer (NSCLC) is surgery. Detection of the disease at an early stage is one factor that determines the prognosis. This study investigates the impact of the novel coronavirus disease pandemic on the diagnosis and surgical treatment of NSCLC.

Materials and Methods: Between March 2019 and March 2021, a total of 163 patients (142 males, 21 females, mean age 62.3 ± 7.8 years; range, 39 to 78 years) diagnosed with NSCLC who underwent surgery were retrospectively analyzed. The patients were divided into two groups according to COVID-19 pandemic period such as before (Group 1) and after (Group 2) March 2020. Demographic characteristics, symptoms, diagnostic method, pathological diagnosis, stage, surgical treatment method, and developing complications of patients were recorded.

Results: During the investigation of COVID-19, NSCLC was detected among 16.8% ($n = 17$) of the patients who underwent surgery. The number of NSCLC patients who underwent surgery during the pandemic period increased by 62.9% compared to the pre-pandemic period. ($p=0.338$). Despite patients with Stage 1 disease (30.6% vs. 9.9%, $p = 0.007$) and the rate of open surgery was significantly decreased (82.3% vs. 52.5%, $p < 0.001$) during the pandemic period for patients diagnosed with NSCLC, those who underwent surgery were most frequently determined on stage 1A when compared with the pre-pandemic period. In contrast, the video-assisted thoracoscopic surgery approach increased (17.7% vs. 47.5%, $p < 0.001$), and a statistically significant difference was found between the distributions of surgical procedures.

Conclusions: In the novel coronavirus pandemic, thorax computed tomography used for lung assessment affected the detection rate of NSCLC. The importance of VATS pulmonary resections increased in this period, thus shortening the hospital stay of the patients.

Keywords; COVID-19, non-small cell lung cancer, pandemic, thoracic surgery.

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Introduction

The World Health Organization declared the novel coronavirus disease (COVID-19) a global pandemic on March 11, 2020 [1]. Most non-small cell lung cancers (NSCLC) are diagnosed in the late stages (Stages IIIB-IV) when 5-year survival is low (less than 36%) [2]. Radiologic imaging techniques such as chest X-ray and thorax computed tomography (CT) are not only used for the detection of asymptomatic early-stage lung cancer but also are used as initial screening tests to diagnose COVID-19 disease and helps for early detection of lung cancer. During the pandemic period, there was a decrease in the number of CT applications in the general population [3]. Especially during the periods when the number of COVID-19 cases has peaked, the rate of radiological examinations have increased in patients with respiratory system complaints. Lesions detected in thorax CT imaging in COVID-19 may appear as a ground-glass opacity, which is also highly similar to the lesions detected in lung cancer [4]. The similarities of the detected lesions suggest may affect the diagnosis and surgical treatment of lung cancer in the COVID-19 pandemic.

In lung cancer, which has more opportunities to be detected in the pandemic process, new decisions were needed for the treatment plan. Guidelines for surgical treatment of lung cancer recommend to defer 3-month in case of early-stage disease and triage of patients for surgery, taking into account the safety of patients and surgical teams [5]. Despite pandemic, oncological surgeries were continued uninterruptedly by providing isolation and protection measures in our clinic. In this study aimed to evaluate the impact of the COVID-19 pandemic on patients with NSCLC who underwent surgical treatment.

Materials and Methods

A total of 163 (142 males, 21 females, mean age 62.3 ± 7.8 years; range, 39-78 years) patients who were diagnosed with NSCLC and underwent surgery between March 2019 and March 2021 were included in our study. The data were analyzed retrospectively.

The patients were divided into two groups. Group 1 was patients with NSCLC who were surgically treated during the one year preceding the pandemic period between March 11, 2019, and March 10, 2020. Group 2 patients with NSCLC were surgically treated during the COVID-19 pandemic between March 11, 2020,

and March 11, 2021. Sixty-two patients were considered in Group 1 and 101 patients were considered in Group 2. Demographic characteristics, symptoms, diagnostic method, pathological diagnosis, stage, surgical treatment method, and developing complications of patients were recorded separately for Group 1 and Group 2. The patients in the group 2 group were evaluated in detail regarding COVID-19 disease. The cases in which NSCLC was detected during investigation for suspected COVID-19 and delayed due to COVID-19 disease in the preoperative period were also evaluated for the effects of COVID-19 on morbidity and mortality in the postoperative period.

This study was conducted with the approval of the Ministry of Health Scientific Research Platform (No:2021-05-17T13_39_03) and our hospital's ethics committee (approval number: 2021/277; approval date: May 20, 2021) approval. Informed consent was obtained from all patients.

Statistical Analysis

Collected data were analyzed using SPSS Statistics for Windows, version 23.0 (IBM Corp., Armonk, NY, USA). The suitability of the data to the normal distribution was examined with the Kolmogorov-Smirnov test. Chi-square and Fisher's Exact tests compared categorical variables according to groups. The Kruskal Wallis test was used to compare quantitative data not normally distributed according to groups of three or more. Mann Whitney U test was used to compare data that did not show normal distribution according to paired groups. Analysis results were presented as mean $\pm$ standard deviation and median (minimum-maximum) for quantitative data and frequency (percent) for categorical data. The significance level was taken as $p < 0.050$.

Results

When the quantitative data were compared according to the groups, no statistical difference was found between the mean age of both groups [62.6 ± 8.1 (42-78) years vs 62.2 ± 7.6 (39-78) years]. No statistically significant difference were detected in terms of tumour size distribution, SUV max values, chest tube pulling time between groups (Table 1). While no statistically significant difference was detected between groups in terms of admission complaints, the rate of NSCLC detected during screening studies increased from 14.5% to 34.7%. In Group 2, the rate of detecting lung lesions on thorax

CT during the COVID-19 evaluation was 16.8% (n = 17). Surgery was postponed for 1 month among 6 patients who were infected or exposed with COVID-19. The number of NSCLC patients who underwent surgery during the pandemic period (Group 2) increased by 62.9% compared to the pre-pandemic period. Median length of hospital stay was detected shorter in Group 2 [6(2-48) days vs 5(1-21) days, $p = 0.014$].

The most common diagnostic method for both groups was trans-thoracic tru-cut biopsy (TT-TCB) (53.2% vs 61.4%), and the most common histopathological diagnosis was adenocarcinoma (58.1% vs 52.5%). The rate of invasive mediastinal staging before resection was increased (45.2% vs. 68.3%, $p = 0.006$). We thought that the probable reason for this increase was intraoperative mediastinal invasive staging (VATS/thoracotomy) (24.2% vs. 46.5%) methods apart from mediastinoscopy ($p = 0.006$). In the pre-pandemic period, NSCLC patients who underwent surgery (Group 1) most frequently were stage 1A; however, it was determined that the rate of Stage 1 disease decreased significantly (30.6% vs. 9.9%, $p = 0.007$) during the pandemic period, and the rate of disease detected in Stage 3B increased significantly (1.6% vs. 10.9%, $p = 0.007$). It was found that a significant difference in the distribution of tumor sizes between the groups. It was determined that T1b size tumors decreased (24.2% vs 7.9%) T1c size tumors increased (14.5% vs 28.7%) ($p = 0.042$). There was a significant difference between the distribution of tumor positive lymph node stations, and a significant increase in the number of N2 positive patients (3.2% vs. 17.8%, $p = 0.028$) (Table 2).

It was determined that the rate of open surgery decreased (82.3% vs. 52.5%, $p < 0.001$). Besides that, the video-assisted thoracoscopic surgery (VATS) approach, increased (17.7% vs. 47.5%) and a statistically significant difference

was found between the distributions of surgical procedures and applied surgical procedures ($p < 0.001$) (Table 3).

There was no significant difference between the complication rates for both groups. However, 5 patients were passed away during the pandemic period. One patient died on the 1st postoperative day due to acute myocardial infarction. The other 4 patients died in the hospital in the first 30 days due to secondary effects after pneumonia of unknown cause. The length of hospital stay during the pandemic period was shortened. ($p = 0.014$)

During the pandemic periods when the number of COVID19 cases peaked, the data were analyzed quarterly to compare the number of patients with NSCLC detected with the previous year. When comparing the number of patients with NSCLC who underwent surgery, it was determined that the number of cases increased by 100% in the March-May period, by 31.25% in the June-August period, by 38.46% in the September-November period, and by 80.95% in the December-February period. It was determined that the proportional increases in the first and last periods were correlated with the peak periods of the daily number of cases shared on the Ministry of Health COVID-19 Information Platform (6) (Figure 1). However, this correlation was not statistically significant.

A statistically significant difference was found between the distribution of the stages among groups in the first 3 months ($p = 0.024$). Patients with Stage 1A who underwent surgery was significantly reduced (58.3% vs 16.7%, $p = 0.024$). There was no statistical difference between groups in terms of the distributions of pathology, stage, surgery, and procedure variables in the June-August period. A statistically significant increase was detected in VATS rates in the September-November (23.1% vs. 66.7%, $p = 0.005$) and December-February (33.3% vs. 63.2%, $p < 0.001$) periods according to the surgical procedures applied (Table 4).

Table 1. Comparison of quantitative variables by groups.

	Group 1		Group 2		Test Stat.	p
	Mean $\pm$ SD	Median (min - max)	Mean $\pm$ SD	Median (min. - max)		
Age (year)	62.6 $\pm$ 8.1	62 (42 - 78)	62.2 $\pm$ 7.6	63 (39 - 78)	3067.0	0.827
Tumor Size (mm)	37 $\pm$ 24.5	34 (2 - 130)	42.9 $\pm$ 24.5	37 (2 - 135)	3638.0	0.083
SUVmax	9.4 $\pm$ 6.5	8.5 (0 - 34.8)	10.3 $\pm$ 7.1	9.1 (0 - 41.3)	3341.0	0.473
Tube Removal (day)	4.5 $\pm$ 2.7	4 (1 - 15)	4.3 $\pm$ 3.3	3 (1 - 20)	2447.0	0.315
Hospital (day)	7.6 $\pm$ 6.3	6 (2 - 48)	6.1 $\pm$ 3.9	5(1 - 21)	2419.5	0.014

Test Statistics: Mann Whitney U test statistic. SUVmax: Maximum standardized uptake value of Positron Emission Tomography).

Table 2. Comparison of categorical variables by groups.

	Group 1 (n=62)	Group 2 (n=101)	Test stat. (χ^2)	p
Sex				
Male	56 (90.3)	86 (85.1)	0.916	0.338
Female	6 (9.7)	15 (14.9)		
Symptom				
Dyspnea	9 (14.5)	11 (10.9)	9.409	0.152
Chest Pain	9 (14.5)	11 (10.9)		
Cough	18 (29)	23 (22.8)		
Hemoptysis	5 (8.1)	9 (8.9)		
Screening	9 (14.5)	35 (34.7)		
Others	12 (19.4)	12 (11.8)		
Diagnosis				
Bronchoscopy	9 (14.5)	12 (11.9)	0.185	0.607
TT-TCB	33 (53.2)	62 (61.4)		
Intraoperative	20 (32.3)	27 (26.7)		
Pathology				
Adenocarcinoma	36 (58.1)	53 (52.5)	6.201	0.185
SCC	22 (35.5)	41 (40.6)		
Others	4 (6.4)	7 (7)		
Mediastinal invasive staging				
Mediastinoscopy	13 (21)	22 (21.8)	10.140	0.006
Intraoperative	15 (24.2)	47 (46.5)		
None	34 (54.8)	32 (31.7)		
Stage				
1A	19 (30.6)a	10 (9.9)b	17.645	0.007
1B	9 (14.5)	21 (20.8)		
2A	5 (8.1)	5 (5)		
2B	11 (17.7)	27 (26.7)		
3A	14 (22.6)	25 (24.8)		
3B	1 (1.6)a	11 (10.9)b		
4	3 (4.8)	2 (2)		
Tumor				
<=10(T1a)	4 (6.5)	4 (4.0)	13.098	0.042
11-20 (T1b)	15 (24.2)a	8 (7.9)b		
21-30 (T1c)	9 (14.5)a	29 (28.7)b		
31-40 (T2a)	12 (19.4)	16 (15.8)		
41-50 (T2b)	10 (16.1)	14 (13.9)		
51-70 (T3)	7 (11.3)	16 (15.8)		
>70 (T4)	5 (8.1)	14 (13.9)		
Node				
None	48 (77.4)	65 (64.4)	9.120	0.028
Hilar	12 (19.4)	16 (15.8)		
Ipsilateral Mediastinal	2 (3.2)a	18 (17.8)b		
N3	0 (0)	2 (2)		
Metastasis				
No	59 (95.2)	99 (98)	---	0.369 ^F
Yes	3 (4.8)	2 (2)		

χ^2 : Chi-square test statistic, F: Fisher's Exact test, a-b: No difference between groups with the same letter. SCC: Squamous Cell Carcinoma.

Table 3. Comparison of surgical variables by groups.

	Group 1 (n=62)	Group 2 (n=101)	Test stat. (χ^2)	p
Resection Type				
Lobectomy	45 (72.5)	87 (86.1)	11.755	0.302
RUL	17 (27.4)	25 (24.8)		
RLL	6 (9.7)	12 (11.9)		
RML	4 (6.5)	3 (3)		
RUBL	1 (1.6)	6 (5.9)		
RLBL	1 (1.6)	2 (2)		
LUL	11 (17.7)	23 (22.8)		
LLL	5 (8.1)	16 (15.8)		
Pneumonectomy	5 (8.1)	2 (2)		
Wedge	5 (8.1)	8 (7.9)		
Segmentectomy	3 (4.8)	2 (2)	0.165	0.684
Extended	4 (6.5)	2 (2)		
Side				
Right	37 (59.7)	57 (56.4)	0.165	0.684
Left	25 (40.3)	44 (43.6)		
Procedure				
Open	51(82.3)	53(52.5)	14.755	<0.001
VATS	11(17.7)	48(47.5)		
Complication*				
None	37 (59.7)	60 (59.4)	4.107	0.534
Prolonged air leakage	13 (21)	24 (23.8)		
Pneumonia	5 (8.1)	6 (5.9)		
AF	2 (3.2)	5 (5)		
Other	9 (14.5)	6 (5.9)		

χ^2 : Chi-square test statistic, *Multiple response. AF: Atrial Fibrillation, LLL: Left Lower Lobectomy, LUL: Left Upper Lobectomy, RLBL: Right Lower Bilobectomy, RLL: Right Lower Lobectomy, RML: Right Middle Lobectomy, RUBL: Right Upper Bilobectomy, RUL: Right Upper Lobectomy, VATS: Video-assisted Thoracoscopic Surgery.

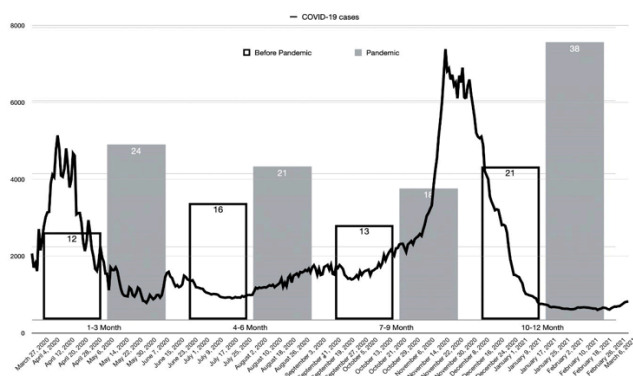


Figure 1. Comparison of the official case numbers obtained from the Ministry of Health's COVID-19 Information Platform and the patients who underwent surgery with the diagnosis of NSCLC in 3-month periods.

Discussion

The main findings of this study were when compared with the pre pandemic period, the number of patients who were incidentally diagnosed with lung cancer were increased during COVID-19 screening. In addition, minimally invasive techniques tend to be preferred

more. The rates of invasive mediastinal staging were increased, and the rate of surgery performed on stage 1A patients decreased at the beginning of the pandemic and increased the risk of death due to postoperative pneumonia during the pandemic period.

While the rate of surgery applied to patients with Stage 1 NSCLC decreased compared to the pre-pandemic period, the rate of surgery applied to Stage 3 patients increased. While symptoms occurring in stage 3 in the pre-pandemic period is not taken into account by patients and are included in advanced stage patient groups who lost the chance for surgery, it is clear that the symptoms that occur during the pandemic period are taken into account due to the risk of COVID-19. Besides, it was shown that the T1b patient group who underwent surgery decreased, and the T factor increased compared to the previous year. At the same time, video thoracoscopic lung resection rates were increased in our clinic by changing surgical procedures according

Table 4. Comparison of Pathology, Stage, Surgery and Procedure variables by groups in quarterly periods

	March-May			June-August			September-November			December-February		
	Group 1	Group 2	Test stat. p	Group 1	Group 2	Test stat. p	Group 1	Group 2	Test stat. p	Group 1	Group 2	Test stat. p
Pathology												
Adenocarcinoma	8 (66.7)	12 (50)		11 (68.8)	9 (42.9)		4 (30.8)	10 (55.6)		13 (61.9)	22 (57.9)	
SCC	3 (25)	11 (45.8)	1.543 0.462	5 (31.3)	11 (52.4)	2.826 0.243	9 (69.2)	6 (33.3)	4.482 0.106	5 (23.8)	13 (34.2)	4.695 0.320
Others	1 (8.3)	1 (4.2)		0 (0)	1 (4.8)		0 (0)	2 (11.1)		3 (14.3)	3 (8.4)	
Stage												
1A	7 (58.3) ^a	4 (16.7) ^b		4 (25)	1 (4.8)		2 (15.4)	1 (5.6)		6 (28.6)	4 (10.5)	
1B	0 (0)	3 (12.5)		0 (0)	5 (23.8)		2 (15.4)	4 (22.2)		7 (33.3)	9 (23.7)	
2A	1 (8.3)	1 (4.2)		0 (0)	0 (0)		1 (7.7)	2 (11.1)		3 (14.3)	2 (5.3)	
2B	1 (8.3)	6 (25)	14.581 0.024	4 (25)	3 (14.3)	9.499 0.091	4 (30.8)	5 (27.8)	3.222 0.781	2 (9.5)	13 (34.2)	12.328 0.055
3A	1 (8.3)	6 (25)		8 (50)	9 (42.9)		3 (23.1)	5 (27.8)		2 (9.5)	5 (13.2)	
3B	0 (0)	4 (16.7)		0 (0)	2 (9.5)		1 (7.7)	0 (0)		0 (0)	5 (13.2)	
4	2 (16.7) ^a	0 (0) ^b		0 (0)	1 (4.8)		0 (0)	1 (5.6)		1 (4.8)	0 (0)	
Resections												
Lobectomy	9 (75)	20 (83.3)		9 (56.2)	20 (95.2)		11 (84.6)	16 (83.3)		16 (76.2)	32 (84.2)	
Pneumonectomy	2 (16.7)	1 (4.2)		3 (18.8)	0 (0)		0 (0)	0 (0)		0 (0)	1 (2.6)	
Wedge	0 (0)	2 (8.3)	14.100 0.119	1 (6.3)	1 (4.8)	15.043 0.130	0 (0)	2 (11.1)	8.184 0.317	4 (19)	3 (7.9)	7.408 0.686
Segmentectomy	0 (0)	0 (0)		2 (12.5)	0 (0)		0 (0)	1 (5.6)		1 (4.8)	1 (2.6)	
Extended	1 (8.3)	1 (4.2)		1 (6.3)	0 (0)		2 (15.4)	0 (0)		0 (0)	1 (2.6)	
Procedure												
Open	12 (100)	19 (79.2)	2.903 0.234	15 (93.8)	14 (66.7)	4.236 0.120	10 (76.9) ^a	6 (33.3) ^b	10.671 0.005	14 (66.7) ^a	14 (36.8) ^b	19.304 <0.001
VATS	0 (0)	5 (20.8)		1 (6.3)	7 (33.3)		3 (23.1) ^a	12 (66.7) ^b		7 (33.3) ^a	24 (63.2) ^b	

(Chi-square test statistic was used as test statistic, a-b: No difference between groups with the same letter. LLL: Left Lower Lobectomy, LUL: Left Upper Lobectomy, RLL: Right Lower Bilobectomy, RLL: Right Lower Lobectomy, RML: Right Middle Lobectomy, RUBL: Right Upper Bilobectomy, RUL: Right Upper Lobectomy, VATS: Video-assisted Thoracoscopic Surgery)

to guidelines published in the pandemic and decreasing the rate of open surgery [7, 8]. While the rate of thoracoscopic surgery was 17% before the pandemic, it was increased to 47.5% during the pandemic period. For this reason, it aims to protect the surgical team and health workers and shorten the patients' hospitalization period.

It is obvious that surgery performed on a more risky patient group in terms of systemic diseases compared to the pre-pandemic period carries a higher risk for COVID-19. Infected with or exposed to COVID-19 were detected in 6% of the patients during the preoperative preparation phase, and to reduce the risks of COVID-19, surgeries were postponed for 1 month.

In the study conducted by Li et al, it was recommended to focus on clinical and pathological findings that require emergency surgery during the pandemic period and perform absolute surgery in resectable lung tumors with significant airway involvement, accompanied by massive hemoptysis and causing severe dyspnea [9]. It was recommended not to apply procedures that allow aerosol propagation, such as bronchoscopy and needle aspiration (EBUS-TBNA) guided by endobronchial ultrasound during the pandemic period [10]. Our findings revealed that the rate of diagnostic bronchoscopy during the pandemic period was 11.9% and without change with the previous period. Nevertheless, TT-TCB was the most frequently used diagnostic method for both groups. Cafarotti et al [11] published an algorithm to determine the urgency classification for patients with lung cancer which was based on TNM staging, the risk of cancer progression and the risks of COVID-19 are considered together. Adenocarcinoma of the lung and squamous cell carcinoma has the potential to progress from early-stage IA to stage IIIB in 1.17 and 2.5 years, respectively [12]. Quebec Lung Cancer Network also shared its recommendations for the surgical treatment of lung cancer during the pandemic [13]. It was emphasized that priority should be given to selected treatments for stage I-III NSCLC. In our study, it was revealed that there was a significant decrease in the T1b patient group who underwent surgery, and surgery was performed on the patients in the T1c patient group, in accordance with the guideline. The fact that the VATS procedure was preferred more as an invasive mediastinal staging method during the pandemic period explains the increase in the number of Stage IIIB patients who underwent surgery.

The high demand for radiological evaluation during the pandemic had a direct impact on patients diagnosed with confirmed or suspected lung cancer [14]. Opinions were advocating that the diagnosis and treatment of lung cancer due to the similarity of ground-glass opacity findings with infiltrates detected in thorax CT in COVID-19 pneumonia, the postponement of cancer screening studies was delayed during the pandemic period [15]. In our study, there was an increase in the number of patients who underwent surgery with the diagnosis of NSCLC. During the pandemic, NSCLC was detected at a rate of 16.8%, with radiological examinations performed with the suspicion of COVID-19. Especially during the peak periods of COVID-19 cases, the number of NSCLC cases that underwent surgery has increased. Compared to the previous year, the number of surgical cases increased by 100% in the Mar-May period and by 80.95% in the Dec-Feb period. In addition, diagnostic procedures such as pulmonary function testing or bronchoscopy, or CT-guided transthoracic biopsy, some preoperative studies were delayed, or certain cases were directly avoided, and this was a strategy that was considered inevitable in line with the measures at the time [16]. It was recommended that patients with nodules with ground-glass opacity (<50% solid component), <2 cm nodules or lung cancer, slow-growing (indolent) histology (carcinoid or slow-growing nodules), and those who may need long-term intensive care should be postponed for 3 months [5]. In accordance with this strategy, according to the T factor in the surgeries performed, it was shown that while the number of patients with T1b tumors decreased in the pandemic period compared to the previous year, there was an increase in the number of patients with T1c tumors. However, when the stages of lung cancer patients who underwent surgery during the pandemic period were compared with the previous year, it was statistically revealed that surgery applied to stage 1A patients was reduced. To keep the surgical time short, especially intraoperative (thoracotomy/VATS), the invasive staging was performed in accordance with the criteria for mediastinal invasive staging [17]. Therefore, it was observed that there was a significant increase in the rate of positive tumor detection in the ipsilateral lymph nodes during the pandemic period in our study.

Since lung surgery is the surgery with the highest aerosol formation, it had a special place in terms of sur-

gical procedures to be applied in the COVID-19 pandemic [7]. Even if all personal protective measures and necessary operating room conditions are provided, it increases the risk of smog, aerosol formation, and spread of infection, especially due to the effect of the energy devices used. Therefore, in all surgical branches, it has been suggested that minimally invasive surgical procedures should be preferred in cases where surgery is unavoidable [18]. Vigneswaran et al argued that aerosolized virus could not cause active infection through the skin [19]. However, we preferred to perform surgical procedures in our clinic in accordance with the guidelines since the situation in question is absorption from the mucous membranes and spread by inhalation. Therefore, it was shown that VATS, which was applied at a rate of 17.7% in the pre-pandemic period, increased to 47.5% during the pandemic period, and this rate was a statistically significant increase. It was determined that the length of hospital stay decreased significantly, especially in correlation with the increasing minimally invasive surgical practice. Notwithstanding, there was no significant difference in surgical complications. Nevertheless, while no mortality was observed in patients who underwent surgery in the pre-pandemic period, mortality occurred in 5 patients during the pandemic period. One patient died on the 1st postoperative day due to acute myocardial infarction. Although the COVID-19 PCR test is negative, in the other 4 patients, COVID-19 pneumonia could not be excluded in computed tomography images. During the pandemic period, the length of hospital stay was statistically significantly shortened. Villena-Vargas et al similarly stated that the duration of hospital stay was shortened during the pandemic period, and this may affect the risk of hospital infection [20].

Our study has some limitations such as it was a single-center retrospective study. Because no surgically applicable lung cancer patient is delayed or referred to non-surgical methods, the effects of the COVID-19 pandemic on the non-surgical NSCLC patient group could not be evaluated. Besides, in the first period of the pandemic, the preoperative COVID-19 exclusion procedures and the difficulties in controlling the postoperative COVID-19 transmission risks directly affected the results.

In conclusion, the novel coronavirus disease, which affected people in all areas of life in the early stages

of the pandemic in our country, directly affected the increase in the detection of patients with NSCLC. The importance of VATS pulmonary resections increased in this period, thus shortening the hospital stay of the patients. It is also thought that up-to-date guidelines can be created with studies to evaluate the effects of COVID-19 vaccine applications on the treatment processes of patients with NSCLC in the future.

Declaration of conflicting interests

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Ethics approval

This study was conducted with the approval of the Ministry of Health Scientific Research Platform (No: 2021-05-17T13_39_03) and Ondokuz Mayıs University Faculty of Medicine ethics committee (approval number: 2021/277; approval date: May 20, 2021).

Authors' contribution

SG: conceptualization, methodology, project administration, writing-original draft, writing-review and editing, visualization, validation, ATS: data curation, formal analysis, investigation, software, writing-review and editing, supervision, validation, MGP: data curation, investigation, formal analysis, YB: data curation, investigation, formal analysis, BC: supervision, validation and writing-review and editing, NGT: data curation and investigation, CI: data curation and investigation, AB: supervision, validation and writing-review and editing. All authors actively contributed to discussion of the results from the study and reviewed and approved the final version to be released.

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Original Article

Pulmonary hydatid cyst disease: a retrospective study

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ABSTRACT

Background: Although the disease progression can be silent, the pulmonary hydatid cyst (HC) may be considered a life-threatening disorder. The mainstay of pulmonary HC treatment is still surgical removal of the cyst. In the current study, we evaluated the results of surgery in patients with pulmonary HC.

Materials and Methods: Thirty pulmonary HC patients who underwent surgery between 2007 and 2020 were analyzed retrospectively. The symptoms, cyst features, surgical procedures, and postoperative findings were recorded. The patients were divided into two subgroups: complicated cyst hydatid (CHC) and simple hydatid cyst (SHC).

Results: Thirteen cases were diagnosed as complicated HC with rupture into the pleural or bronchial cavity (CHC group, 43.4%), whereas 17 cases (56.6%) involved intact cysts (SHC group). Cystotomy and capitonnage were performed in the majority of the patients (n = 24, 80.0%). No mortality was observed during the one-year follow-up period.

Conclusions: Surgical management is an effective and safe method for the treatment of pulmonary HC.

Keywords: pulmonary hydatid cyst, complicated cyst, surgery, outcomes

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Introduction

Echinococcosis is a zoonotic disease caused by *Echinococcus* species in the cestode class. It is an important cause of morbidity and mortality in many parts of the world [1]. Lung involvement with hematological spread is one of the most common accompanying conditions, and it may present with extrapulmonary involvement in this region due to the close proximity of the lung [1-3]. The aim of the present study was to compare our results with the relevant literature.

Materials and Methods

Thirty patients that were diagnosed and operated on between 2007 and 2020 due to simple hydatid cyst (SHC) or complicated hydatid cyst (CHC) were evaluated respectively. The study took place at Tokat Government Hospital. The clinical manifestation, age, sex, symptomatology, additional organ involvement, diagnostic methods, surgical interventions and preoperative and postoperative complications, duration of hospital stay, and one-year follow-up findings were recorded.

After a detailed history was obtained, chest X-ray (Figures 1A-C) and thorax computed tomography (Figures 2A,B) were used to establish the diagnosis. Serologic tests were not used routinely due to their low diagnostic value.



Figure 1. The PA x-ray show; a single cyst in superior zone of left lung (black arrows) (A), bilateral hydatid cysts; multiple hydatid cysts in the right lung (white arrows) (B), bilateral pulmonary hydatid cysts (white arrows) (C).

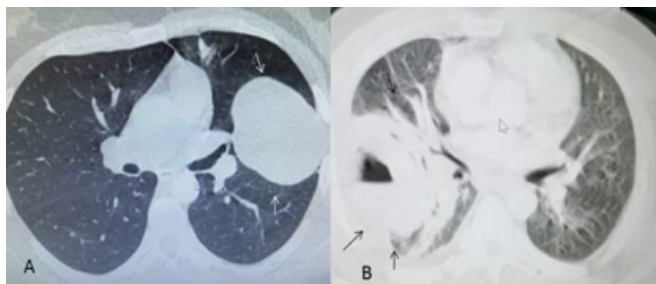


Figure 2. The chest CT show; a giant hydatid cyst (white arrows) (A), a ruptured hydatid cyst (black arrows) (B).

Rupture of the hydatid cyst into the pleural cavity or bronchus was defined as CHC and an intact cyst was defined as SHC. In complicated cysts, tube thoracostomy was performed.

The routine surgical procedures were cystotomy and capitonnage. Posterolateral thoracotomy was performed in all patients under general anesthesia. Compresses soaked with hypertonic NaCl (10%) as a scolicidal agent were placed around the cyst field to prevent spread. The cyst content was aspirated to decrease the intracystic pressure and the laminated membrane was removed with care. The cyst cavity was irrigated with saline solution with immediate aspiration and each sub-segment opening was sutured meticulously with absorbable sutures; then the cavity was obliterated from bottom to top. The pleural surface was finally sutured for prevention of any air leakage.

Wedge resection and enucleation were also performed using similar protective measures. (Figures 3A-D). Decortication and pleurectomy were the additional procedures performed for thickened pleural surfaces

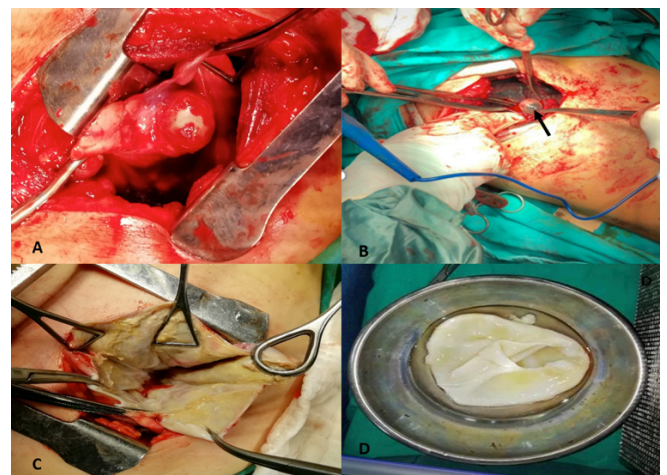


Figure 3. Peroperative images showing; wedge resection (A), enucleation of intact hydatid cyst (black arrow) (B), cystotomy-cappitonnage (C), laminated membrane (D).

No mortality or serious morbidity was observed in our study. Prolonged air leakage was observed in three patients (10%), atelectasis in two patients (6.6%), and superficial wound infection in one patient (3.3%). Post-operatively all CHC patients with a rupture received albendazole treatment with a dose of 10-15 mg/kg/day in 2 or divided doses. The treatment was continued intermittently for 3 months. In the monthly follow-up, liver function tests were performed to detect possible toxicity. The study was approved by the local ethics committee of TOGU Medical School (Approval No. 22-Kaek-111).

Statistical Analysis

All statistical evaluations were made using SPSS ver 22.00 (IBM, Armonk, NY, USA). Continuous variables were presented as mean $\pm$ standard deviation (SD) and ranges. Rates were expressed as percentages.

Results

The average age was 30.8 ± 17.3 . Sixteen patients (53.3%) were female and 14 (46.6%) were male. There were 17 (56.6%) SHC and 13 CHC (43.4%) patients in our case series. The main complaints were cough (40%), dyspnea (26.6%), sputum (26.6%), fever (16.6%), chest pain (16.6%), hydoptysis (6.6%), and hemoptysis (6.6%) in the entire patient population. Fever and productive cough (in 4 (30.7%) of the patients) were the most prominent complications in complicated cases (CHC). Four patients (13.3%) had liver involvement (primary lesion) and one patient had subcutaneous HC (3.3%). The imaging methods revealed multiple HCs in six patients (20.0%), bilateral HCs (10.0%) in three patients, and a single lesion in 21 patients (70%). The HCs were localized in the left lung in 13 patients (43.4%) and right-sided HC was detected in 14 patients (46.6%). Six cases (20.0%) required chest tube thoracostomy before surgery. A preoperative biopsy was not taken from any of these patients. Three patients had been operated on previously due to HC disease (10%). Pneumonia was detected in 5 (16.6%) patients that required antibiotic therapy during the preoperative period. Moreover, in the CHC group, 3 patients had empyema (23.1%) and 3 patients hydropneumothorax (23.1%). Additionally, cyst rupture into the bronchopulmonary tree was detected in 15.4% of these cases. The main findings of the patients are summarized in table 1.

The surgical procedures were cystotomy and capitonnage in 24 (80.0%) patients, wedge resection in 2 (6.6%) patients, and enucleation in 4 (13.3%) patients. Additional pleurectomy was applied in 4 patients (13.3%). Two of them had larger HC size (≥ 12 cm). Mean hospital stay was 15.46 ± 6.1 days (201 days) for CHC, whereas it was 7.35 ± 2.4 days for SHC (125 days).

The postoperative complications observed were atelectasis, prolonged air leakage, and superficial wound infection (in 6 (20.0%) of all patients). Two (6.6%) patients underwent rigid bronchoscopy due to atelectasis after the operation.

Table 1. The main findings of patients.

All patients (n=30)	n/%
Symptoms	
Cough	12/40.0
Dyspnea	8/26.6
Sputum	8/26.6
Fever	5/16.6
Chest pain	5/16.6
Hydoptysis	2/6.6
Hemoptysis	2/6.6
Accompanying Lesion	
Liver	4/13.3
Subcutaneous	3/3.3
Localization	
Multiple cyst	6/20.0
Bilateral cyst	3/10.0
Left lung	13/43.4
Right Lung	14/44.6
Complicated patients (n=13)	n/%
Fever	4/30.7
Productive cough	4/30.7
Hydropneumothorax	3/23.1
Empyema	3/23.1
Ruptured cyst	2/15.4

Discussion

The diagnosis of HC is based on suspicion depending on patient history, occupation, and imaging method. Serological tests such as direct hemagglutination, latex agglutination, immunoelectrophoresis and ELISA could be used for the diagnosis [4,5]. However, the exact diagnosis is performed preoperatively and confirmed via pathological examination of material. Chest radiography, computed tomography (CT), and magnetic resonance imaging (MRI) are very effective in identifying the cyst size and localization [4,6]. Although chest X-ray is the primary diagnostic method due to its simplicity, availability, and cost, it is insufficient for evaluating the surrounding tissue and the exact localization and associated complications.

The symptoms include non-specific cough, chest tightness, and pain, which can be seen in all lung diseases [1,7]. It can even be diagnosed incidentally as it may be asymptomatic until reaching large sizes. Acute symptoms may occur in complicated cysts. Spontaneous or traumatic rupture may be the cause of admission

to the emergency department with chest discomfort, hemoptysis, fever, and productive cough [8-11]. The main symptom in our series was cough. Fever and productive cough were prominent in complicated patients.

The prediagnosis can be performed with imaging methods such as chest X-ray, CT, and MRI and can be strengthened with serological tests [4-6]. However, seronegative results are highly probable and therefore not essential [12,13]. In particular, direct chest X-ray gives information about the general structure of the cyst, but MRI and CT provide clearer data for the exact location, adjacent anatomical positions, and surgical planning [4-6]. Chest radiography was used as the first imaging test in our series and thorax CT was performed in all. We did not routinely use thorax MRI and serological tests in our clinic. It is important to examine the cyst material for a definitive diagnosis. However, needle aspiration biopsy is not recommended due to the risk of rupture of the infected cyst content and the formation of new cysts in the same area or anaphylactic shock. However, we performed thoracentesis for complicated cysts with acute onset symptoms that ruptured into the pleura.

The mainstay treatment for pulmonary hydatid cysts is surgery. The basic methods include excision of the cyst while preserving the tissue. The surgical technique is based on closure of air leakages from subsegmental/segmental bronchi and removal of the cyst and its contents [14]. Although lung-sparing surgery is recommended as much as possible, tissue loss may be inevitable with segmentectomy and lobectomy, depending on the extent of the cyst and the severity of complications [15,16]. This risk is slightly higher with large cysts. Cysts greater than 10 cm in diameter are defined as gigantic and have been associated with an increased risk of complications. In many aspects such as hospital duration, mean chest tube removal time, and especially postoperative prolonged air leakage large cysts are considered a different entity when compared to smaller cysts. In large cysts non-capitonnage is used as a surgical method. Variable high morbidity rates have been reported in different series; however, according to Özpolat et al, regardless of the size and surgical method surgery provides excellent outcomes [17,18]. Pulmonary hydatid disease and liver disease may occur simultaneously in 4% to 25% of cases [19]. In patients simultaneous lung and liver hydatid disease may be managed through a transdiaphragmatic approach or in some cases via a

thoracoabdominal approach. This procedure is safe and spares the patient from undergoing another surgical intervention.

Currently, minimally invasive treatment in thoracic surgery is being used extensively, including in surgery for hydatid cysts. When compared to open surgery, it has obvious advantages such as less postoperative pain, fast recovery, and reduced postoperative complications [20].

When the decision concerning surgery is made carefully and meticulous planning is performed with preoperative imaging methods, unnecessary tissue loss is prevented and the incidence of postoperative adverse events is reduced [21].

The most common complication related to surgery was reported to be atelectasis and other complications indicated were wound infection, prolonged air leak, empyema, pneumothorax, and secondary hydatidosis [22]. Surgical methods were applied to all patients in our series. No mortality was observed during the one-year follow-up. Although two patients had cysts larger than 12 cm in diameter, no serious complication developed in these patients. The most common complications after surgery in our cases were atelectasis, prolonged air leak, and superficial wound infection.

In conclusion, surgery is sufficient for curative results in pulmonary hydatid cyst.

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

The study was approved by the local ethics committee of TOGU Medical School (Approval No. 22-Kaek-111).

Authors' contribution

KC: Conceived and designed the analysis, collected the data, developed the methodology, wrote the paper.

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Original Article

A thoracic surgery consensus statement for performing thoracic surgery in SARS-CoV-2 pandemic

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ABSTRACT

Background: Spreading to almost all countries in the world, the COVID-19 pandemic impacted healthcare and prevented or postponed major thoracic surgeries. In this study, members of the Turkish Society of Thoracic Surgery report a consensus reached by the members for providing thoracic surgery safely during the COVID-19 pandemic.

Materials and Methods: In this study, a questionnaire included categories of questions aimed to explore the aspect and perspectives of the professionals in the field. The survey was distributed through the email platform of the National Association which includes active members of the society and it was kept open between the 8th of April and the 7th of May 2021.

Results: Eighty members of the national association completed the survey. Most of the respondents were academic surgeons from university hospitals (43.8%) or teaching hospitals for thoracic surgery (36.3%). Almost all respondents agreed that patients with lung cancer and prolonged air leaks should be operated during the pandemic (98.8% and 96.3% respectively). The majority required that patients should be quarantined at the hospital or home up to 1 week before surgery. Academic surgeons statistically significantly more frequently thought that the quarantine period was unnecessary ($p = 0.045$). Most respondents indicated that stage IA to IIIA lung cancer patients should be operated. All respondents agreed that patients can be operated on provided that a PCR-negative swab test. Most of the professors thought elective surgeries should be delayed 1 to 4 weeks while some surgeons believed the delay should be more than a month ($p = 0.028$).

Conclusions: Thoracic surgery services for patients with thoracic should be maintained during the COVID-19 pandemic. We as thoracic surgeons are responsible to perform good surgical management of patients during this and any future pandemic.

Keywords: COVID-19, thoracic surgery, consensus, report, survey

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Introduction

Severe acute respiratory syndrome coronavirus 2 (SARS-CoV-2) has spread to almost all countries, with WHO declaring a COVID-19 pandemic on March 11, 2020 [1]. It has prevented major thoracic surgical surgeries in most countries redirecting almost all sources to patients with COVID-19. However, as necessary care for patients with COVID-19 is provided, there is also a concern regarding how management for cases needing treatment for other diseases can be provided.

The specialty of general thoracic surgery is expected to maintain the management of important entities such as lung cancer, esophageal cancer, inflammatory pulmonary diseases, and mediastinal and chest wall diseases. Appropriate treatment of these diseases is time-dependent. A number of guidelines have been published for the management of surgical patients during the SARS-CoV-2 pandemic [2,3]. However, they are based on expert opinions. A very large-multi-national study reported the outcomes of patients who had undergone surgery with perioperative SARS-CoV-2 infection indicated that; postoperative pulmonary complications occur in half of the patients they are associated with high mortality [4]. The necessity of timely treatment of thoracic malignancies is arguably higher than that of other malignancies where elective resections can be postponed [5,6]. It is therefore a surgical ethical challenge to maintain appropriate surgical treatment for patients with lung cancer despite limited resources during the continuing COVID-19 pandemic [7].

In this study, the Turkish Society of Thoracic Surgery reports a consensus reached by the members of society on the best possible strategy for providing thoracic surgery safely during the COVID-19 pandemic.

Materials and Methods

A questionnaire was designed on the Google Forms platform. The survey was designed by the Istanbul Branch of the Turkish Association of Thoracic Surgery. It included categories of questions that aimed to explore prerequisites of thoracic surgery during the pandemic, safety considerations when offering operations, and the setting where thoracic surgical services should be provided.

Multiple-choice answers were offered for the members to select from. The survey was distributed through the email platform of the National Association which includes active members of the society. The survey was kept open between the 8th of April and the 7th of May 2021. Every survey could be answered by every member only once based on the email that is used by the member. Multiple-choice answers were offered for the experts to select from. Survey questions aimed to define the decision for operation, preoperative work-up and taken precautions, surgical planning, and postoperative course.

Results from the survey formed the basis of the current Consensus Statement. The consensus was described as more than 50% agreement among the panel of experts [8]. Clinical practice was described as “recommended” if 50%-74% of the members reached an agreement.

Statistical Analysis

Data analysis was performed using IBM® SPSS Statistics 25.0(NY, USA) software. The chi-square statistics were used. Yates’ correction was made in certain comparisons. A p-value of less than 0.05 was considered statistically significant.

Results

Eighty members of the national association completed the survey. Most of the respondents were members who have been working at university hospitals (43.8%) or teaching hospitals for thoracic surgery (36.3%) (Figure 1). Professors comprised 36.3% of respondents whereas 68.8% of respondents have at least an assistant professor post (Figure 2).

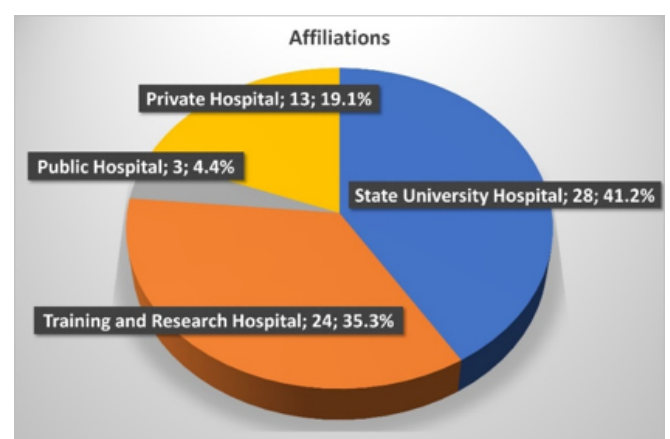


Figure 1. Distribution of affiliations of the respondents.

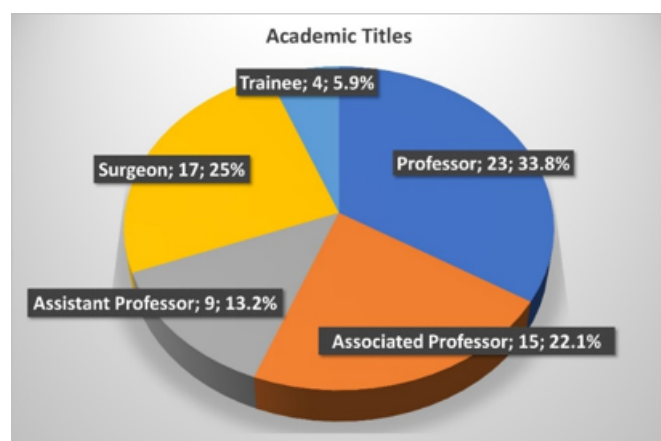


Figure 2. Distribution of academic titles of the respondents.

Almost all respondents agreed that patients with lung cancer and prolonged air leaks should be operated during the pandemic (98.8% and 96.3% respectively). Ninety percent of surgeons indicated that tracheal surgery should be done during the COVID-19 era. Most members recommended surgery for mediastinal diseases (Table 1).

The majority of respondents required that patients should be quarantined at the hospital or home up to 1 week

before surgery while 25.0% of surgeons declared that it is unnecessary (Table 2). Associate professors and professors statistically significantly more frequently thought that the quarantine period was unnecessary ($p = 0.045$) (Table 3). There was a consensus regarding the operable stages of lung cancer: Most respondents indicated that stage IA to IIIA patients should be operated. More non-professor respondents indicated that even patients with stage IIIB and stage IV lung cancer could be operated ($p = 0.066$) (Table 3).

All respondents agreed that patients can be operated on provided a PCR-negative swab test. There is a consensus that (i.e., 80%) patients should have negative thorax CT for COVID-19 before surgery (Table 2). There was no statistically significant difference among participants ($p = 0.938$) (Table 3).

Most of the members of the society agreed that FFP3 masks should be used during surgery, whereas other protective measures were deemed to be unnecessary (Table 2). Yet 23% of the surgeons believed that standard surgical gowns and surgical masks might be enough for protection (Table 3).

Table 1. Summary of responses regarding surgical indications during the COVID-19 pandemic.

Question	n	%
Which of the following surgeries should be performed during the Covid-19 pandemic (you can make multiple choices)?		
Stage IA - IIIA operable lung cancer surgery	79	98.8
Posterior mediastinal mass	56	70.0
Intact pulmonary hydatid cyst	39	48.8
Pneumothorax complicated with prolonged air leak	77	96.3
Thymoma resection	69	75.0
Thymectomy for Myasthenia Gravis	43	53.8
Sleeve resections	72	90.0
Tracheal surgeries	69	86.3
Chest wall deformities	1	1.3
Hyperhidrosis	1	1.3
If it is necessary to make surgical resections of lung cancer patients during this period, which stage patients should be performed without postponement (you can make multiple choices)?		
Stage I	72	90
Stage II	70	87.5
Stage IIIA	62	77.5
Stage IIIB – Stage IV	19	23.8
All operable lung cancer patients should be postponed	9	11.3
Which of the following types of thoracic surgery approach should be preferred during the pandemic?		
VATS and/or RATS	34	42.5
Thoracotomy/sternotomy	2	2.5
Only endoscopic procedures (bronchoscopy, esophagoscopy)	1	1.3
Surgical approach selection should not matter	43	53.7
Abbrev: ::VATS: Video-assisted thoracic surgery; RATS: Robotic-assisted thoracic surgery		

Table 2. Summary of responses regarding patient workup and safety during the COVID-19 pandemic.

Question	n	%
How long patients should be kept in quarantine prior to surgery?		
1-3 days	13	16.3
5-7 days	28	35.0
10 days	10	12.5
14 days	8	10.0
No need for quarantine	21	26.2
Which of the following is required before planned thoracic surgery (you can make multiple choices)?		
Negative SARS-CoV-2 RT-PCR result	80	100.0
A chest CT scan with no COVID finding	65	81.3
A chest X-ray scan with no COVID finding	21	26.3
Absence of signs/symptoms of COVID-19 (sore throat, shortness of breath, cough, fever, etc.)	66	82.5
Absence of a biochemical finding of COVID-19 (ferritin, D-dimer increase, lymphopenia, etc.)	49	61.3
When can a patient, who got infected by COVID-19 and turn to RT-PCR negative, undergo a planned thoracic surgery?		
1-2 weeks	13	16.3
2-4 weeks	34	42.5
4-6 weeks	23	28.7
More than 7 weeks	8	10.0
Avoid any thoracic surgery in patients with a COVID-19 infection background	2	2.5
Is it necessary to take an additional precaution other than a normal surgical mask during the surgery, even if the patient is COVID-19 negative (you can make multiple choices)?		
N95 / FFP3 mask	58	72.5
Wearing full protective equipment	8	10.0
Goggles	32	40.0
A surgical apron and mask is enough	22	27.5

Minimally invasive surgical procedures were recommended to be performed by 42.5% of respondents while 53.8% of surgeons pointed out that there should be no surgical approach preference (Table 1). Professors were in favor of minimally invasive approaches and there was statistical significance compared to other surgeons ($p = 0.031$) (Table 3).

Eighty-one percent of respondents indicated that a patient's companion was necessary after surgery (Table 4). Only seven participants were strictly against it during this period. More than half of the respondents (64%) thought that patients should be kept in quarantine after surgery to avoid post-operative COVID-19 infections (Table 3). One-fifth of surgeons recommended 3-day

quarantine. Sixty percent of members required that patients should be discharged as early as possible during the pandemic (Table 4).

In addition, almost all respondents recommended that (95.7%), some period of time should be passed after COVID-19 infection in patients with COVID-19 infection (Table 2). Ten percent of respondents recommended at least 7 weeks of waiting time after negative COVID-19 PCR. Most of the professors responded as 1 to 4 weeks would be enough while more than half of the other surgeons believed the delay should be more than a month to avoid any complications due to the infection ($p = 0.028$) (Table 3).

Table 3: Comparison of certain responses from professors and others

Q/A	Professors	Other	p
The quarantine period prior to surgery is...			
...necessary	10	30	0.045
...not necessary	19	22	
Lung cancer surgery during the pandemic			
Stage I-III A Non-small cell lung cancer	20	31	0.066
All operable cases	9	21	
CT scan in addition to swap to avoid COVID-19 for preoperative work-up...			
...is indicated	23	43	0.938
...is not indicated	6	9	
Personal protection during surgery to avoid any risk of contamination			
Personal protective equipment is necessary	23	39	0.869
A standard gown and surgical mask is enough	6	13	
The safe and recommended surgical approach during the pandemic			
Minimally invasive approaches	15	16	0.031
Not relevant	14	36	
During the pandemic should any companion be allowed in wards?			
No	1	6	0.214
Yes, if necessary	28	46	
Post-surgical quarantine			
No need	11	18	0.967
Yes for a couple of days	20	32	
Rapid discharge should be the main strategy for post-operative patient care during the pandemic			
Disagree	1	6	0.407
Agree, depends on the procedure	28	46	
How long should one wait for surgery after a COVID-19 infection?			
1 to 4 weeks	22	25	0.028
More than a month	7	27	

Table 4. Summary of responses regarding post-surgical care during the COVID-19 pandemic.

Question	n	%
Is it safe and necessary to let a companion during the hospital stay after the surgery?		
Yes	17	21.3
No	15	18.8
It depends on the surgical procedure.	48	60.0
Is it necessary to discharge the patients earlier in the pandemic era?		
Yes	48	60.0
No	6	7.5
It depends on the surgical procedure.	26	32.5
Do the patients need to stay in quarantine after discharge? If yes for how long?		
No	33	41.3
3 days	0	0
5 days	4	5.0
7 days	17	21.3
15 days	21	26.2
more than 15 days	5	6.2

Discussion

This study represents a collective definition of how to provide thoracic surgical services to patients with thoracic surgical diseases during the COVID-19 pandemic. The survey was conducted during the peak stage of the Omicron-variant surge across the world and Turkey and reflects the real-time and real-world decisions being taken in Turkish thoracic surgical units. It has been well recognized that healthcare resources including surgical wards and operation rooms due to reclaiming some operation rooms into the intensive care units stretched thin [9]. For this reason, a balance may have to be reached between the necessity to manage patients with COVID-19 and the need to treat patients with operable lung cancer.

In our survey, there was a consensus that resectional surgery should proceed for treating lung cancer, mediastinal tumors, and tracheal lesions. Similarly, ASCVTS recommended by consensus that those patients should be operated during the pandemic [7,10]. Most respondents agreed that thoracic surgery should be performed after a period of quarantine. No thoracic surgery publication recommended a quarantine. However, Ralhan and colleagues stated that high-risk contacts should be tested for COVID-19 using RT-PCR after 7 days of quarantine [11]. Quarantine followed by a COVID-19 test theoretically decreased the COVID-19 infection that may develop perioperatively.

Covid-Surg collaborative study showed that COVID-19 infection developed perioperatively resulting in almost 25% of mortality [4]. For this reason, every effective measure should be taken before planned surgery. However, more members of the thoracic surgery society with high academic ranking (i.e., professors and associate professors) tended to perform surgery without any quarantine period. This statistically significant difference in strategy might be attributed to having a better and established multidisciplinary care system such as infectious diseases and multidisciplinary intensive care units by high-ranking academic personnel.

On the other hand, patients who had NSCLC and cases who might be considered marginally operable might be treated with chemoradiation followed by immunotherapy as suggested by the PACIFIC trial [12]. A fraction of patients with stage 1 NSCLC had delayed operation assuming the delay could not affect the survival of those patients [13].

Most respondents preferred to perform surgery for lung cancer patients provided certain requirements such as quarantine, negative PCR of nasal swabs, and a negative thorax CT for COVID-19. The consensus was also reached that FFP3 masks should be used during surgery, whereas other protective measures were deemed to be unnecessary. This selection of protective measures was also recommended before [10]. It should be highlighted that; the majority of respondent surgeons indicated that there was no selection of surgical approach (i.e., VATS or thoracotomy) whereas more than 40% of surgeons preferred VATS over thoracotomy during the pandemic. The experts of ASCVTS concluded that open surgery was not deemed to be safer than a minimally invasive surgery [10], however, other surgical specialties suggested a higher risk for laparoscopy due to the use of high-pressure carbon dioxide during the operation [10]. However, carbon dioxide is rarely used by thoracic surgeons. Of note, shorter hospital stays, and shorter drainage time associated with VATS procedure may be helpful in decreasing the postoperative exposure of the patients to hospital cross-infection [14].

The necessity of quarantine after thoracic surgery is yet to be described in the literature [15]. However, in our study, the majority of respondents suggested a quarantine period after surgery in order to prevent possible contamination. It has been well established that perioperative COVID-19 leads to very high mortality as high as 23.8% in 30 days. For this reason, every attempt should be made to prevent such a perioperative COVID-19. However, a national surgical guideline did not recommend any type of postoperative quarantine [16]. Nevertheless, it is preferable that the patients should be hospitalized for as short as possible. Of note, it has been well established that the implementation of enhanced recovery after thoracic surgery allows a reduction of postoperative stay and complication [17,18]. However, the effectiveness and safety of enhanced recovery after surgery protocols in the COVID-19 era are yet to be established.

In our study, a survey indicated that, for the surgical patients who had COVID-19 before planned surgery, there is a consensus that surgery should be delayed. Almost all thoracic surgeons agreed on the delay before planned surgery. In a very large study that analyzed 140,231 patients from 116 countries [19], authors concluded that surgery

should be delayed for at least 7 weeks following COVID-19 in patients. It has been found that, if surgery was performed more than 7 weeks after COVID-19, mortality risk was associated with a similar mortality risk to baseline (i.e., OR: 1.5) whereas it was 4.1 in patients having surgery within 0-2 weeks of the diagnosis. However, the impact of delay on the mortality and survival of patients with cancer has been well investigated [20]. Even a four-week delay in cancer treatment was found to be associated with increased survival in surgical treatment [20]. It could be proposed that neoadjuvant therapy can be initiated in those patients with SARS-CoV-2 infection. However, many patients could not be fit for even neoadjuvant chemotherapy during this period.

There are some limitations of the study. Survey respondents were the members of the national society who accepted to answer the questionnaire. The experience and seniority of the respondents varied. We did not use a well-established model such as the Delphi process due to a large number of participants. Nevertheless, it would be reasonable to suggest that the size of respondents was large as it could have been. In the study, decisions were made as the interpretations of the responses.

The take-home message here is to evaluate the conditions and risks regarding the current pandemic. Since the survey took place in the beginning period of the global recovery from the pandemic and the first applications of vaccinations, the responses should be valid until this day when we are still discussing new waves, variants, and any possible precautions such as masks to be mandatory again. WHO did not still remove the 'pandemic' status of the disease. Probably, SARS-CoV-2 will continue to infect humans although the preventive measures have been effective for the last year [21]. In addition to that, there may be additional viral threats that may cripple the caregiving system and force us to make critical decisions once again [21]. This statement is therefore crucial in showing an example of a collaborative effort to fight against a pandemic and give thoracic surgical care to those who need them the most.

In conclusion, the results of the survey that were described here should provide thoracic surgeons a guide-

line when representing how patients can be operated for lung diseases. During the pandemic, patients who are to undergo thoracic surgery rely on thoracic surgeons to not only implement appropriate surgical treatment but to advocate for their right to receive it.

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Authors' contributions

AT,IS: qualify for authorship according to the following criteria, analyzed and interpreted data, final approval. AT: contributed to concept and design, revised the manuscript, IS: acquired data, drafted the manuscript, AD,KB,DS,MY: administration and mentoring the study.

Availability statement

The data that support the findings of this study are available from the corresponding author upon reasonable request.

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Original Article

A retrospective analysis of chest wall deformities incidentally detected in emergency department patients: an observational study

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ABSTRACT

Background: The most frequently observed chest wall deformities are pectus excavatum and pectus carinatum. The purpose of this observational study was to examine the prevalence of chest wall deformities in adult patients presenting for various reasons to the emergency department in our institution, a regional reference center for chest deformities, the demographic characteristics of patients with such malformations, emergency presentation symptoms, and the effects of deformities on patients' psychological state.

Materials and Methods: Two hundred forty cases presented to the İzmir Katip Çelebi University Atatürk Training and Research Hospital tertiary emergency department and meeting the inclusion criteria were investigated retrospectively. The cases' demographic data, presence of family histories of similar deformities, presenting symptoms, deformity depths, Haller indices, and patients' psychological deformity levels were determined and subjected to statistical analysis.

Results: The prevalence of pectus excavatum among the patients presenting to our center was 1/308, while the prevalence of pectus carinatum was 1/698. The patients' mean age was 24.75 years, and 84.6% were men. The accompanying psychological deformity was present in only 28.33% of cases. The presence of family history was determined at 38.8%. The patients' mean Haller Index value was 3.16, and the mean depth of deformity was 2.5 cm. Haller Index values were significantly higher among patients with moderate or severe psychological deformity than in the other patients ($p < 0.001$). The Haller Index exhibited a high correlation with depth of deformity ($p < 0.001$) and a negative correlation with patients' body mass index values ($p = 0.024$).

Conclusions: Chest wall deformities are a very common societal health problem, one that exhibits psychological effects and familial predisposition, and that can lead to various physiological problems. Familial disposition levels may vary among societies. Adult patients begin to tolerate the presence of low body mass index and psychological deformity. Prospective studies are now needed for greater clarification of these issues.

Keywords: pectus excavatum, pectus carinatum, chest wall anomalies, psychological deformity, Haller Index

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Introduction

The shape of the human chest cavity varies from individual to individual [1]. Differences outside normal limits occurring for various reasons during growth in the development of osseous or cartilaginous tissues that comprise the chest wall are known as anterior chest wall deformities. The most common chest wall deformities are pectus excavatum (PE) and pectus carinatum (PC) [2]. PE (sunken chest or cobbler's chest) was first described by Eggel in 1870. It is the most frequently seen chest deformity (80-90%), characterized by varying degrees of depression of the sternum and costal cartilages into the thorax [3]. PC (pigeon chest), characterized by varying degrees of anterior protrusion of the sternum and sternocostal cartilages, is the second most common chest deformity. Although these deformities are congenital, they generally produce findings in adolescence.

These malformations emerging in childhood can cause varying degrees of structural and psychological disturbance [4]. While no life-threatening functional problem is observed in the majority of pectus patients, various system anomalies may be observed in severe cases. The most frequent symptom in patients with chest deformities is dyspnea on exertion, which is more common in individuals with PE. Various cardiac problems and respiratory system disorders associated with structural anomalies may also be seen in these patients. Studies have also reported that the life span may be shorter in these patients [5,6].

Individuals with thoracic deformities are a cause for concern for families from the outset. Individuals with deformities gradually begin to experience various psychological problems deriving from their physical appearance [7]. Psychiatrists have performed numerous studies on this subject since 1960 and have employed the term 'psychological deformity' in reference to it. Problems such as anxiety, motivation disorder, and labeling anxiety were observed at a rate of 82% in one study, lack of self-esteem and timidity at 78%, indecisiveness at 72%, and aggression at 66% [8].

This observational study was designed to examine the prevalence of chest wall deformities in adult patients presenting for various reasons to the emergency department in our institution, a regional reference center for chest deformities, the demographic characteristics of patients with such malformations, emergency presentation symptoms, and the effects of deformities on patients' psychological states.

Materials and Methods

This retrospective observational study was performed in the tertiary emergency department of the Izmir Katip Çelebi University Training and Research Hospital, Turkey. Approval for the study was granted by the Izmir Katip Çelebi University Non-Interventional Clinical Research Ethical Committee (no: 0360 Date: 25/05/2022).

A total of 60,725 patients randomly selected using computer software [PASS 11(Power Analysis Sample Size)] were screened from among all patients aged over 18 and presenting to the emergency department for any reason within the previous five years. The sample size was determined as a result of prehoc analysis at a 95% confidence interval and 0.05 margin of error. Patients with chest wall malformations were identified, and the study was conducted with these.

Patients who refused to participate, with psychiatric diseases other than psychological deformity, with systemic disease, whose imaging records or contact information were unavailable, who underwent surgery, or with an additional musculoskeletal disease were excluded from the study.

Patients' age, sex, family history, disease history, body mass index (BMI), deformity type and depth, and presenting symptoms were recorded on the basis of information in our hospital's data management system. The presence of chest deformities in first- and second-degree relatives was regarded as a positive family history. Deformity depths were calculated and recorded from the physical examination notes for patients with PE in the hospital system, and Haller indices from computed tomography (CT) images in the hospital system.

Depth of deformity refers to the value in centimeters on the measurement bar at the intersection of a transversely placed ruler passing over the most protuberant points on both hemithoraces after the measurement rod has been placed perpendicular to the sternum at the deepest point of the chest with the patient in a standing position. The Haller Index value was determined by proportioning the maximum lateral diameter of the chest at thoracic CT and the distance between the sternum and vertebra at the maximum depression point [5].

The Beck Depression Inventory (BDI) was applied to identify whether any depressive component was present in patients' psychological deformities and to classify the level thereof, if applicable. The BDI, developed by Beck et al.

in 1961, is a self-report scale used to evaluate behavioral findings of depression in adolescents and adults [6]. The individual's level of depressive symptoms is determined by means of this scale. The individual is asked questions under 21 headings, with four options for each. The form is completed in the light of the respondent's answers, and the level of depression is classified in the light of the score obtained [5]. Scores of 0-9 are regarded as normal status, 10-18 as indicating mild depression, and 19-29 as indicating moderate depression. Scores of 30-63 indicate severe depression or risk thereof. Depression can be distinguished at a rate of 90%, depending on the score obtained [7]. In order to be able to perform this measurement with the patients in the present study, they were contacted using the details in the system, and the psychological effects of their diseases were investigated. Depression was accordingly classified as none, minimal, moderate, or severe.

Statistical Analysis

All the study data were analyzed on IBM SPSS Statistics Standard Concurrent User version 26 statistical software (IBM Corp., Armonk, NY, USA). Descriptive statistics were presented as unit number (n), percentage (%), mean $\pm$ standard deviation ($\bar{x} \pm SD$), median (M), minimum (min), and maximum (max) values. The distribution of data for numerical variables was assessed using the Shapiro Wilk normality test. Since the data did not meet normal distribution conditions, comparisons between two groups were performed using the Mann-Whitney U test, while three-way comparisons were performed with the Kruskal Wallis H test. The Pearson and Fisher exact tests were applied in the comparison of categorical variables. Spearman's Rho correlation coefficient was used to examine relationships between numerical variables. P values < 0.05 were considered statistically significant.

Results

Investigation of the 60,725 patients screened for the study showed that chest deformity accompanied emergency department presentation symptoms in 284 cases. In terms of the cross-sectional prevalence of the sample, chest deformity was present in a mean one in 214 patients. One hundred ninety-seven (69.37%) of the patients with deformity were diagnosed with PE and the remaining 87 (30.63%) with PC. Prevalence was thus 1/308 for PE and 1/698 for PC.

Twenty-four of the 284 patients with chest wall deformity were excluded since they could not be contacted for various reasons or else declined to take part in the study, 12 due to imaging results being unavailable, one patient due to a diagnosis of schizophrenia, and seven due to various accompanying systemic or musculoskeletal diseases (diabetes, rheumatoid arthritis, Marfan syndrome, and osteogenesis), and the study was therefore finally conducted with 240 patients.

As shown in table 1 the patients' mean age was 24.75 years, and 203 (84.58%) were men. Scoliosis was observed as a structural anomaly in 20 patients (8.33%). No psychological deformity was encountered in 68 patients (28.33%), while at least minimal accompanying psychological deformity was present in the remaining 172 (71.67%). Examination of emergency department presentations in terms of symptoms that may occur in association with chest deformity revealed that the most common complaint was back pain, in 23 cases (9.58%), followed by effort dyspnea in 19 (7.91%), and chest pain in 16 (6.66%). The patients' mean BMI value was 21.52.

As shown in table 2, Haller Index values were higher in patients with moderate and severe psychological deformity than in the other patients ($p < 0.001$).

As shown in table 3, Haller Index values exhibited a significant correlation with the depth of deformity in patients with PE (the one increasing with the other at a value of 76.00%) ($p < 0.001$).

However, in other patients, there was a negative statistically significant correlation of 57.70% between the Haller index and the BMI value, with one increasing and the other decreasing ($p = 0.024$). As shown in table 1, the mean age of the patients with PE alone was 25.17. Male patients represented one hundred fifty (85.71%) of the patients with PE, with male gender predominating over female. A family history was present in 56 patients (32.00%). Ten (5.71%) patients exhibited structural anomalies diagnosed as scoliosis. No psychological deformity was encountered in 62 patients (35.43%), while at least minimal accompanying psychological deformity was present in all the remaining cases. The patients' mean Haller Index value was 3.16, and their mean depth of deformity was 2.50. The most common symptom potentially associated with chest deformity was also back pain, observed in 17 (9.71%) cases. BMI values ranged between 15 and 35..

Table 1. Descriptive values.

Variables	Values	Patients with pectus excavatum
Group, n (%)	175 (72.9)	
Pectus excavatum	29 (12.1)	
Pectus carinatum	36 (15)	
Other		
Age, years		
$\bar{x} \pm SD$	24.75 $\pm$ 6.14	25.17 $\pm$ 6.22
M (min-max)	24 (18-55)	24 (18-55)
Gender, n (%)		
Female	37 (15.4)	25 (14.28)
Male	203 (84.6)	150 (85.71)
Family history, n (%)		
Yes	93 (38.8)	56 (32.00)
No	147 (61.2)	119 (68.00)
Structural anomalies, n (%)		
ASD	6 (2.50)	2 (1.14)
Aortic insufficiency	6 (2.50)	4 (2.28)
Dextrocardia	3 (1.25)	2 (1.14)
Mitral valve insufficiency	8 (3.33)	3 (1.71)
Scoliosis	20 (8.33)	10 (5.71)
VSD	3 (1.25)	2 (1.14)
Psychological deformity, n (%)		
None	68 (28.33)	62 (35.43)
Minimal	97 (40.41)	58 (33.14)
Mild	41 (17.08)	31 (17.71)
Moderate	26 (10.83)	17 (9.71)
Severe	8 (3.33)	7 (4.00)
Emergency presentation Symptom, n (%)		
Dyspnea	12 (5.00)	10 (5.71)
Palpitation	15 (6.25)	12 (6.85)
Effort dyspnea	19 (7.91)	11 (6.28)
Chest pain	16 (6.66)	9 (5.14)
Syncope	3 (1.25)	1 (5.20)
Back pain	23 (9.58)	17 (9.71)
Other reasons for presentation	152 (63.33)	115 (65.71)
BMI		
$\bar{x} \pm SD$	21.51 $\pm$ 3.07	21.71 $\pm$ 3.13
M (min-max)	21 (15-35)	21 (15-35)
Haller Index		
$\bar{x} \pm SD$		3.16 $\pm$ 0.54
M (min-max)		3.00 (2.60-5.60)
Depth of deformity		
$\bar{x} \pm SD$		2.50 $\pm$ 0.83
M (min-max)		2.40 (1.00-5.60)

Abbrev.; %: Row percentage, M: Median, $\bar{x}$: Mean, SD: Standard Deviation

Table 2. A comparison of psychological deformity and Haller Index values.

	Haller Index	Test statistics	
	M (min-max)	H value	p value
Psychological deformity	2.9 (2.6-3.6) ^a	84.279	<0.001
None	3.0 (2.6-3.8) ^b		
Minimal	3.2 (2.7-4.3) ^c		
Mild	3.9 (3.2-4.7) ^d		
Moderate	4.9 (3.8-6.0) ^d		
Severe			

Abbrev.; M: Median, H: Kruskal Wallis test. The letters a b c d represent multiple comparisons. There is no statistical difference between groups containing the same letter, there is a statistically significant difference between groups containing different letters.

Table 3. Correlation relationships.

Group		Age	Haller Index	BMI
Pectus excavatum	Haller	rho	-0.077	0.010
	Index	p	0.344	0.900
	Depth of deformity	rho	-0.026	0.760
		p	0.753	<0.001
Other	Haller	rho	0.182	-0.577
	Index	p	0.517	0.024
	Depth of deformity	rho	-0.059	0.417
			0.836	0.122

Abbrev.; rho: Spearman Correlation Coefficient

Discussion

The most common chest wall malformation is PE, followed by PC. Reported incidences are 1/300-400 for PE and 1/1000 for PC [1,2,8]. Both deformities are five times as common in males [1,2,9]. Prevalence among patients presenting to our hospital emergency department for various reasons in the present study was 1/308 for PE and 1/698 for PC. The frequency of the male gender was 84.6% for all chest wall deformities and 85.7% for PE. Accordingly, the prevalence of PE in the present study was consistent with the previous literature, while that of PC was higher. In addition, a slightly higher frequency of male gender was determined for both deformities.

Examination of family histories in the patients with chest wall deformities included in the study revealed a rate of chest deformity in first- and second-degree relatives of 38.75%, and a figure of 32% among the isolated

PE patients. The genetic basis of PE is still unknown, although the presence of similar genetic deformities in 40% of family members is clinical evidence corroborating the idea that PE is a genetic disease [4]. The familial dominance in PC is 25% [9]. Lower familial disposition was determined for PE among the patients in the present study, while such disposition was higher than previously reported for PC. These rates may vary among countries and cultures since the frequency of all diseases with such familial disposition increases in societies with higher rates of consanguineous marriage.

Posture disorders accompany deformity in 50-80% of patients with PE. They are thin and tall in appearance, with a narrow chest, increased dorsal lordosis, shoulders slumped forward, and a protruding abdomen. Accompanying scoliosis is present in 26-30% of cases [10]. Although PC may appear as an isolated deformity, 51.6% of patients exhibit one or more associated musculoskeletal system abnormalities (posture impairment), with forward head posture, uneven shoulders, rounded shoulders, thoracic kyphosis, lumbar lordosis, anterior pelvic tilt, and scoliosis having been reported [11]. Comparatively lower rates of scoliosis and other structural anomalies were encountered in the present series.

Mean and median BMI values of 21 were determined in both the PE and PC groups. These values, within normal ranges for an adult individual, were surprising, since patients with chest deformities exhibit lower BMI values than in society in general. In a study published in 2016, Park et al. investigated 1371 patients and reported a mean BMI of 18 [12]. However, it should not be forgotten that the majority of patients seen in clinical practice and in Park et al.'s study are in the pediatric population, while we included patients in the adult age group. Although chest deformities cause some degree of BMI depression in the early age group, patients can tolerate this and normalize their BMI levels at later ages.

The most frequent emergency department presentation symptoms potentially associated with chest deformity are back pain, effort dyspnea, and chest pain. We think that presentations to the emergency department due

to back pain are particularly common because such pain is prevalent in society and in trauma patients. Dyspnea with exertion, generally the most common symptom, was the second most frequent symptom in our series.

The Haller Index is calculated as the ratio of the maximal transverse diameter of the chest to the minimal anteroposterior diameter; values are lower than 2.0 in healthy individuals, while values exceeding 3.5 have been reported as an indication for surgery [13,14]. Values between 2.0 and 3.2 are defined as 'mild excavatum,' 3.3-3.5 as 'moderate excavatum,' and values of 3.6 or higher as 'severe excavatum.' The mean Haller Index value in the present series was 3.16, with a median value of 3. This shows that we screened individuals from all patient groups, not solely those with severe excavatum. This also confirmed the data obtained concerning the depth of deformity. A depth of less than 1 cm is regarded as minimal, 1-3 cm as moderate, and greater than 3 cm as severe deformity (1.0-5.6). From that perspective, the Haller Index and depth of deformity values obtained were mutually consistent.

Chest wall deformities are problems that generally require correction. Indications for corrections occur in two groups – functional (cardiac, pulmonary, and postural-orthopedic) or psychosocial-esthetic indications. In other words, even in the absence of any functional complaint, patient dissatisfaction with his appearance is by itself an indication for correction. It is therefore of great importance to establish the prevalence of 'psychological deformity, even in an observational study such as the present research. We employed a depression inventory in the context of psychological deformity in this study, and determined at least minimal depression in all patients with chest wall deformity, and in 64.6% of those with PE. From that perspective, the rate of psychological deformity revealed by our data differed only minimally from that in the previous literature. Due to the low number of presentations among patients under 18, since ours is an adult hospital, this patient group was excluded from the study, and this may account for the lower rate of psychological deformity than in the previ-

ous literature. This is because older adult patients may be expected to be more reconciled to their deformities than the adolescent group aged under 18.

Another finding emerging from this study is that the severity of psychological deformity increased in line with Haller Index values. This correlation was highly significant. This suggests that the Haller Index is a good indicator not only of the patient's physiological characteristics, but also of his psychological state. Another advantage of the Haller Index lies in the fact that BMI values decrease as Haller values rise. This is because, as an indicator of the severity of the deformity, the Haller Index is also correlated with the problem of low BMI emerging secondary to the deformity. This makes the Haller Index particularly significant.

The significant correlation between the Haller Index and depth of deformity confirmed that-, similarly to the index itself, the latter is also an important marker. In addition, while radiological imaging is required to calculate Haller Index values, measuring the depth of deformity is noteworthy as a highly practical method that can be applied anywhere.

Limitations

The principal limitations of the present study are its retrospective and single-center nature, the fact that patient randomization was not performed, and that pediatric cases were not included. Since the study was retrospective, patients were reached through their contact information. This has led to the possibility that the depression level of the patients may change over time. Further randomized, multi-center clinical studies consisting of large series and patients from all age groups are now needed to clarify the situation.

In conclusion, chest wall deformities are a relatively common health problem in society, involving psychological effects that frequently result in psychological deformity, involving a familial predisposition, and are capable of leading to problems such as low BMI in adult patients. The Haller Index and the depth of deformity are both potentially good indicators of both psychological and physical state. Further prospective studies are now needed to shed greater light on this subject.

Declaration of conflicting interests

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Ethics approval

The study was approved by the Izmir Katip Çelebi University Non-Interventional Clinical Research Ethical Committee (no: 0360 Date: 25/05/2022).

Authors' contributions

OSC, HE; conceptualized and designed the study, collected analyzed, and interpreted the patient data regarding pulmonary metastasis, co-wrote the paper, MGE; collected and analyzed data, revised the final version of the manuscript, and wrote the paper. All authors read and approved the final manuscript.

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Original Article

Efficacy of thoracic ultrasonography in thoracostomy tube removal

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ABSTRACT

Background: Tube thoracostomy is the most commonly performed procedure in thoracic surgery. Follow-up with daily chest X-ray is performed for both pneumothorax patients and postoperative patients, but residual pneumothorax can often be missed on portable imaging in supine position. This study aimed to evaluate the efficacy and safety of ultrasonography and chest X-ray by comparing these two methods in chest tube removal and thus determine whether ultrasonography is suitable for routine use in thoracic surgery clinics.

Materials and Methods: This prospective study included a total of 28 patients who underwent tube thoracostomy in our center between December 2019 and December 2020 due to spontaneous pneumothorax (n = 16) or after wedge resection for different indications (n = 12). Chest X-ray and thoracic ultrasonography were performed before and after chest tube removal. The efficacy of thoracic ultrasonography compared to chest X-ray in the detection of residual pneumothorax was evaluated using specificity, sensitivity, positive predictive value (PPV), and negative predictive value (NPV).

Results: Twenty-four (85.7%) of the patients were male. The mean age was 40.25 ± 19.75 years (median, 39.5). Tube thoracotomy was performed on the right side in 18 patients (64.3%). Ultrasonography before tube removal had a PPV of 50%, NPV of 100%, sensitivity of 100%, and specificity of 96.3%. After tube removal, ultrasonography had a PPV of 100%, NPV of 96%, sensitivity of 75%, and specificity of 100%.

Conclusions: The results of our comparison of chest X-ray and thoracic ultrasonography examinations performed before and after chest tube removal showed that thoracic ultrasonography was as effective as chest X-ray and can be used safely in clinics.

Keywords: Thoracic ultrasonography, tube thoracostomy, pneumothorax

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Introduction

Tube thoracostomy (TT) is the most commonly performed procedure in thoracic surgery. It is among the most frequent interventions for preventing potentially fatal complications and postoperative pneumothorax (PTX) in particular. However, standard management guidelines for this procedure are still lacking. Different centers have varying practices regarding approaches to reduce morbidity, follow-up protocols, the timing of chest tube removal, and length of hospital stay [1,2]. Clinical follow-up of both PTX patients and postoperative patients is performed with daily chest X-ray (CXR), but many cases of PTX may be missed in portable imaging acquired in the supine position [3,4]. Although the most effective method of monitoring for PTX is computed tomography (CT), its use in daily practice is limited for several reasons, including that it is costly, involves intense radiation exposure, and is not portable, which causes difficulties in patient transport [4-7]. Thoracic ultrasonography (US) has been used in the evaluation of PTX for 30 years, and with recent advances in technology, it has become much more sensitive than CXR in the diagnosis of PTX [8,9]. Especially in recent years, its superiority over CXR in the detection of pathologies such as PTX and hemothorax in patients with chest trauma has been recognized. Thoracic US was also reported to be effective in demonstrating drain position during and after TT [10-12]. Advantages of bedside US include that it is easy to perform, can be readily repeated, is portable, has superior sensitivity and specificity, is low-cost, does not cause radiation exposure, and effectively guides the decision to remove a chest tube [1]. Despite the many articles on this subject in the literature, there has been little research on thoracostomy tube removal.

This prospective study aimed to evaluate the efficacy and safety of US and CXR by comparing these two methods in chest tube removal and thereby determine whether US is suitable for routine use in thoracic surgery clinics.

Materials and Methods

A total of 28 patients who underwent TT in the thoracic surgery department of the Recep Tayyip Erdoğan University Faculty of Medicine Training and Research Hospital between December 2019 and December 2020

were included in the study. Of these, 16 patients had spontaneous PTX and 12 underwent wedge resection for different indications. Patients under 18 years of age, pregnant women, patients who underwent TT due to trauma, and patients who underwent anatomical resection (lobectomy and segmentectomy) due to infectious or malignant causes were not included in the study.

The decision to remove the chest tubes was made upon observing reduced oscillation in the drain and the resolution of the air leak. On the day of removal, the tube was clamped for 6 hours, during which CXR and concurrent bedside US were performed. Three hours after drain removal, CXR and concurrent US examination were repeated once more. All CXRs were obtained with the patient standing. Bedside US was performed at the second and third intercostal spaces on the midclavicular line with the patient in supine position with head raised 45 degrees. All US examinations were done by the same radiologist who was experienced in thoracic radiology and had not seen the patients' CXRs. The procedure was performed in the longitudinal plane using a high-frequency superficial probe (DC-3; Mindray; 5-10 MHz). The appearance of lung lines and the presence of artifacts in the aerated lung tissue were evaluated in B-mode US imaging. The "lung sliding" sign was interpreted as lung expansion. Using the same device in the same area, the presence or absence of movement with the expansion of the lung tissue was examined under M-mode imaging. The "sandy beach" sign confirmed lung expansion, while the "stratosphere" sign was accepted as an indicator of air in the pleural space (Figure 1,2) [14].

Approval for this prospective, single-center descriptive study was obtained from the Clinical Research Ethics Committee of Recep Tayyip Erdoğan University Faculty of Medicine Training and Research Hospital (date: September 3, 2020; decision no: 2020/193).

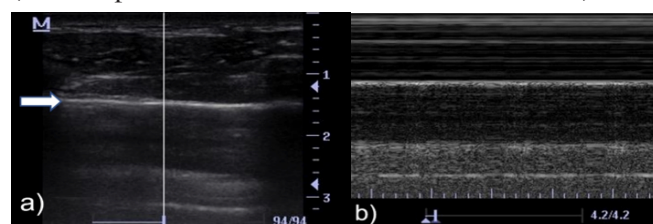


Figure 1. B- mode two-dimensional ultrasonographic image of normal lung tissue. The pleural line is seen as a hyperechogenic interface (arrow) (a), "seashore sign" on the M-mode image at the same level. There is no pneumothorax in this patient (b).

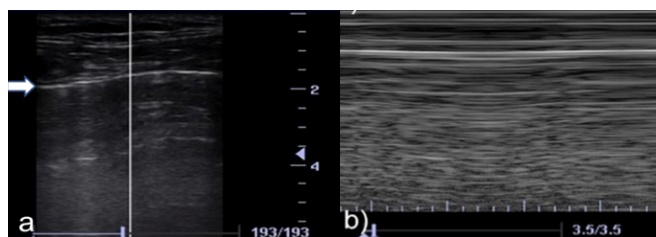


Figure 2. B- mode two-dimensional ultrasonographic image from 2nd intercostal space. A pleural line is seen (arrow) but lack of sliding was detected (a), “stratosphere sign” on M-mode image at 2. intercostal space, apical pneumothorax was defined on lung US (b).

Statistical Analysis

Statistical analyses were performed using R Software version 4.0.3 (R foundation, Vienna, Austria). The data were presented as number and percentage values for categorical variables and as mean and standard deviation for numerical variables. The efficacy of US in the evaluation of lung expansion was assessed by calculating its sensitivity, specificity, negative predictive value (NPV), and positive predictive values (PPV) compared to CXR.

Results

Of the 28 patients who underwent TT in this study, 24 (85.7%) were men and 4 (14.3%) were women. The mean age was 40.25 ± 19.75 years (range, 19-75) and the median age was 39.5. TT was performed on the right side in 18 patients (64.3%) and on the left side in 10 patients (35.7%). The indication for TT was spontaneous PTX in 16 (57.1%) of the patients, while the other 12 patients (42.9%) underwent TT after surgery. Of these surgical procedures, 5 (41.6%) were video-assisted thoracoscopic surgery (VATS) wedge resection for lung biopsy, 4 (33.4%) were VATS bullectomy, and 3 (25%) were VATS metastasectomy. The mean length of hospital stay was 7.71 ± 2.92 days for nonsurgical patients (those who underwent TT due to pneumothorax) and

8.07 ± 2.87 days (median: 7) among postoperative patients (wedge resection) (Table 1).

The chest tube was clamped for 6 hours before removal, during which CXR and US examinations were performed. In our comparison of the methods when used before thoracostomy tube removal, the US had a PPV of 50%, NPV of 100%, sensitivity of 100%, and specificity of 96.3% (Table 2). Of the 28 patients, PTX was detected in 1 patient with CXR, whereas 2 patients were evaluated as having PTX according to the US. This was due to the presence of apical bullae in one of the patients whose US findings were interpreted as PTX, which is the reason for the 50% PPV. In the comparison made after tube removal, the US had a PPV of 100%, NPV of 96%, sensitivity of 75%, and specificity of 100% (Table 3). PTX was detected on CXR in 4 patients after tube removal. PTX was detected in 3 of these patients in the US. Two of these 4 patients were followed with nasal oxygen for 24 hours and discharged, while the chest tubes were replaced in the other 2 patients.

Table 1. Characteristics of patients (n = 28).

	Mean $\pm$ SD or n (%)
Age	40.25 $\pm$ 19.57
Gender	
Male	24(85.7)
Female	4(14.3)
Side of PSP	
Right	18(64.3)
Left	10(35.7)
Length of hospital stay	
Wedge resection	8.07 $\pm$ 2.87
Tube thoracostomy	7.71 $\pm$ 2.92
Reason for tube thoracostomy	
Spontaneous pneumothorax	16(57.1)
Post-surgery (wedge resection)	12(42.9)

Abbrev.; SD: Standard deviation, PSP: Primary spontaneous pneumothorax

Table 2. Comparison of thoracic US examination compared with chest x-ray findings before tube thoracostomy removal.

		Chest x-ray		Sens	Spes	PPV	NPV
		PTX	Non PTX				
Torax US	PTX	1	1	100.0%	96.3%	50.0%	100.0%
	Non PTX	0	26				

Abbrev.; Sens: Sensitivity, Spes: Specificity, PPV: Positive predictivity value, NPV: Negatif predictivity value

Table 3. Comparison of thoracic US examination compared with chest x-ray findings after tube thoracostomy removal.

		Chest x-ray		Sens	Spes	PPV	NPV
		PTX	Non PTX				
Torax US	PTX	3	0	75.0%	100.0%	100.0%	96.0%
	Non PTX	1	24				

Abbrev.; Sens: Sensitivity, Spes: Spesificity, PPV: Positive predictivity value, NPV: Negatif predictivity value

Discussion

Thoracic US is an important post-trauma imaging modality, having 95% sensitivity and 100% specificity in the diagnosis of PTX and hemothorax in trauma patients [13,15]. Although there are many publications about the use of the US in the diagnosis of trauma patients, very few studies have investigated its effectiveness in planning thoracostomy tube removal [5,6,16]. In general, studies have only focused on the diagnostic efficacy of the US and how it compares to CXR.

Thoracic US is helpful in many stages of the TT procedure. Authors have discussed the efficacy of using the US at every stage, from preventing intercostal injury and tube malposition during placement to daily monitoring and deciding the most appropriate time for tube removal [11,12,17]. Lavingia et al emphasized that physicians can easily make these evaluations with basic US training [18].

Complications that may occur after tube removal can be listed simply as PTX recurrence and the development of hemothorax. Hernandez et al. reported a three-fold increase in cost when these complications occur after tube removal. They emphasized that increased cost was a result of missing the findings on CXR, whereas this rate was quite low in patients evaluated with US [19]. However, in our series, there were no patients with recurrent PTX or fluid collection that was missed on CXR but detected by the US.

US evaluation of lung expansion is based on the movement of the pleural line. In the presence of pneumothorax, movement of the pleura is not observed in B-mode imaging. In M mode, parallel lines form in the absence of movement, which is called a barcode or stratosphere sign. As a control, when the lower intercostal spaces or symmetrical lung space are evaluated

in M mode, the parallel line structure is disrupted and a complex dotted appearance occurs upon lung expansion (sandy beach sign) [20].

Studies have suggested that the US of the chest has similar accuracy to plain radiography. The main advantages of US are that it can be performed at the bedside, has a short evaluation time, and does not involve ionizing radiation [14]. In addition to emergency medical units, the use of the US in thoracic surgery wards to examine patients during PTX follow-up and before thoracostomy tube removal has been discussed in the literature in recent years [21]. In a 2018 study conducted with 50 patients, it was suggested that follow-up imaging of patients after tube removal could be performed by the US [20]. Galbosis et al reported in their study that residual air that was undetectable on plain radiography could be demonstrated by lung US. Only patients with residual PTX detected by the US required tube replacement [22]. In recent years, we have started routinely using thoracic US in the diagnosis and treatment of effusions that require intervention in patients with hemothorax, as well as for other thoracic catheterizations that require prior marking. With support from experienced radiologists, we have also started increasing our experience with the use of bedside US in chest tube removal to protect our patients from additional radiation.

Regarding the use of US during chest tube removal, a meta-analysis of thoracic US emphasized that the adequacy of US depends on the examiner [9]. In our study, we also attempted to identify factors that may affect the results of evaluations made by the US. The presence of the chest tube makes it difficult to assess pleural movement and reduces its prominence in the US. Before the procedure, the patient's head should be elevated and time allowed for any air present in the pleural space to move cranially. Turning on the device after adjusting the pa-

tient's position provides the optimal time required before starting the procedure. Observing the pleural movement of the fellow lung can prevent false positive results. US evaluation of pleural movement is difficult in patients with bullae, diffuse interstitial lung diseases, or a prior history of chest tube placement. Movements of the chest wall and the effect of heart pulsation on the left lung can cause an appearance that mimics the pleural movement and lead to false negative results [8,16,20,22].

The main limitation of our study is that despite this being a prospective study, we were unable to conduct multivariate analyses because of the small number of patients. Due to the COVID-19 pandemic, we limited the sample size to avoid unnecessary risks to the health of both patients and physicians.

In conclusion, the data obtained in our study support the conclusion that US is as effective as CXR in thoracostomy tube removal and the evaluation of residual pneumothorax. Larger prospective studies with homogeneous groups are needed in this regard. The most important advantages of bedside US are that it is easy to perform, involves no radioactive adverse effects, reduces the need for patient transport, and can provide a rapid diagnosis. Apart from residual PTX, its ability to reveal fluid collections, especially in postoperative patients, is also valuable.

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 prospective, single-center descriptive study was obtained from the Clinical Research Ethics Committee of Recep Tayyip Erdoğan University Faculty of Medicine Training and Research Hospital (date: September 3, 2020; decision no: 2020/193).

Authors' contributions

KT, NH; conceptualized and designed the study, collected analyzed and interpreted the patient data, GS, HT; collected

and analyzed data and revised the final version of the manuscript. All authors read and approved the final manuscript.

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Case Report

Coexistence of an esophageal bronchogenic cyst and a thyroglossal duct cyst: a case report and review of literature

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ABSTRACT

An esophageal bronchogenic cyst is a rare form of a bronchogenic cyst, a foregut developmental anomaly in embryological age. Herein, we present a case that underwent surgery for the complicated mediastinal esophageal bronchogenic cyst and a thyroglossal duct cyst, another anterior foregut anomaly at the neck. Bronchogenic cysts should be operated on as early as possible due to the complications and a detailed and radiological examination should be kept in mind for other foregut anomalies that may accompany it. To the best of our knowledge, this is the first case reported as the coexistence of two foregut anomalies.

Keywords: esophageal bronchogenic cyst, thyroglossal duct cyst, foregut anomalies

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Introduction

A bronchogenic cyst is an anomaly, derived from the anterior foregut. They usually appear in the mediastinum or lung parenchyma and are rarely found in the esophageal wall. Multiple extrapulmonary localizations have also been reported [1-3]. Esophageal or mediastinal bronchogenic cysts usually cause symptoms due to compression to the surrounding tissues. Particularly, the cyst may eventually rupture when infected or due to the accumulation of mucus secretion. In addition, malignant transformation is possible after chronic inflammation [4,5]. The general treatment approach for the bronchogenic cyst is surgical resection.

Case Report

A 58-year-old female patient was referred to our clinic with symptoms of dyspnea and chest pain for over three years. With the progression of her complaint, she was admitted to the emergency service several times with pleuritic chest pain and dyspnea symptoms. The patient's vital signs showed a heart rate of 85 bpm, a respiration rate of 16 breaths per minute, and an oxygen saturation of 95% in room air. On auscultation, no pathological finding was detected. Laboratory results revealed a normal complete blood count and basic metabolic profile. Thorax CT scan located a 26x28 mm cystic lesion in the subcarinal region containing homogeneous, low-density fluid (Figure 1).

In the operation preparation period, ultrasonography of the neck was made due to her history of a multinodular goiter, which showed a submental cyst that revealed a thyroglossal duct cyst. The radiology and Ear-Nose-Throat specialist recommended a follow-up.

A cervical and thoracic MRI was performed to evaluate the relationship between subcarinal and submental cysts with the surrounding tissues or organs (Figure 2). The first lesion measuring 6x13 mm was located in the middle line of the submental region, and it was isointense in T1W1s and hyperintense in T2W2s. The other lesion that was located paraesophageal in the subcarinal region showed a hyperintense image on T1W1s, and isointense in T2W2s, and was measured 26x28 mm (Figures 1,2). By comparing with the previous CT scan reported, it was determined that the subcarinal cyst had grown in size within 3 years.

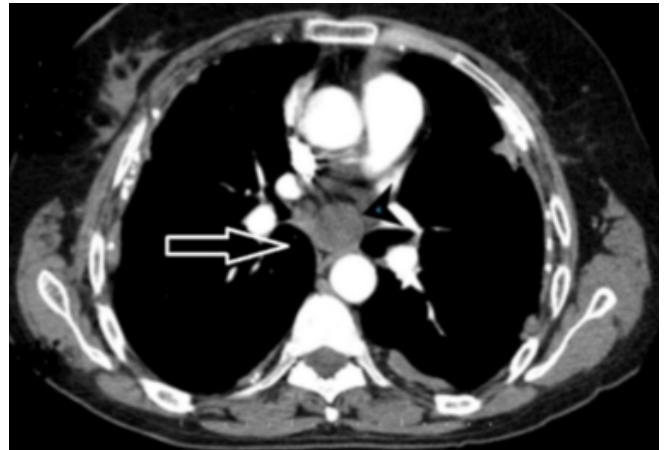


Figure 1. Thorax CT showing the subcarinal cyst.

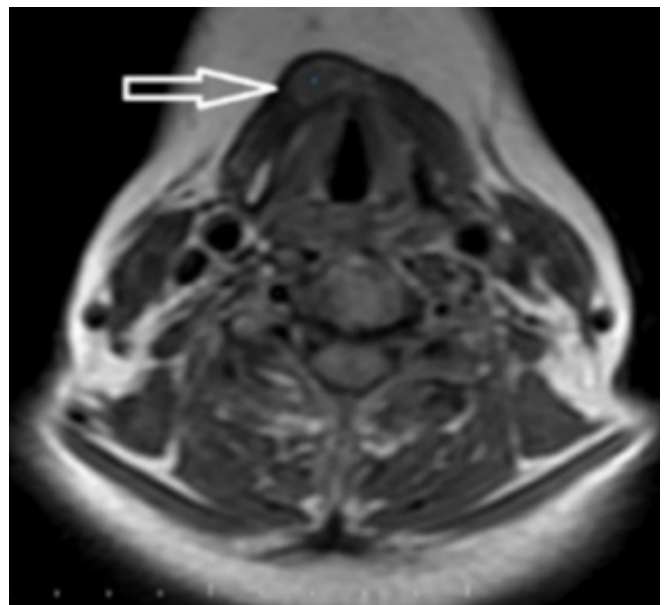


Figure 2. Cervical MRI showing the submental cyst.

A right posterolateral thoracotomy was performed for the excision of the mediastinal cyst. The mediastinal pleura was dissected at the subcarinal-hilar level. It was observed that the lesion was partially attached to the esophageal muscle layer with the appendix. No connection with the bronchial system was observed. The appendix was not explored. The cyst ruptured spontaneously without opening its contents. The greenish cyst contents were discharged. At the level of the lesion, the esophagus was suspended and dissected circumferentially. The appendix of the cyst, which was partially embedded in the esophageal muscular layer, was excised by ligation with a 2/0 silk suture without any damage to the esophageal mucosa. The esophageal wall incision was closed with 2/0 silk interrupted sutures (Figure 3). The postoperative period was uneventful. The patient was followed up for three years.

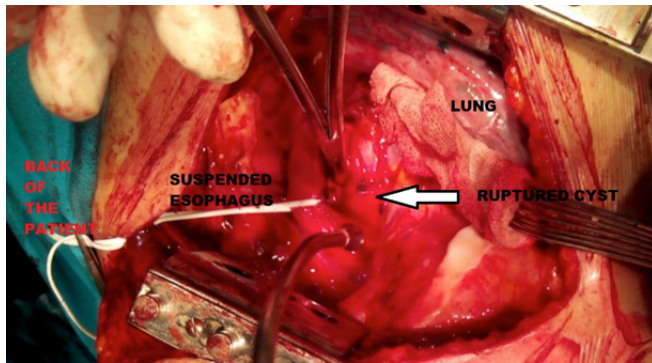


Figure 3. The image of the subcarinal cyst during the operation.

Pathological examination observed a fibrous cyst wall that was edematous that covered with pseudostratified ciliated respiratory epithelium which included bronchial glands, smooth muscle, and inflammatory cells (Figure 4). All these findings were reported in agreement with the bronchogenic cyst. There was no finding suggestive of malignancy. Written informed consent was obtained from the patient for the publication of her data.

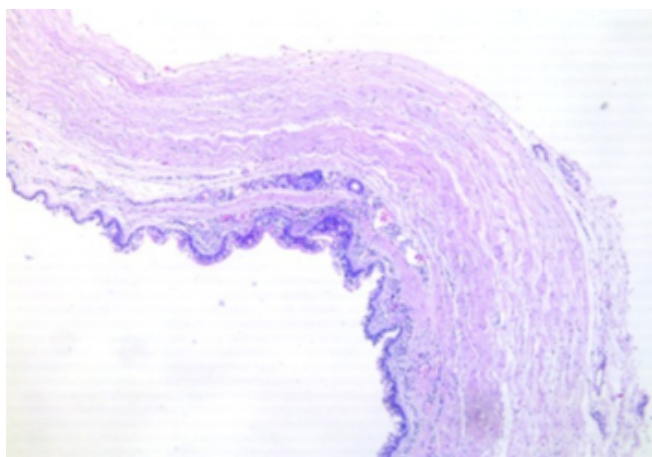


Figure 4. The microscopic image showing a fibrous cyst wall, covered with pseudostratified ciliated respiratory epithelium including bronchial glands, smooth muscle, and inflammatory cells (HEx40).

Discussion

In humans, a range of foregut anomalies can occur depending on the region of the foregut. Many of these defects result from abnormal morphogenesis of endodermal tissue, suggesting that there is a common developmental mechanism between the organ systems.

On the 28th day of the embryonic period, the esophagus and laryngotracheal groove are together in the same circular canal. The inner part of this canal is composed of endodermal cells, while the outer part consists of mesodermal cells. The cells in the foregut

endoderm are multipotent cells that develop the thyroid, thymus, esophagus, lung, liver, pancreas, stomach, and biliary systems. The transformation of these organs and systems initiates from the endoderm and mesoderm, depending on the dose, by the Transforming Growth Factor B (TGF β) signaling released from the primitive node [6]. The foregut endoderm cells start polarizing to the ventral and dorsal regions. The development of the trachea from the endoderm cells occurs by polarizing to the ventral area and the development of the esophagus occurs from the dorsal regionally endoderm cells when foregut endoderm cells start to split into two regions. Certain molecules play an important role in polarizing and splitting this dorsal/ventral. Previous studies showed that the Sox2 gene in the dorsalization of the esophagus, the Nkx2.1 gene is effective in the ventralization of the trachea [7,8]

This balance between Nkx2 and Sox2 is provided by the proteins Noggin, FGF, Wnt, and Shh that are found in the mesoderm surrounding the endoderm and play a role in the development of body tissue. Genetic and epigenetic mutations in these genes cause foregut separation distortion/failure or epithelial/mesenchymal abnormal differentiation. Although the pathogenesis of bronchogenic cysts is currently unclear, several theories have been proposed. It has been suggested that the development of the bronchogenic cyst is caused by the mutations of the Wnt gene [8].

A thyroglossal duct cyst was reported incidentally in the neck USG was performed due to a previous history of thyroid disease in the patient who did not have a primary neck complaint. In our case, a thyroglossal duct cyst was detected with a bronchogenic cyst. To the best of our knowledge, this case is the first report featuring two foregut anomalies. The thyroglossal duct cyst is one of the foregut anomalies that is commonly seen in the cervical region. The primordial thyroid gland develops from the endoderm that originates at the base of the tongue (foramen cecum) and descends through the anterior of the hyoid bone to arrive at its final position [9]. The pyramidal lobe on the thyroid gland is a residue of the persistent thyroglossal duct. Additionally, the aperture of the duct anywhere through the descent of the thyroid gland can cause the development of a thyroglos-

sal duct cyst. The most typical presenting symptom is an upper midline mass intimately associated with the strap muscles around the neck, which is mobile upon tongue protrusion or swallowing.

Bronchogenic cysts are one of the most common foregut malformations, usually occurring along the tracheobronchial tree in the mediastinum. Approximately two-thirds of it is located in the mediastinum and the one-third in intraparenchymal. It can also be located in the mediastinum in the paratracheal, subcarinal, hilar, and esophageal regions. Other unusual locations, such as the pericardium, thymus, subcutaneous, diaphragm, retroperitoneum, gastroesophageal junction, and cervical region, have also been reported [1-3].

The symptoms of the bronchogenic cyst may vary depending on its location, size, and complications (e.g., pressure symptoms, cyst hemorrhage, or infection). Typically, bronchogenic cyst infections are observed when they are associated with the trachea-esophageal lumen. However, a connection between the trachea-esophageal lumen and the cyst may not always be demonstrated radiologically. The heterogeneity of the cyst and the increase in cyst size in the radiological follow-up may be the best indicator to confirm that it's infected or associated with the trachea-esophageal lumen.

Additionally, cysts are isointense on T1W1s MR images. However, when infected, the cyst's intensity is increased on T1W1s images [10]. In our case, T1W1s images were hyperintense. This situation explained the reason for the recently increased symptoms of our patient. Although the relationship of the cyst with the esophageal lumen could not be fully demonstrated in our case, the increase in cyst's density in the MRI images, the increase of the cyst's size in follow-up thorax CT scans, and the infection findings in histopathological evaluation suggested that there may be a millimeter-sized connection between the esophagus and the cyst.

Bronchogenic cysts cause symptoms associated with compression of the surrounding organs and tissues [11-13]. The most serious complication in this is infection and cyst rupture. There are reports of malignancy in the cyst due to recurrent infections [2].

In conclusion, bronchogenic cysts should be operated on at the earliest as possible due to the complications and should also be evaluated in terms of other accompanying foregut anomalies. A detailed examination and radiological examination should be kept in mind for other foregut anomalies that may accompany it in the evaluation of bronchogenic cyst cases.

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Authors' contributions

HGB,SK,AZ: 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

Fatal pulmonary sequestration supplied by the pulmonary artery in a neonate

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ABSTRACT

Pulmonary sequestration is defined as a segment of the lung that is supplied by one or more systemic arteries and has no connection with normal bronchial structures. This rare lung condition is basically classified into two different types: extralobar sequestration (ELS), which has its own pleura, and intralobar sequestration (ILS), which is in structural continuity with normal lung tissue. While aberrant arterial supply is usually derived from the thoracic aorta, abdominal aorta, celiac artery, splenic artery, and intercostal artery, it can also rarely be provided from the subclavian, left gastric, phrenic, and coronary arteries. This patient group can be asymptomatic but may present with complaints such as cough, fever, hemoptysis, and chest pain when symptomatic. Surgery is currently the treatment of choice for such patients.

Here, we present a rare and fatal case of pulmonary sequestration supplied by the pulmonary artery in a neonate who was born at 32 weeks of gestation weighing 1980 g, required mechanical ventilatory support, and was operated on while intubated on the 32nd day of birth to provide curative treatment.

Keywords: pulmonary sequestration, congenital diseases, pulmonary arterial supply, surgery

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Introduction

Pulmonary sequestration (PS) is defined as a segment of the lung that is supplied by one or more systemic arteries and has no connection with normal bronchial structures. Although it was first described by Huber (in 1777), the term “sequestration” was first introduced in 1946 [1].

Pulmonary sequestration is classified into intrapulmonary and extrapulmonary sequestrations depending on the presence of its own pleura. Although most of the sequestrations are supplied by the thoracic aorta and abdominal aorta alone, they can also receive blood supply from more than one abnormal systemic artery [2].

Both types of PS can be asymptomatic; however, the most common clinical symptoms include cough, sputum, fever, and hemoptysis when they are symptomatic [3].

Case Report

Our patient was born at 32 weeks of gestation weighing 1980 g from a 33-year-old mother who became pregnant through in vitro fertilization. The patient, who was non-invasively followed up with a pre-diagnosis of adenoid cystic malformation in the intrauterine period, was admitted to the neonatal intensive care unit and intubated due to severe dyspnea immediately after birth. His chest x-ray showed an appearance consistent with respiratory distress type 1 and a pulmonary mass (Figure 1).



Figure 1. The initial chest x-ray of the patient after birth showing a mild cardiac shift to the right, and an appearance consistent with type 1 respiratory distress syndrome (RDS).

Surfactant therapy was administered to contribute to lung maturation. The patient with oxygen desaturation on mechanical ventilation was suspected to have pulmonary hypertension, therefore underwent a cardiac echo, which revealed PDA. Contrast-enhanced computed tomography of the chest (Figures 2,3) taken for the mass appearance was reported as a space-occupying formation of soft tissue density, extending from the left lung lower lobe superior segment to the basal segments, is notable. The soft tissue lesion appears to have no connection with the bronchial structure. The inferior branch of the left main pulmonary artery is markedly large and enters the soft tissue area in the lower posterior basal segment of the left lung. The clinically unstable patient with a persistent desaturation problem developed cardiopulmonary arrest once. He responded to cardiopulmonary resuscitation (CPR), achieving partial recovery in the clinic during the follow-up. After interviewing the patient’s family, a surgery decision was taken on the 32nd day of birth. The patient underwent a left thoracotomy. Exploration showed extrapulmonary sequestration with its own pleura on the left. It was easily isolated from the surrounding lung. It was observed that the sequestration artery originated from the pulmonary artery and the vein was drained into the pulmonary vein by extending along the same track. The artery and vein of the sequestration area that was found to have no connection with the bronchial structure were cut using a stapler, and sequestrectomy was completed. Mechanical ventilatory support was discontinued due to the dramatic improvement in respiratory parameters on the fourth day of the follow-up of the patient, who was again on mechanical ventilation postoperatively. The chest tube was removed on the sixth day of the follow-up. The patient was discharged in an extubated state on the 43rd day of hospitalization (Figure 4). Written informed consent was obtained from the parents for the publication of his data.

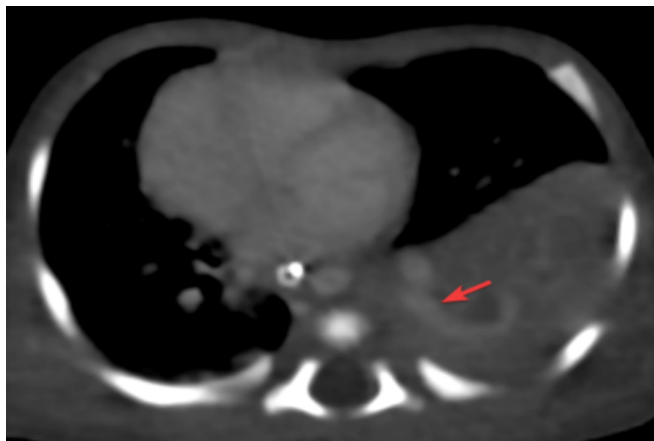


Figure 2. Contrast-enhanced computed tomography showing a mass appearance on the left and the branch of the pulmonary artery supplying blood to the sequestration extending into the mass marked with an arrow.



Figure 3. Coronal section showing the branch of the pulmonary artery (arrow) supplying the sequestration.

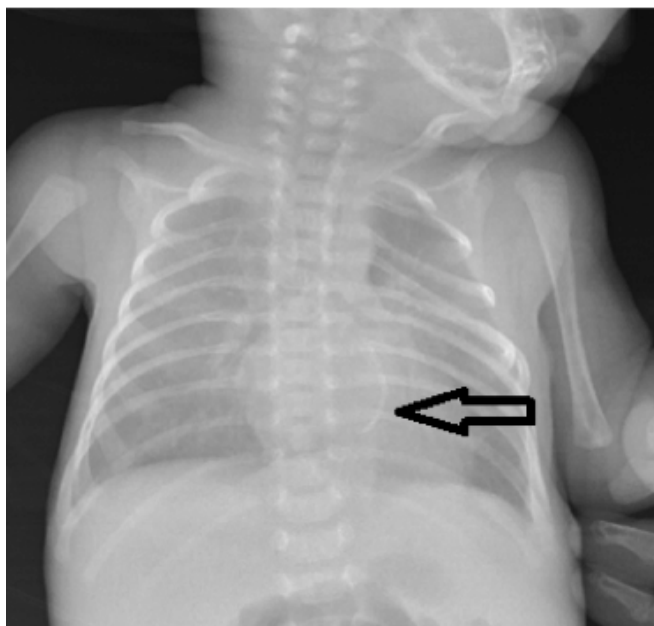


Figure 4. Post-sequestrectomy chest x-ray (the stapler line was marked with arrow).

Discussion

Considering that some cases of PS are asymptomatic and may not be detected at all, it is not possible to state a precise incidence. However, post-mortem pediatric and antenatal studies have reported an incidence of 0.15-1.8% for PS [4].

The most common site of involvement is the left lower lobe, followed by the right lower lobe. While PS is most commonly located in the abdomen other than the chest, it can also occur in different locations such as the neck [5,6].

Considering pediatric cases reported in the literature, approximately one-third of the patients are asymptomatic. The manifestations of symptomatic PS include respiratory tract infections [6].

Although large series of PS in the literature does not mention that the clinic of the patients may be severe enough to require respiratory and circulatory support [3,6], there are rare cases with such severe clinical manifestations presented in the form of a case report. Among these, there is a case report of PS supplied by the pulmonary artery in a premature infant with a severe clinic requiring early surgery, as in our patient [7,8,9].

Although chest x-ray is the first-line diagnostic test, contrast-enhanced computed tomography is sufficient for diagnosis today. This technique can also clearly demonstrate arterial supply. For this reason, pulmonary angiography has nowadays lost its former popularity. Apart from that, MRI and ultrasound are used as diagnostic techniques. However, diagnostic studies on both pediatric and adult patients have reported a quite high rate of misdiagnosis for these techniques [3,6].

Our patient was also followed up as having adenoid cystic malformation in the intrauterine period. The diagnosis could only be clarified by postnatal tomography.

One of the basic characteristic features of sequestration is systemic arterial supply. As in our patient, sequestration that receives blood supply from the pulmonary artery is very rare. As a matter of fact, a study of 2625 patients with PS in the literature identified arterial supply in 1808 patients, reporting pulmonary arterial supply in only 5 of them. Furthermore, a study of 208 pediatric patients found no pulmonary arterial supply at all [3,6].

As is understood from this information, pulmonary arterial supply is very rare in cases of PS. There is no comment in the literature about the relationship of this condition with the clinical conditions of the patients.

Surgery appears to be the most effective treatment approach currently. Apart from this, it is necessary to be cautious about congenital diaphragmatic hernia and other anomalies accompanying ELS, especially in ELS. When diagnosed, it may be appropriate to surgically treat these in the same session [6].

Another important point in surgery is that more than one arterial supply is seen in 20% of PS cases. Although a second arterial supply was not noted in our patient, it should be kept in mind that the arteries travel in the sub-pleural area. These are the main points to keep in mind to avoid a complication.

In fact, the debates on the surgery are two-dimensional. The first is whether to intervene in asymptomatic patients, while the other is about the timing of surgery.

The prevailing belief is that all patients should be referred to surgery after the diagnosis is confirmed. This approach seems reasonable to prevent future complications [3,6].

Nevertheless, there is no clear approach in the literature on timing. Infections have been shown to cause mortality and morbidity in children. While this frequency was the highest in the pediatric group in the first six months, it was found to be the lowest between 6 months and 1 year of age. Therefore, the recommended timing of surgery is between 6 months and 1 year of age [3].

As mentioned earlier in the literature, the diagnosis of our patient was challenging due to the rarity of pulmonary supply. The patient had a cardiac arrest while waiting for PDA closure and an appropriate time for surgery. Accordingly, the patient was operated on after discussing with the family, without discontinuation of ventilatory support. The patient achieved a dramatic improvement postoperatively. Accordingly, ventilation could be withdrawn, the chest tubes were removed, and finally, he was discharged.

Thoracotomy is the traditional approach adopted in surgery. However, it is of note that video-assisted thoracic surgery (VATS) has provided very good results in

the pediatric and adult groups [6,10]. We preferred thoracotomy for our patient because of severe respiratory problems and low birth weight. However, VATS should not be excluded as a surgical treatment technique even for these patients.

In conclusion, it should be kept in mind that PS can be severe enough to cause severe respiratory distress in newborns, requiring emergency surgical intervention even though some of the cases are asymptomatic. Moreover, pulmonary arterial supply can be seen in sequestrations. We achieved curative results in the patient after surgery. Therefore, surgery should be performed as soon as possible after the clinical stabilization of the patients, albeit partially. It should be noted that surgery is the only known curative treatment for these patients.

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Authors' contributions

OFD,OO,OT: 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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