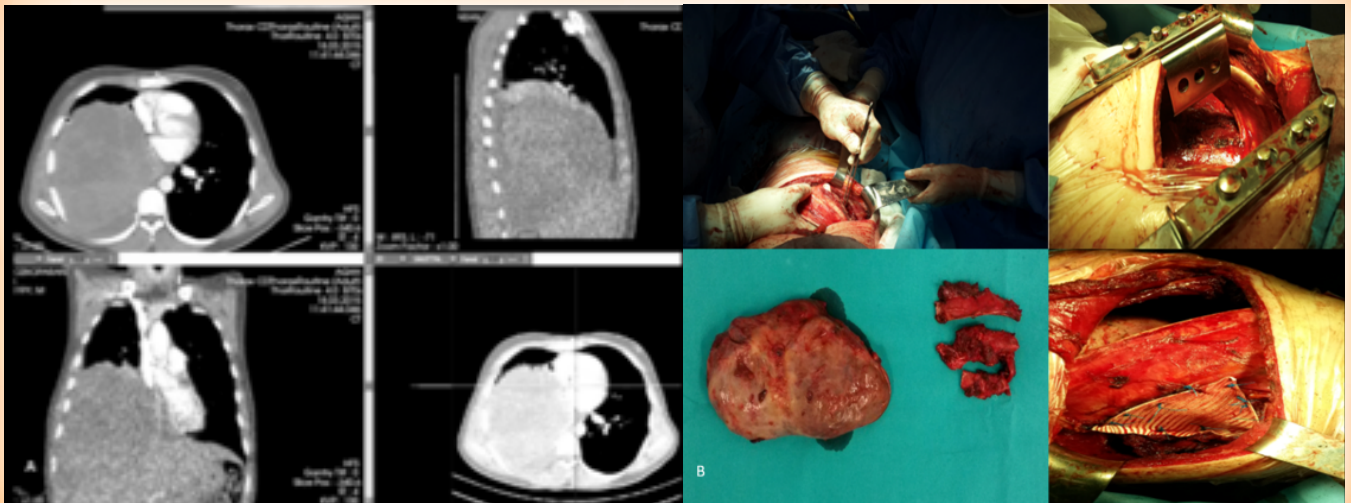


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Original Article

Is primary thoracic Ewing sarcoma aggressive than others? Seventeen-years' experience

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ABSTRACT

Background: Ewing sarcoma (ES) is an uncommon aggressive malignant tumor of the bone and/or soft tissue and belongs to peripheral primitive neuroectodermal tumor (PNET) family of tumors. Bone PNETs primarily arise from the diaphysis of long bones, whereas soft tissue PNETs are most commonly found in the chest wall. Multidisciplinary treatments, including chemotherapy, surgery, radiotherapy, or all three combined, improve the survival of patients with localized ES. However, the best approach to achieve local control remains controversial.

Materials and Methods: We retrospectively analysed the medical records and pathology data of 14 patients (8 male, 6 female; mean age, 23.2 [range, 4-54] years) with primary thoracic ES who underwent surgery in our clinic between January 2002 and December 2019. In addition, the treatment modalities and tumor-related factors of chest wall ES and lung parenchyma were evaluated.

Results: The most frequent complaint was chest pain (n = 7). In 10 patients, the tumor originated from the ribs, whereas the remaining 4 patients had lung parenchymal tumors. Ten patients underwent complete tumor excision with chest wall resection, one patient underwent lower lobectomy with chest wall resection, and three patients underwent complete tumor excision via wedge resection. All patients were treated with chemotherapy, except two who underwent bone marrow transplantation. The median follow-up was 31.6 (range, 2-84) months. Relapses were seen in 5 cases in the median 19.8th (range, 4-60) month.

Conclusions: Complete tumor resection is the most effective treatment for thoracic ES and multimodal therapy (surgical resection, chemotherapy, and local radiation therapy), which is recommended when indicated, constitutes the optimal treatment for ES. Although relapses occur within the early postoperative period, late relapses are not uncommon. The follow-up periods must be short and should be maintained long term for late relapses.

Keywords: Ewing's sarcoma, chest wall, chemotherapy, surgery, recurrence

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Introduction

Ewing sarcoma is an aggressive malignancy with a high incidence of local recurrence and distant metastasis, which are important prognostic factors. Surgical resection, chemotherapy, and local radiation therapy, when indicated, constitutes the optimal treatment. Thoracic sarcomas frequently involve the ribs, characterized by indolent progression. Hereby, these tumors are often diagnosed at a locally advanced stage, with massive pleural cavity involvement, or even at a metastatic stage (in 25% of cases) [1]. Although the relapses occur within the early postoperative period, late relapses are not uncommon. The follow-up periods must be short and should be maintained long term for late relapses.

ES primarily affects the bones and/or soft tissues and mainly occurs in children and young adults. Approximately 10% of ES arises from the chest wall [2]. Although ES can develop in any bone or soft tissue, the most common sites are flat bones or long bones of the leg, such as the pelvis, axial skeleton, and femur. Extraskeletal ES is by far less common than skeletal ES. ES located in the duodenum, neck, and kidney was reported in some case series. However, primary ES arising from the lung is extremely rare [3]. Along with malignant small-cell tumors of the thoracopulmonary region (Askin tumor), primitive neuroectodermal tumor (PNET), extrasosseous ES (EES), and atypical ES, and these malignancies are part of a spectrum of neoplastic diseases known as the ES family of tumors. These tumors were considered to be derived from a common cell of origin due to their similar histopathological and immunohistochemical characteristics and shared non-random chromosomal translocations [4].

The patients have metastasis at diagnosis about 20%–30%, and the presence of metastasis at the time of diagnosis is the most unfavorable prognostic factor. The treatment of chest wall ES involves intense local therapy with surgical resection and/or radiation therapy, and chemotherapy is used to treat systemic diseases. In addition to surgery, chemotherapy improves the outcome from 15%–20% to 60%–70% [5]. In this study, we retrospectively analysed the findings of the cases with primary thoracic ES, and total resection surgery was performed.

Material and Methods

We retrospectively analysed the medical records and pathological data of 14 patients (8 male, 6 female; mean

age, 23.2 [range, 4–54] years) with primary thoracic ES who underwent surgery in Ataturk Chest Disease and Thoracic Surgery Training and Research Hospital, Thoracic Surgery Department between January 2002 and December 2019. The pathological material was reviewed by an experienced sarcoma pathologist. The presence of small round cell tumors with CD99 positivity without lymphoma, rhabdomyosarcoma, or neuroblastoma pathological features was assessed as ES. A panel of immunohistochemical stains, including CD99, synaptophysin, chromogranin, neuron-specific enolase, S-100, vimentin, leukocyte common antigen, cytokeratins (AE1/AE3), desmin, and actin, was used.

Data of patients who underwent surgical resection in our clinic were evaluated retrospectively for age, gender, clinical features, tumor size and localization, clinical-stage, surgical procedure, resection margins, chemotherapy, radiotherapy, and treatment results. Informed consent was obtained from all patients. Approval was obtained from Ataturk Chest Disease and Thoracic Surgery Training and Research Hospital of the local institutional review board (No: 03.12.2020/ 703).

All cases were staged with clinical history, physical examination, routine laboratory tests, chest radiograph, and computed tomography (CT) of the chest and brain. Positron emission tomography (PET)–CT was used for staging in three cases, while others were evaluated using additional abdominal ultrasound for metastasis. Bronchoscopy and transthoracic needle biopsy with CT were applied in all patients preoperatively.

In this study, all patients had primary thoracic ES and underwent complete resection with safe margins. Event-free survival (EFS) was assessed using the date of disease progression, relapse, death, or last follow-up visit from the date of diagnosis. The overall survival was calculated from the date of diagnosis to the date of death or last follow-up.

Thirteen patients with Thoracic ES were diagnosed using tomography-assisted transthoracic biopsy (n = 3), incision biopsy (n = 3), metastasectomy (n = 5), supraclavicular lymph node biopsy (n = 1), and endobronchial cryobiopsy (n = 1); however, these patients were excluded from the study due to distant metastasis or unresectable/inoperable tumor. Patients with isolated thoracic ES who underwent total resection were included in the study.

Results

The most frequent complaint was chest pain ($n = 7$, 50%), followed by cough ($n = 4$, 28.5%), swelling of the chest wall ($n = 2$, 14.2%), and hemoptysis ($n = 1$, 7.1%). Table 1 shows the clinical features and demographics of patients. All patients were evaluated using chest CT. PET/CT was used for five patients. Three of the patients with PET scan were screened preoperatively and median fluorodeoxyglucose (FDG) uptake was 4.03 (2.5-6.4). A 19-year-old male who was operated on due to post-traumatic pleural hematoma and ES was detected during surgery, and a 4-year-old female patient was evaluated using PET/CT in the postoperative period for staging; both these patients showed no evidence of distant metastasis.

In 10 patients, the tumor originated from the ribs, whereas the remaining 4 patients had lung parenchymal tumors. Surgical procedures included complete tumor excision with chest wall resection ($n = 10$), lower lobectomy with chest wall resection ($n = 1$), and complete tumor excision via wedge resection ($n = 3$). All the patients were treated with chemotherapy, except for two patients who underwent bone marrow transplantation. The median follow-up was 31.6 (range, 2-84) months. Relapse was seen in 5 cases (35.7%) in the median 19.8th (range, 4-60) month (Table 2).

Table 1. General characteristics of patients who were operated for thoracic Ewing sarcoma.

Patient No	Age	Gender	Complaint	Localization	Size (cm)
1	54	F	Cough	RLL	12
2	36	M	Pain	LLL	12
3	14	M	Pain	LLL	13
4	20	F	Cough	RLL	15
5	27	M	Cough	LLL	2
6	13	M	Pain	6-7th rib, RLL	8
7	11	F	Pain	RLL	5
8	25	M	Hemoptysis	RLL	5
9	21	M	Pain	10th right rib	25
10	16	F	Swelling	RLL	17
11	19	M	Pain	RLL	14
12	39	M	Pain	RLL	6
13	26	F	Pain	RLL	7
14	4	F	Swelling	LUL	5

Abbrev.: F; female, M; male, RLL; right lower lobe, LLL; left lower lobe, LUL; left upper lobe

ES was located on the right side in 9 cases (64.2%). The mean tumor size was 10.4 (range, 2-25) cm. All cases showed diffuse membranous staining only for CD99.

A 14-year-old male patient had a 2-cm lymph node on the superior to the pancreas. Abdominal ultrasound and magnetic resonance imaging were used to determine the nodule, and histopathological signification could not be done due to the localization of the nodule; however, no progression was seen on the 48th month follow-up period.

The endobronchial lesion was determined in a 20-year-old female patient via bronchoscopy, and a biopsy was performed (Figure 1). Three cycles of neoadjuvant chemotherapy were administered to the patient.

A 19-year-old male patient was admitted to the hospital due to chest pain, 45 days following a traffic accident. Chest x-ray revealed pleural effusion and tube insertion was performed due to hemothorax. Thorax CT revealed pleural effusion and a 14x12 cm heterogeneous solid lesion, suggesting a suspicious hematoma. Following this, a right thoracotomy was performed confirming the tumor. The tumor was totally excised with wedge resection of the 9-11th ribs. Postoperative PET/CT was used to determine distant metastasis and no abnormal FDG uptake was found (Figure 2). Surgical procedures included complete tumor excision with chest wall resection ($n = 10$), lower lobectomy with chest wall resection ($n = 1$), and complete tumor excision via wedge resection ($n = 3$).



Figure 1. Thorax CT showing a Ewing sarcoma in a 20-year-old female patient at the right lower lobe (Right lower lobectomy and 9th rib resection was performed).

Table 2. Treatment strategies and results of the patients who were operated for thoracic Ewing sarcoma.

Patient No	Surgical Procedure	Chemotherapy	Radiotherapy	Follow-up (months)	Recurrence	Secondary surgery	Mortality
1	Wedge resection	10	-	72	-	-	-
2	9th rib and surrounding soft tissue excision	12	-	17	-	-	-
3	5, 6, 7th rib resection	10	+	48	-	-	-
4	Lower lobectomy and 9th rib resection	6	-	16	On surgical incision site, 4th month	Metastectomy	16th month
5	Wedge resection	8 and bone marrow transplantation	-	84	Right lower lobe, 60th month	Wedge resection	-
6	5, 6, 7, 8th rib resection	8 and bone marrow transplantation	-	36	-	-	-
7	6, 7, 8th rib resection	15	-	48	-	-	-
8	Lower lobectomy	10	-	36	Multiple, bilateral nodules, 24th month	-	-
9	9, 10, 11th rib resection	4	+	18	Local recurrence, 16th month	-	18th month
10	3, 4, 5 rib resection	6	-	12	-	-	-
11	9, 10, 11th rib resection and wedge resection	10	-	23	-	-	-
12	Wedge resection	2	-	19	Pleura, 5th month	Decortication	-
13	2, 3rd rib resection and wedge resection	-	-	2	-	-	-
14	5th rib resection and wedge resection	6	-	12	-	-	-

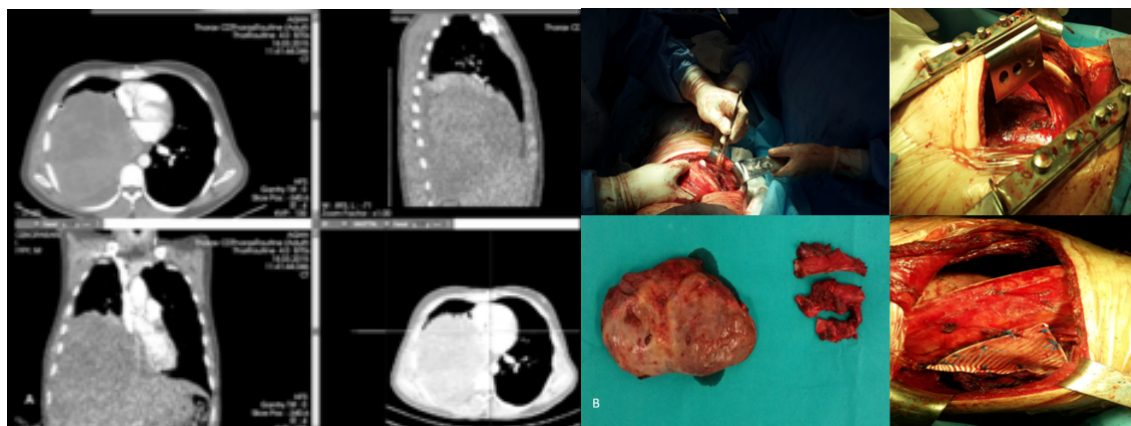


Figure 2. Thorax CT findings of a 19-year-old male patient. A 14 cm diameter tumor was seen in the right lower lobe causing compression on the heart and great vessels (A), Right thoracotomy and tumor excision with wedge resection and 9-10-11th rib resection was performed. A polypropylene mesh was used to repair the 10x15 cm chest wall defect (B).

A 21-year-old male patient had a giant tumor measuring 25 cm adjacent to the right lower lobe (Figure 3). Total tumor excision and 9-10-11th rib excision was performed. However, the recurrence was occurred on the 16th month and he died on the 18th month of the follow-up.

The 10 patients with ES originating from the ribs underwent complete tumor excision with chest wall resection, one patient underwent lower lobectomy with chest wall resection, and 3 patients without chest wall invasion underwent complete tumor excision via wedge resection. All patients were treated with chemotherapy postoperatively. Three cycles of neoadjuvant chemotherapy were used for a 20-year-old female patient (Table 1, case #4). Multi-drug chemotherapy, which is a minimum of four drugs such as vincristine, doxorubicin, dactinomycin, and cyclophosphamide, was used for all patients, except for two patients who underwent bone marrow transplantation. Radiotherapy was used for two patients due to narrow surgical margins and large tumor sizes.

Relapses were seen in 5 patients: 4 male and 1 female with a median age of 26.4 (range, 20-39) years. The average tumor size was 10.4 cm (range, 2-25 cm). The clinical features and treatment modalities are shown in Table 1. Chemotherapy was used in all patients, except for one patient who underwent additional bone marrow transplantation. The median follow-up was 31.6 (range, 2-84) months. Relapses were seen in 5 cases in the median 19.8th (range, 4-60) months. Three of them underwent secondary thoracic surgery procedures (redo-thoracotomy metastasectomy, contralateral wedge resection, and decortication). A 20-year-old female patient who underwent redo-thoracotomy and a 21-year-old male patient with a 25-cm chest wall tumor died on the 16th and 18th months, respectively, after diagnosis with a mortality rate of 14.3%.

Discussion

In this study, we report the clinical features, treatment strategies and outcomes, complications, and survival of 14 patients with thoracic ES who underwent surgical resection. In addition, we found that there was a male predilection. ES mainly occurs in children and young adults. Reportedly, ES is the 3rd most common primary malignant neoplasm of bone and soft tissues seen in children after osteosarcoma and rhabdomyosarcoma, respectively [6]. Extraskel-

(p)NETs) are usually present in the 2nd decade of life [6]. Familial cases have been reported, and Extraskel-etal ES/pNET may rarely occur as a 2nd malignant neoplasm [7,8].

The most common sites for ES are flat bones or long bones of the leg, mostly located in the pelvis, axial skeleton, and femur. Extraskel-etal ES incidence is variable and ranges from 1/5,000,000 to 1/10,000,000. In our study, ES developed or invaded the ribs in 8 patients (72.7%), whereas 27.3% of them were extraskel-etal. The typical clinical presentation for ES is pain or swelling around the tumor site [8]. In this study, cough was a more frequent complaint than swelling.

EES/pNET is often large at presentation, ranging from 1 to 40 cm in diameter and frequently larger than 10 cm [6]. In this study, the mean tumor size of extraskel-etal and skeletal ES was 6.3 (2-12) cm and 13.6 (5-25) cm, respectively.

Classic ES/pNET has patterns of formless sheets and lobules of uniform round to oval cells, and spindle cell areas may be present [9]. Mitoses are present in variable numbers and are inconspicuous in some cases [6]. The overlapping histologic, immuno-histochemical, and cytogenetic, and molecular genetics features cause diagnostic challenges despite significant clinical and prognostic differences. EES and PNETs are pathologically characterized by diffuse sheets of small round cells that express vimentin as well as CD99 (a MIC2 gene product). The diagnosis can be confirmed by the non-random chromosomal translocation involving the ES breakpoint region 1 gene; t (11;22) (q24;q12) [10].

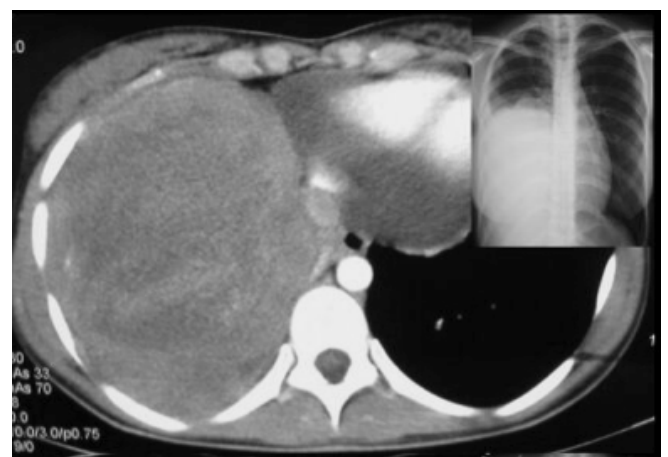


Figure 3. Thorax CT showing a huge Ewing sarcoma that was originated from 10th rib and 25 cm diameter, in a 21-year-old male patient.

A capsule or pseudo-capsule frequently appears in the resected tumor. In some cases, it was difficult to identify any residual gross tumor in the margin, which is important for the decision of radiotherapy [11,12]. Positive or close margins are generally managed by radiotherapy. In this study, radiotherapy was used for 2 patients with 13 and 25 cm ES on the chest wall due to close surgical margins. A 21-year-old male patient with a 25-cm tumor had a recurrence on the 16th month of the follow-up and eventually died on the 18th month (case # 9). Furthermore, a 14-year-old male patient with a 13-cm tumor had a 48-month EFS postoperatively.

There are limited studies concerning the management of primary chest wall ES. The treatment of ES is multimodal, which includes definitive surgery, radiation therapy, or a combination of both, followed by induction chemotherapy. Radiation therapy is applicable in unresectable tumors or those that prove to have margins of resection involved or closely approached by a tumor. Treatment condensation for high-risk patients may consist of high-dose chemotherapy, followed by stem cell transplant. In this study, 2 male patients aged 13 and 27 with 8 and 2-cm tumor sizes, respectively, underwent bone marrow transplantation [6,13,14]. The 27-year-old patient had a recurrence in the 60th month following left lower lobectomy, and wedge resection for the right lower lobe was performed. Both the patients were followed up for 36 and 84 months, respectively without mortality.

Prognosis is associated with age, localization, metastasis, tumor size, origin tissue of tumor, chemotherapy administered, and response to chemotherapy [15,16]. The current 5-year EFS rate for non-metastatic EWS/pPNET ranges from 60%–75% [17].

Surgical resection of chest wall ES can be performed as the initial therapy (up-front) or after the patient has received neoadjuvant chemotherapy (delayed). Delayed surgery can provide smaller resections and better margin negativity rates compared to up-front surgery [18]. Besides reducing the tumor volume and increasing the rate of negative margin resections (R0), the other benefit of neoadjuvant chemotherapy is decreasing the use of radiotherapy [1]. A study of 98 patients with chest wall ES showed that patients who underwent delayed surgery had a higher frequency of successful complete resection of the tumor and decreased requirement of radiation therapy to the chest [19]. The primary aim is to provide complete resection with adequate margins. Neoadjuvant

chemotherapy prevents the patient from radiation therapy and its potential long-term complications, including pulmonary fibrosis, increased incidence of coronary artery disease, and a significant incidence of secondary tumors [19]. Radiation therapy in doses recommended for the treatment of ES/PNET has been associated with a significant incidence of secondary tumors in these patients (10%–30%), and delayed resection resulted in a significant decrease in the proportion of patients requiring chest radiation therapy [19]. However, in this study, only one patient received neoadjuvant chemotherapy and relapse occurred in the 4th month postoperatively.

The main limitation of this study was its retrospective nature and the benefit of total resection of the affected ribs could not be proved.

In conclusion, although ES is an aggressive disease with a high incidence of local recurrence and distant metastases, long-term survival is achievable with appropriate and aggressive multimodal therapy. Relapses are not rare, but if they occur, surgical resection for secondary tumors can help improve survival. Surgical treatment with adequate margins in association with appropriate use of combined chemotherapy and postoperative radiotherapy or bone marrow transplantation, when indicated, is necessary for preventing relapses and achieving longer survival rates. Further prospective and larger sample size studies are recommended to determine the prognostic factors and treatment results of thoracic ES.

Declaration of conflicting interests

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Ethics approval

The study was approved by the institutional review board of Health Sciences University, Ataturk Chest Disease and Thoracic Surgery Training and Research Hospital (No: 03.12.2020/ 703).

Authors' contributions

SH, YA; co-wrote the paper, collected the data, performed the analysis, KA, PB; contributed data/analysis tools, GF; conceived and designed the analysis, co-wrote the paper, SŞEG, MŞD; collected the data, SK; conceived and designed the analysis.

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Original Article

Comparing the outcomes of video-assisted thoracoscopic surgery and re-thoracotomy in the management of postoperative hemorrhage

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ABSTRACT

Background: Although postoperative hemorrhage after thoracic surgery is uncommon, it is the most common indication for revision surgery after these procedures. Most postoperative hemorrhages are due to surgical technique, although some comorbidities can predispose the patient to bleeding. We investigated whether video-assisted thoracoscopic surgery (VATS) and re-thoracotomy had the same outcomes in the management of postoperative hemorrhage in patients who underwent open thoracotomy or VATS.

Materials and Methods: We retrospectively analyzed patients with postoperative hemorrhage after thoracotomy (n = 659) or VATS (n = 883) between 2018 and 2020. Revision surgery was performed after thoracotomy in 22 patients (3.3%) and after VATS in 4 patients (0.4%). Of these, 11 patients (42.3%) were re-operated by re-thoracotomy (Re-thoracotomy Group) and 15 patients (57.7%) by revision VATS (VATS Group).

Results: Revision due to postoperative hemorrhage was required significantly more frequently after thoracotomy than VATS (3.3% vs. 0.4%, p < 0.001). In patients with hemorrhage after pneumonectomy (n = 14), revision by VATS was preferred to re-thoracotomy (n = 10, 71.4% vs. n = 4, 28.6%). The mean time to discharge after revision surgery was 5.1 ± 2.2 days (range, 2-12 days) overall and was significantly shorter in the revision VATS Group than in the Re-thoracotomy Group (4.4 ± 1.5 days vs. 6.2 ± 2.5 days, p = 0.004).

Conclusions: VATS has similar results to re-thoracotomy and is advantageous in terms of earlier recovery and shorter hospital stay. Therefore, VATS should be the preferred method for postoperative hemorrhage management.

Keywords: VATS, re-operation, thoracic surgery, thoracotomy, postoperative hemorrhage

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Introduction

Postoperative hemorrhage is one of the most important complications of thoracic surgery and is reported in 1% to 3% of patients after elective thoracotomy procedures [1]. Hemorrhagic blood loss in excess of 1000 mL is referred to as massive hemorrhage and generally requires revision thoracic surgery, although the decision to re-operate depends on the patients' hemodynamic features, the presence of hematoma on chest x-ray, and the hourly rate of hemorrhagic drainage [2,3]. Re-thoracotomy and video-assisted thoracoscopic surgery (VATS) are both options for revision surgery. While emergency re-thoracotomy is still the gold standard for life-threatening postoperative hemorrhage, VATS is increasingly being used in clinically stable patients [4].

Our aim in this study was to compare the results of revision VATS and re-thoracotomy in the management of postoperative hemorrhage.

Materials and Methods

We reviewed the records of patients who underwent re-thoracotomy, VATS, or re-VATS due to postoperative hemorrhage between 2018 and 2020. During this period, a total of 1542 patients underwent primary VATS ($n = 883$) or thoracotomy ($n = 659$). Revision was required due to postoperative hemorrhage in 22 patients (3.3%) after thoracotomy and 4 patients (0.4%) after VATS. Of these, 11 patients (42.3%) underwent re-thoracotomy (Re-thoracotomy Group) and 15 patients (57.7%) underwent revision by VATS (VATS Group). The study was approved by the institutional review board (number/date: 2020-35/15.10.2020) and conducted in accordance with the principles of the Declaration of Helsinki.

Patients who were treated with intrapleural thrombolytic agents or were followed up medically instead of undergoing surgical revision were excluded from this study.

Before the primary surgery, all patients were routinely evaluated for respiratory capacity and cardiac functions. If resection was planned, a forced expiratory volume in 1 second (FEV1) of at least 60% was sought. Cardiac functions were monitored via electrocardiography, and patients were referred to the cardiology department for echocardiography when necessary. Prothrombin time, international normalized ratio, and partial thromboplastin time were checked routinely in preoperative evaluations. Management of patients using antico-

agulant therapy was arranged before surgery. Warfarin was discontinued 3 days before and antiplatelets such as acetylsalicylic acid and clopidogrel were discontinued 7 days before the procedure [6]. For patients undergoing coronary procedures (e.g., angioplasty, stenting), their cardiologists were contacted before interrupting antiplatelet therapy and their management was planned according to the nature of the planned surgery (elective or emergent). Enoxaparin sodium was routinely initiated at 30 to 40 mg/day subcutaneously for prophylaxis before planned surgery unless the procedure was short and/or the patient was young [5]. If lung resection was planned, enoxaparin sodium was initiated regardless of the patient's age.

As neoadjuvant chemotherapy and radiotherapy may pose a risk for postoperative hemorrhage, surgery was performed 4 to 6 weeks after neoadjuvant therapy. Patients who underwent resection and elderly patients with comorbidities were admitted to the intensive care unit postoperatively for at least one night. Drainage and hemodynamic parameters were routinely monitored by intensivists, intensive care nurses, and thoracic surgery residents. If hemorrhagic drainage from the chest tube exceeded 100 mL/hour, the on-call team was alerted to the possibility of hemorrhage in the patient. Patients with ongoing drainage at a rate of 100 mL for 8 hours or 200 mL for 2 to 4 hours were considered for revision. The decision to re-operate and using which surgical method was made by the thoracic surgery clinical council in consideration of the amount of residual hematoma on x-ray, the patient's hemodynamic parameters, and the volume of hemorrhagic drainage per hour. While re-thoracotomy remains the first option in life-threatening emergencies, VATS has become the preferred choice for revision in recent years, especially in pneumonectomy patients.

In re-thoracotomy, the previous incision was completely re-opened. For revision VATS in patients with primary thoracotomy, the chest tube incision was enlarged slightly for insertion of the VATS instrumentation, and another thoracoport incision was made if necessary. For re-VATS in patients who underwent primary VATS, the chest tube incision was used and the previous utility thoracotomy was re-opened if necessary. After the clots were evacuated, the remaining lobe was re-expanded in patients who underwent primary lobectomy and the chest cavity was carefully evaluated. If a specific bleeding site was detected, it was managed using cau-

terization, stitches, hemoclip, or by applying oxidized regenerated cellulose (Surgicel® Ethicon, Somerville, NJ), depending on location.

Regardless of the primary surgery, all patients were monitored in the intensive care unit after revision surgery and transferred to the ward the following day if hemodynamically stable. Chest tubes were removed when drainage was less than 100 mL/day and no air leak was detected. Patients were discharged after chest tube removal if they were clinically stable.

Patients were called to the thoracic surgery outpatient clinic for follow-up between 7 and 10 days after discharge and routinely evaluated using chest x-ray and laboratory tests. Chest computed tomography was requested if necessary.

Statistical Analysis

The patients' demographic characteristics and operative data were entered into IBM SPSS Statistics version 25.0 (IBM Corp, Armonk, NY) for analysis. Qualitative variables were expressed using frequency and percent distribution. Normally distributed continuous variables were reported as mean $\pm$ SD and Student's t-test was used for comparison of groups. Chi-square was used for the analysis of qualitative variables and Fisher's exact test was used if the group was small ($n < 5$). Nonparametric continuous variables were recorded as median and minimum - maximum values and were compared by using Mann-Whitney U tests. P value less than 0.05 was accepted as statistically significant. Odds ratio (OR) was used for the risk ratios of the differences detected in the comparisons.

Results

Between 2018 and 2020, a total of 1542 patients underwent VATS ($n = 883$) or thoracotomy ($n = 659$). The mean age was 53.5 ± 8.6 years (range, 14-82 years) and the majority of the patients were male ($n = 1058$, 70.1%). There were 22 patients (3.3%) who required revision after thoracotomy, while only 4 patients (0.4%) required revision after VATS ($p < 0.001$) (Table 1).

Of the 659 primary thoracotomies, 446 (67.7%) were anatomical lung resections (120 pneumonectomies, 326 lobectomies). Of the 883 VATS procedures, 141 (16%) were anatomical lung resections (1 pneumonectomy and 140 lobectomies). Revision was required due to postoperative hemorrhage after 3 (1 pneumonectomy,

2 lobectomies) of the 141 VATS resections (2.1%) and after 19 (13 pneumonectomies, 6 lobectomies) of the 446 thoracotomy resections (4.2%). There was no significant difference in postoperative hemorrhage rates between resection with VATS and thoracotomy ($p = 0.315$) (Table 1).

Postoperative hemorrhage occurred more frequently after pneumonectomy than lobectomy (14 revisions after 121 pneumonectomies [11.5%] vs. 8 revisions after 466 lobectomies [1.7%]). There was a significant difference in the frequency of postoperative hemorrhage between lobectomy and pneumonectomy patients ($p < 0.001$) (Table 1).

A total of 26 patients (84.6% men) underwent revision after thoracotomy or VATS due to postoperative hemorrhage. Of these 26 patients, 15 (57.7%) underwent revision by VATS (VATS in 12 patients, re-VATS in 3 patients) and 11 (42.3%) by re-thoracotomy. The mean age was 57.8 ± 12.1 years (range 24-74 years) for all re-operated patients, 59.6 ± 9.7 years (range 37-71 years) in the revision VATS Group, and 55.5 ± 15.0 years (range 24-74 years) in the Re-thoracotomy Group. There was no significant difference between the revision VATS and Re-thoracotomy Groups in terms of patient number, age, or sex ($p > 0.05$, table 2).

Revision was performed after lobectomy in 8 patients (revision VATS in 4 [50%], re-thoracotomy in 4 [50%]) and after pneumonectomy in 14 patients (revision VATS in 10 [71.4%], re-thoracotomy in 4 [28.6%]). Although the difference was not statistically significant, VATS was more preferred for the management of post-pneumonectomy hemorrhage ($p = 0.386$).

Postoperative hemorrhage was observed in 4 patients after operations other than anatomic lung resection (VATS thymectomy in 1 patient, decortication in 2 patients, and surgical treatment of penetrating injury in 1 patient). The patient with penetrating injury underwent re-thoracotomy due to persistent hemorrhage after the primary operation, and the source of the bleeding was identified as oozing from the parenchyma.

The mean drainage volume was 1250 ± 327.7 mL (range, 900-2300 mL) overall, 1166 ± 252.6 mL (range, 900-1700 mL) in the revision VATS Group, and 1363 ± 393.1 mL (range, 900-2300 mL) in the Re-thoracotomy Group ($p = 0.259$).

Table 1. Distribution of patients who underwent primary and revision surgery.

Variables	Total (n=1542)	VATS (n=883)	Thoracotomy (n=659)	P value	OR	95%CI
Age, mean±SD	53.5±8.6	50.3±6.5	56.5±4.3	0.001	na	na
Sex, n (%)				0.01	na	na
Male	1058 (70.3)	562 (63.6)	496 (75.2)			
Female	484 (29.7)	321 (36.4)	163 (24.8)			
Revision required due to postoperative hemorrhage, n (%)	26 (1.7)	4 (0.4)	22 (3.3)	<0.001	7.589	2.603 - 22.131
Anatomical lung resections, n (%)	587 (38)	141 (16)	446 (67.7)	na	na	na
Lobectomy						
Pneumonectomy	466 (79.4) 121 (0.6)					
Revision required after anatomical lung resection due to postoperative hemorrhage, n (%)	22 (37.4)	3 (2.1)	19 (4.2)	0.315	2.047	0.597 - 7.022
Postoperative hemorrhage, n (%)				<0.001	7.491	3.064
After lobectomy	8 (1.7)	2 (1.4)	6 (1.8)			-
After pneumonectomy	14 (11.5)	1 (100)	13 (11.6)			18.310

Abbrev.: CI: Confidence interval, n: Number, na: Not applicable, OR: Odds ratio, pneumty: Pneumonectomy, VATS: Video-assisted thoracoscopic surgery, SD: Standard deviation

Table 2. The demographic characteristics and revision surgery details of the patients.

Variables	Total (n=26)	VATS Group (n=15)	Re-thoracotomy Group (11)	P value	OR	95%CI
Age (years), mean±SD	57.8±12.1	59.6±9.7	55.5±15.0	0.610	na	na
Sex, n (%)					1.00	1.00
Male	22 (84.6)	13 (86.6)	9 (81.8)	1.00		
Female	4 (15.4)	2 (13.4)	2 (18.2)			
Primary resection*				0.386	2.500	0.410
Lobectomy	8 (36.3)	4 (50.0)	4 (50.0)			-
Pneumonectomy	14 (63.7)	10 (71.4)	4 (28.6)			15.230
Mean amount of drainage (mL)	1250±327.7	1166.6±252.6	1363.6±393.1	0.259	na	na
Mean revision time, (days)	2.2±1.4	2.3±1.3	2.0±1.5	0.540	na	na
Mean discharge time after re-operation (days)	5.1±2.2	4.4±1.5	6.2±2.5	0.004	na	na

Abbrev.: CI: Confidence interval, n: Number, na: Not applicable, OR: Odds ratio, VATS: Video-assisted thoracoscopic surgery, SD: Standard deviation

* Only includes resection patients who underwent postoperative revision.

The overall mean time from primary surgery to revision was 2.2 ± 1.4 days (range, 1-6 days). This interval was 2.3 ± 1.3 days (range, 1-4 days) in the revision VATS Group and 2.0 ± 1.5 days (range, 1-6 days) in the Re-thoracotomy Group ($p = 0.540$).

The mean discharge time after revision was 5.1 ± 2.2 (range, 2-12) days for all re-operated patients, 4.4 ± 1.5 days (range 2-8 days) in the revision VATS Group, and 6.2 ± 2.5 days (range, 3-12 days) in the Re-thoracotomy Group. This time was significantly shorter in the VATS

Group than the Re-thoracotomy Group ($p = 0.004$).

Details related to the patients' demographic characteristics and revision surgeries of are shown in table 2.

A specific bleeding site was identified in 11 (42.3%) of the patients during revision. This site was the chest wall in 4 patients (36.3%), intercostal vessels in 2 patients (18.2%), bronchial vessels in 2 patients (18.2%), the subcarinal lymphatic bed in 1 patient (9.1%), the brachiocephalic vein in 1 patient (9.1%), and lung parenchyma in 1 patient (9.1%).

Three of the patients had a history of neoadjuvant chemotherapy. All 3 patients underwent primary pneumonectomy by thoracotomy followed by revision VATS.

Discussion

Hemothorax is one of the most important postoperative complications in thoracic surgery, especially after resections [6]. The decision to re-operate depends on the amount of hemorrhagic drainage, the presence of hematoma in the hemithorax, and the hemodynamic stability and clinical condition of the patient. Although there are some guidelines for the management of postoperative hemothorax, they are controversial. Medical follow-up may still be an option for patients with approximately 1000 mL of blood loss if the hemorrhage seems likely to stop [7]. With recent advances in VATS experience, it has begun to replace re-thoracotomy except in cases of emergency life-threatening bleeding.

According to the literature data, the incidence of postoperative hemorrhage is approximately 2% to 3% after thoracotomies and 0% to 1.7% after VATS procedures [8-10]. In the present study, re-operation rates due to postoperative hemorrhage were 3.3% after thoracotomy and 0.4% after VATS procedures. Hemorrhage rates after VATS may be lower because patients selected for VATS have relatively easier surgical procedures compared to the thoracotomy patient group. Consistent with the literature, the lung tumors in our VATS patients were smaller and more peripherally located [11]. In addition, there was only 1 pneumonectomy patient in the revision VATS Group. In the National Veterans Affairs Surgical Quality Improvement Program study, 3500 patients were retrospectively evaluated and the frequency of postoperative hemorrhage was 2.9% after lobectomy and 3% after pneumonectomy [12]. In the present study, postoperative hemorrhages were more common after pneumonectomy ($n = 14$, 11.5%) compared to after lobectomy ($n = 8$, 1.7%). Our center is one of the high-volume hospitals in Turkey for lung cancer surgery. Patients who are receiving neoadjuvant therapy and require extended pneumonectomy are referred to our hospital from many clinics. In addition, the prevalence of squamous cell carcinoma is higher in Turkey than in European countries due to our high smoking rate. Because patients with squamous cell carcinomas present with large tumor size, they often require pneumonectomy. Therefore, our higher rate of postpneumonectomy

hemorrhage can be attributed to our high percentage of patients undergoing pneumonectomy and receiving neoadjuvant therapy.

In the present study, VATS was preferred over re-thoracotomy for patients with postoperative hemorrhage due to pneumonectomy. This is reasonable considering VATS provides excellent visibility of the entire hemithorax. In addition, clots can easily be evacuated through a single port, and we used the previous chest tube incision for this purpose.

In general, the reported surgical indications for posttraumatic hemothorax also apply to postoperative bleeding. The criterion for considering revision surgery is ongoing hemorrhagic drainage of 100 mL for 8 hours or 200 mL for 2 to 4 hours [13]. The mean volume of hemorrhagic drainage in this study was 1250 mL (1166 mL in the revision VATS Group and 1363 mL in the Re-thoracotomy Group). The volume of hemorrhagic drainage was lower in the revision VATS Group because most were pneumonectomy patients and some of the blood collected in the pneumonectomy space as hematoma. It should be noted that large amounts of blood, as much as 30% to 40% of the circulating blood volume, can accumulate in the pleural space [13]. Beyond the guidelines, physician observation is crucial in routine practice. In some cases, careful observation may prevent unnecessary surgical revision if hemorrhagic drainage is approximately 1000 mL but appears likely to stop. However, a patient may also present with hemorrhage volume below the indication for revision surgery, but in fact the hemorrhage may be retained as a hematoma. Even if such a hemorrhage could resolve with follow-up alone, it might turn into empyema and fibrothorax. Therefore, the decision to re-operate or follow up is crucial in this patient group. Surgical revision may be an option for removing clots even when the bleeding has stopped [14,15]. Six of the patients in this study underwent revision for the purpose of clot removal even though the bleeding had stopped 4 to 6 days earlier.

In the current study, the mean time to revision surgery was found to be 2.23 days, with no significant difference between the revision VATS and Re-thoracotomy Groups. The timing of VATS is very important, and it may be more effective for evacuating clots within the first 2 to 3 days [16]. Some authors have suggested that

VATS would not be feasible after day 7 due to denser clots, thicker pleura, and adhesions that would prevent safe insertion of the VATS equipment. However, when performed before day 7, authors have reported that VATS has significant benefits over thoracotomy [17,18]. Morales et al. [19] reported a 15.8% rate of conversion to open thoracotomy after day 6. We did not encounter any difficulties using VATS for revision within a 5 to 6 day period. However, these patients were pneumonectomy patients. On the other hand, one patient required conversion to thoracotomy on the first postoperative day of hemorrhage. This patient had undergone VATS thymectomy and was bleeding from the junction of the brachiocephalic vein and vena cava superior where the thymic vein originates. Because it could not be controlled with VATS, the procedure was converted to an open thoracotomy.

VATS requires selective intubation, which adds time before the surgery can be initiated. For this reason, thoracotomy should be the first choice rather than VATS for the management of hemorrhage in unstable patients [20]. There were no such cases in the present study.

The specific site of bleeding usually cannot be identified during revision surgery. Sirbu et al. [21] found that 23% of hemorrhages originated from a mediastinal or bronchial vein and 17% from intercostal veins. However, they could not find any specific bleeding site in 41% of the cases. Hemorrhage may also occur as oozing from vascular stapler lines, as bleeding from lymphatic beds, peribronchial tissue, parenchyma, or adhesions, or as a muscle bleed [22]. Similarly, we did not find any specific site of bleeding in most of our patients. We just evacuated the clots and cauterized suspicious areas, mostly on the chest wall.

Many studies have pointed to the benefits of VATS over thoracotomy: shorter hospitalization, less pain, earlier recovery, lower costs, better lung function, better visualization, and a higher rate of detection of small injuries [23,24]. In the present study, the length of hospital stay after revision surgery was significantly shorter in patients who underwent revision VATS group compared to those who underwent re-thoracotomy (3 days vs. 5 days).

As with every retrospective data analysis, this study also has its limitations. Another strategy for managing postoperative hemothorax is to use intrapleural throm-

bolytic agents, but the possible adverse effects of this management strategy are controversial, as it may increase bleeding [25]. Although we have experience with this treatment technique, we did not include these patients because the aim of this study was to determine the difference in outcomes between VATS and re-thoracotomy procedures in postoperative hemorrhage management. Previous studies comparing VATS with thoracotomy in the management of hemothorax have mostly focused on traumatic hemothorax. Investigating the management of postoperative hemorrhage alone is one of the notable features of this study.

In conclusion, VATS was found to be superior to re-thoracotomy in the management of postoperative hemorrhage in terms of earlier recovery time and shorter hospital stay. Considering the advantages of this minimally invasive approach and its similar outcomes with re-thoracotomy, we believe that VATS should be the preferred technique in postoperative hemorrhage management, especially in pneumonectomy patients.

Declaration of conflicting interests

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Ethical approval

The study was approved by the Ethics Committee of Health Sciences University (No: 2020-35/15.10.2020) and conducted in accordance with the principles of the Declaration of Helsinki.

Authors' contributions

VE; Conceived and designed the analysis, co-wrote the paper, MSO; conceived and designed the analysis, AÇ, SE; collected the data, AP, EYE; contributed data/analysis tools, YA, MVD; performed the analysis, MM, ACK; co-wrote the paper.

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Original Article

Comparison of thoracotomy and videothoracoscopy for intrathoracic bronchogenic cysts

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ABSTRACT

Background: Bronchogenic cyst is a generally benign and rare congenital anomaly. The mediastinum and lung parenchyma are the most common sites the cysts occur. This study aimed to compare the effectiveness of thoracoscopy and thoracotomy for the treatment of bronchogenic cysts.

Materials and Methods: Twenty-one patients were operated on in Dr. Suat Seren Thoracic Surgery Clinic between 2004 and 2019 with the diagnosis of bronchogenic cyst. Patients were divided into two groups according to the surgery method as video-assisted thoracoscopic surgery (VATS) or thoracotomy. Demographic and operative features, radiological findings, and complications were retrospectively analyzed.

Results: Patients underwent either VATS (10 patients) or thoracotomy (11 patients). Sixteen patients underwent simple cyst excision, whereas parenchymal resection was performed for five patients. The mean Visual Analogue Scale (VAS) value was 3.5 ± 1.1 for the VATS group and 6.5 ± 1 for the thoracotomy group. Duration of hospital stay and chest drainage was 4.2 ± 1.7 and 3.2 ± 1.7 days, respectively, in the VATS group, while in the thoracotomy group, it was 7.1 ± 4.5 and 6.1 ± 4.5 days. The time of hospitalization stay, duration of chest drainage, operative time, and VAS values were found statistically significantly lower in the VATS group.

Conclusions: Both VATS and thoracotomy are preferred surgical methods in bronchogenic cyst surgery. VATS is superior to thoracotomy in terms of shorter hospital stay, chest drainage duration, operative time, and lower postoperative pain level. Minimally invasive methods can be chosen safely for the treatment of bronchogenic cyst in appropriate cases.

Keywords: bronchogenic cyst, videothoracoscopy, thoracotomy

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Introduction

The bronchogenic cyst is a rare congenital anomaly resulting from abnormal branching of the tracheobronchial tree developing from the ventral part of the embryological foregut between the 26th and 40th days of gestation [1]. The bronchogenic cyst is generally located in the middle and posterior mediastinum or lung parenchyma. It is diagnosed incidentally since it is often asymptomatic in adults.

Some authors claim conservative treatment for asymptomatic patients. However, video-assisted thoracoscopic surgery (VATS) and thoracotomy are the most preferred treatment options for symptomatic or asymptomatic patients of which can be complicated or developing malignancy. Robotic surgery can also be considered an option in recent years.

In this study, 21 patients operated on with the bronchogenic cyst diagnosis were analyzed in terms of demographic and surgical features. It is aimed to compare the surgical methods accompanied by a literature review.

Materials and Methods

Data Collection

The patients' records operated in the thoracic surgery clinic of Dr. Suat Seren Chest Diseases and Surgery Hospital between 2004 and 2019 with the diagnosis of mediastinal, parenchymal, or diaphragmatic lesions were reviewed retrospectively. Twenty-one patients whose histopathological diagnosis confirmed as bronchogenic cysts were included in the study. The patients were divided into two groups as those operated by VATS or thoracotomy. The clinical features of these patient groups were reviewed as demographic characteristics at the time of operation (age, gender), preoperative symptoms, anatomical location of the cyst, cyst diameter, operative method, chest tube drainage duration, postoperative hospital stay, postoperative complications, and recurrence. All patients underwent routine blood tests, posteroanterior (PA) and lateral chest X-ray, and thorax computed tomography (CT) preoperatively. Magnetic resonance imaging (MRI) is used for determining the nature of the cyst. The study was approved by the institutional review board (No: 2019/5.1) and conducted per the principles of the Declaration of Helsinki.

Operation Method

Following the double-lumen intubation, all patients

were operated on in the right or left lateral decubitus position. One of the patients with a mediastinal bronchogenic cyst of subcarinal position was operated in the supine position. As the operation method, thoracotomy was performed before 2010, whereas VATS was preferred for suitable cases after this year. Depending on the cyst's anatomical location, fourth or fifth intercostal space was used for thoracotomy. In the VATS approach, a utility incision (4th or fifth intercostal space) and a camera port of length 1 cm on the sixth or seventh intercostal space of the midaxillary line were used. The absence of air leakage for at least 24 hours and pleural drainage less than 200 mL per day were considered as the criteria to remove the chest tube. The air leakage that lasted seven days or more was considered as prolonged air leakage. The patients' postoperative pain level was determined according to the subjective assessment method of the Visual Analogue Scale (VAS).

Statistical Analysis

Statistical Package for the Social Sciences (SPSS) version 22.0 program was used to analyze data. Continuous data were specified as mean $\pm$ standard deviation, and median (minimum-maximum) values. Mann-Whitney U test was used to compare differences between groups and Fisher's Exact test for dichotomous variables. A p-value less than 0.05 was considered as statistically significant.

Results

The mean age of 11 female and ten male patients was 41.3 ± 18.6 years (18-78). Eighteen of the patients (85%) were asymptomatic consistent with the literature. Fever, chest pain, dyspnea, cough were the complaints of symptomatic patients. All the patients underwent a thorax CT. MRI was also used for eight patients in which the nature of the cyst could not be clearly identified. MRI confirmed all the lesions as cyst successfully. Radiological imaging examples of mediastinal and parenchymal cysts are presented as figures 1 and 2.

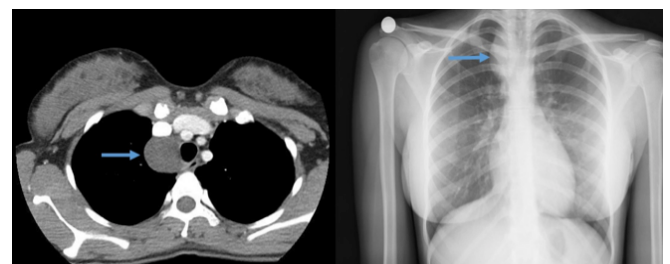


Figure 1. Preoperative imaging of a mediastinal cyst.

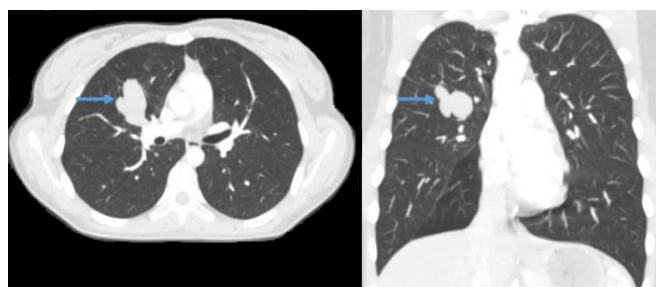


Figure 2. Preoperative imaging of a parenchymal cyst.

The median cyst diameter was 4.5 cm (1-10). None of the cysts had any tracheobronchial connection. Comparative characteristics of patients who underwent VATS and thoracotomy are summarized in table 1.

Patients underwent either thoracotomy (11 patients) or VATS (10 patients). Simple cyst excision (16 patients), wedge resection (3 patients), lobectomy (1 patient), and lobectomy and segmentectomy (1 patient) were the surgical procedures performed. The anatomical localization of the cyst is presented in table 2. A bronchial laceration was repaired in one patient intraoperatively, and that patient's bronchial system was observed to be intact by bronchoscopy in the postoperative period. Five patients had prolonged air leakage as the postoperative complication (23%). The median duration of postoperative hospitalization was 6 days (2-19). VATS patients' hospital stay duration ($p = 0.03$), drainage duration ($p = 0.03$), operative time ($p = 0.013$), and VAS score ($p < 0.001$) of the patients operated by VATS were found to be statistically significantly lower than thoracotomy group. There was no statistically significant difference between the two groups regarding age, gender, cyst diameter, and postoperative complications.

No recurrence was observed in any of the patients during the median follow-up of 94.5 months (15-190).

Discussion

The bronchogenic cyst is a rare, benign lesion that arises from abnormal budding of the tracheobronchial tree. The most commonly affected site is mediastinum, but the location may be parenchymal in 15-20% of the cases [2]. It is mainly located in the paratracheal and subcarinal regions of the mediastinum. Although mediastinum and parenchyma are the primary localizations, they may be seen on ectopic locations such as retroperitoneum, neck, cranial fossa, pericardium [3, 4]. The majority of the patients in our study (71%) had mediastinal cysts, and most of them were located at the paratracheal region, similar to the literature review. Besides, the cyst was observed on the diaphragm in one patient.

The clinical manifestations may vary, and the cysts are usually asymptomatic in adults until they reach large sizes and compress other structures or be infected due to having a bronchial connection. The most common symptoms are chest pain, cough, dyspnea, dysphagia, hemoptysis, depending on compression and location of the cyst [5]. Also, it has been reported in the literature that they may cause arrhythmia due to cardiac pressure when they reach large dimensions [6]. Several studies reported that the majority of asymptomatic patients will develop symptoms over time [7]. Only three (14%) patients had symptoms in our series, all located in the pulmonary parenchyma. Fever, chest pain, cough, and dyspnea were the complaints of symptomatic patients, consistent with the literature.

Table 1. The characteristics of patients who underwent VATS and thoracotomy.

	VATS (n=10)		Thoracotomy (n=11)		p-value
	n (%)		n (%)		
Gender (male)	5 (50.0)		5 (45.4)		0.55
Presence of symptoms	0 (0.0)		3 (27.2)		0.72
Side (right)	8 (80.0)		8 (72.7)		0.81
Postoperative complications	1 (10.0)		4 (36.3)		0.23
	Mean ± SD	Median (min-max)	Mean ± SD	Median (min-max)	
Age (years)	46.9±23.5	44 (18-78)	38.1±13.5	33 (20-68)	0.47
Cyst Diameter (cm)	3.8±1.7	4 (2-8)	5.28±2.9	4.8 (1-10)	0.14
VAS value	3.5±1.1	4 (2-6)	6.50±1	6 (3-8)	<0.001
Hospitalization (days)	4.2±1.7	4 (2-7)	7.0±4.5	7 (5-19)	0.03
Drainage duration (days)	3.2±1.7	3 (1-5)	6.1±4.5	6 (3-8)	0.03
Hounsfield Unit	33.6±10	32.2 (10-80)	44.4±27.1	42.8 (20-90)	0.66
Operative time (minutes)	102.7±36.3	100 (60-170)	200.5±111.1	180 (150-260)	0.013

Table 2. The anatomical localization of the cysts.

Mediastinum (n=15)	Parenchyma (n=5)	Diaphragm (n=1)
Paratracheal	9	Right lower lobe 2
Subcarinal	4	Middle lobe 1
Hilar	1	Left upper lobe 1
Paraesophageal	1	Left lower lobe 1

Preoperative diagnosis of bronchogenic cysts may be challenging due to having a wide range of radiological and clinical features. Thoracic computed tomography (CT) and magnetic resonance imaging (MRI) have an essential value in determining the size, shape, content, and relationship of the cyst with surrounding tissues [8]. Attenuation coefficient or Hounsfield Unit (HU) value on CT can be a marker to determine the lesions as solid or cystic [9]. While cyst at mediastinum localization is generally presented as well-defined masses, the air-fluid level is the radiological finding of parenchymal cysts connected with bronchi. In case of infection, higher HU values can be obtained on CT due to protein and calcium within the cyst in contrast to low HU like water or soft tissue of cystic lesions. Higher HU may be evaluated in favor of semisolid or solid masses. MRI provides more valuable information than CT when solid or cystic lesions cannot be differentiated [11]. The median and mean HU values were obtained as 34 (10-90) and 39 ± 20.7 , respectively, in our study. This value was higher than the water density and 20 HU in which was evaluated in favor of a cyst. This result may be related to the proteinous content in the cyst. Cases of bronchogenic cysts with high HU values have been reported in the literature similarly to our study [11].

The primary treatment for symptomatic or complicated cases is surgery to relieve symptoms and prevent complications such as infection and compression. However, on the other hand, there is still no consensus regarding the management of asymptomatic patients. Wang et al. suggested that a conservative approach may be appropriate provided that radiological follow-up is performed for this group of patients since the malignant transformation is not very frequent in asymptomatic patients [12]. On the other hand, opposite opinions have claimed that surgery should be performed instead of conservative treatment [13] since adenocarcinoma may arise from bronchogenic cyst [14]. Surgery was performed for 18 patients in our study, even if they had no symptoms, considering the possibility of developing

malignant degeneration and symptoms over time. As we did not follow up asymptomatic patients conservatively, we do not have sufficient data on long-term outcomes of non-operated asymptomatic patients. We favor surgery for patients even though they are asymptomatic since the malignant transformation is a possibility. Although conservative treatment may be an option for elderly asymptomatic patients, thoracotomy was performed on a 78-year-old symptomatic male patient to relieve his symptoms in our study.

The appropriate treatment approach is the complete removal of the lesion for mediastinal cysts and anatomical or wedge resection for parenchymal cysts. Wedge resection is adequate for small-sized peripheral parenchymal lesions. Lobectomy should be chosen for centrally located parenchymal lesions. While lobectomy is usually performed as anatomical resection, segmentectomy may also be preferred in appropriate cases. Segmentectomy can be performed if the cyst does not exceed the boundary of the segment of the involved lobe. In our study, simple cyst excision was performed for mediastinal and diaphragmatic cysts, while patients with parenchymal cysts underwent pulmonary resection.

Complete removal of the cyst should be the primary goal since recurrence in postoperative follow-up may occur in a cyst that has not been excised totally [15]. However, adhesions with major structures may prevent complete excision of the cyst, especially in infected patients. In this case, destruction of the mucosal layer of the cyst by using electrocautery is suggested to prevent recurrence [16]. According to Fievet et al [17], early excision of the cyst between the 6th and 12th month of life could prevent surgical complications and incomplete cyst excision, eliminating the need for electrocautery use.

The cyst was totally excised in all patients of our study. No recurrence was obtained in any patient during the median follow-up of 94.5 months (15-190).

The preferred surgical approaches are generally VATS and thoracotomy. Duration of chest drainage for the VATS group is reported to be shorter than thoracotomy group in the literature [18]. In addition, the VATS group is superior to thoracotomy in terms of shorter operative time, shorter postoperative hospital stay, and less intraoperative blood loss [19]. When comparing VATS and thoracotomy groups in our study, the VATS patients had statistically significant shorter hospital stay, shorter

operative time, lower VAS scores, and shorter chest drainage duration ($p < 0.05$). VATS clearly has advantages, including low pain severity and good exposure to the thoracic cavity compared to thoracotomy [20].

Adhesions due to infection can lead to complications, especially in symptomatic patients. One patient with a mediastinal cyst located on the subcarinal region suffered an iatrogenic injury of the bronchus in our series, similar to Granoto et al, who reported bronchus injury on two patients due to sharp dissection for adhesions of mediastinal cysts [21]. The bronchial laceration was repaired intraoperatively by thoracotomy before the patient was discharged without any other complication in the postoperative period. Besides, a prolonged air leak was observed in five patients.

Since adhesions due to infection sometimes make the thoracoscopic approach difficult, a thoracotomy may be needed. At this point, robotic surgery can be considered an alternative to thoracotomy in the advantages of 3D image quality and the manoeuvrability of the instruments used [22].

Today, bronchogenic cyst operation, with the development of thoracoscopic surgery, is performed safely in a minimally invasive manner, and patients are discharged in a short time without any severe complications.

In conclusion, surgery should be performed to both confirm the diagnosis and prevent complications and malignant transformations. Since videothoracoscopy is superior to conventional thoracotomy in the ways of shorter hospital stay, shorter chest drainage duration, shorter operative time, and lower VAS score, minimally invasive methods can be chosen safely as the first choice for the treatment of bronchogenic cyst in appropriate cases.

Declaration of conflicting interests

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

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Ethics Approval

The study was approved by the Ethics Committee of University of Health Sciences, Dr. Suat Seren Chest Disease and Thoracic Surgery Training and Research

Hospital (No: 2019/5.1) and conducted in accordance with the principles of the Declaration of Helsinki.

Authors' contributions

HM; Collected the data, co-wrote the paper, conceived and designed the analysis, KCC; Co-wrote the paper, performed the analysis, ŞÖK; Contributed data or analysis tools, AGY: Contributed data or analysis tools.

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Original Article

Efficiency of vocal cord medialization for recurrent laryngeal nerve dysfunction following pulmonary resections

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ABSTRACT

Background: Evaluating the incidence of postoperative vocal cord dysfunction after pulmonary resections and the impact of timing for vocal cord medialization on preventing postoperative pulmonary complications for these patients.

Materials and Methods: Patients developing vocal cord dysfunction (VCD)/unilateral vocal cord paralysis (UVCP) after pulmonary resection were examined retrospectively, in terms of postoperative pulmonary complication (PPC) rates and hospital length of stay. Total of 2740 patients underwent anatomical pulmonary resection for malignancy. Eleven patients were referred to otolaryngology team with pre-diagnosis of VCD following the operation. UVCP diagnosis was confirmed with indirect laryngoscopic examination.

Results: UVCP diagnosis was confirmed in 8 (0.3%) with indirect laryngoscopic examination. Performed resections were left upper lobectomy in 3 and left pneumonectomy in 5 patients. Atelectasis necessitating bronchoscopy and pneumonia were the PPC, seen in 3 (37.5%) patients. Calcium hydroxyapatite injection for 6 patients and polytetrafluoroethylene graft implantation for 2 patients was performed. Mean duration between pulmonary resection and medialization was 5.3 days in patients developing PPC and 3.6 days in patients with no PPC ($p = 0.011$). All patients were discharged within an average of 8.1 (6-13) days, uneventfully. One patient required re-injection of calcium hydroxyapatite on 5th month. Throughout a mean follow-up duration of 14.8 months, all patients had stable vocal cord position.

Conclusions: Vocal cord medialization can be performed safely for postthoracotomy UVCP. In order to minimize phonetic and respiratory complications, this procedure must be applied on early postoperative period.

Keywords: acquired vocal cord palsy, complications, medialization laryngoplasty, thoracotomy

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Introduction

Postoperative pulmonary complications (PPC) are the major source of morbidity in patients undergoing pulmonary resections due to malignancy. Atelectasis, bronchospasm and pneumonia incidence rate vary between 5% and 80% in literature, with 20% leading to respiratory failure and mortality among those patients [1-3]. Many risk factors associated are listed: Postoperative pain, immobilization, insufficient respiratory exercise, concomitant pulmonary diseases (e.g. chronic obstructive pulmonary disease or asthma), compromised tracheal ciliary activity, impaired glottis function due to endotracheal intubation, and orogastric aspiration [2-6].

Recurrent laryngeal nerve (RLN), which is a branch of Vagus nerve, is responsible for innervations of the larynx and vocal cords. Damage to this nerve during a thoracic operation, especially on the left hemithorax due to its anatomy, can cause phonetic problems, aspiration and respiratory distress. Repositioning of the paralytic cord is recommended to prevent these serious complications [7-10].

Aim of our study was to evaluate our rate of recurrent laryngeal dysfunction after pulmonary resections, and clinical outcomes of these patients following vocal cord medialization.

Materials and Methods

After the approval of Institutional Review Board (14.01.21/179) the clinical files of all non-small cell lung carcinoma (NSCLC) patients undergoing elective surgery via thoracotomy between January 2011 and January 2020 were examined retrospectively. Data of patients developing postoperative unilateral vocal cord paralysis (UVCP), confirmed by indirect laryngoscopic examination and have undergone vocal cord medialization procedure was evaluated. Examined variables were pulmonary complication incidence, timing and effectiveness of procedure, and hospital length of stay (LOS). Patients with prior tracheostomy or known pre-operative phonetic/vocal cord dysfunction are excluded from the study. Written informed consent is waived from all patients prior to surgery.

Statistical Analysis

All analyses were performed using the MedCalc version 12.7 (MedCalc Software bvba, Belgium). Categorical data were examined using Pearson's Chi-square test.

For continuous variables, Student's-t and Kolmogorov-Smirnov tests were preferred for comparison. P values <0.05 were considered as statistically significant.

Results

Total of 2740 NSCLC patients underwent pulmonary resection via thoracotomy in 10 years. Eleven patients developed severe phonetic (hoarseness or dysphonia) alteration, postoperatively. After laryngoscopic examination (Figure 1), UVCP diagnosis was confirmed in 8 (0.3%) patients and the vocal cords of other 3 were found edematous.

Operations performed on these 8 patients were left pneumonectomy in 5 (62.5%) and left upper lobectomy in 3 (37.5%). Gender distribution of the patients was 6 (75%) males and 2 (25%) females, with an age average of 62.4 ± 22.7 . Final pathology revealed squamous cell carcinoma in 7 and adenocarcinoma in 1 patient.

One (12.5%) patient developed atelectasis necessitating bronchoscopy on postoperative day (POD) 2, and 2 (25%) patients developed pneumonia on POD 3 and 4, respectively. Overall PPC incidence was 37.5% in our UVCP cohort.

Calcium hydroxyapatite injection (Radiesse®; Bioform Medical Inc., USA) for 6 patients under general anesthesia, and polytetrafluoroethylene (PTFE) graft implantation under local anesthesia for 2 patients was performed. Choice of medialization technique was made upon the institution's actual amenities. Mean duration between pulmonary resection and medialization procedure was 5.3 ± 3.8 days in patients developing PPC and 3.6 ± 3.1 days in patients with no PPC ($p = 0.011$). No complication about medialization procedure or 30-day mortality was seen. All patients were discharged, uneventfully, with an average hospital LOS of 8.1 ± 4.9 days. Patient demographics and clinical data are summarized in table 1.

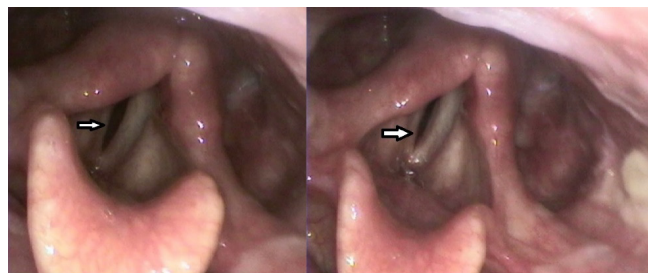


Figure 1. Paralytic left vocal cord on laryngoscopic examination.

Table 1. Patient demographics.

Patient #	Age	Gender	Resection	Pathology	Medialization technique and interval (days)	PPC	Hospital LOS (days)
1	72	Male	LUL	SCC	PTFE / 3	-	6
2	64	Male	LP	SCC	CaHA / 3	-	7
3	51	Male	LP	SCC	CaHA / 4	-	6
4	55	Female	LP	SCC	PTFE / 4	-	7
5	66	Male	LUL	SCC	CaHA / 6	Pneumonia	13
6	64	Female	LP	AC	CaHA / 5	Atelectasis	9
7	69	Male	LUL	SCC	CaHA / 4	-	7
8	58	Male	LP	SCC	CaHA / 5	Pneumonia	10

Abbrev.: AC: Adenocarcinoma, CaHA: Calcium hydroxyapatite, LOS: Length of stay, LUL: Left upper lobectomy, LP: Left pneumonectomy, PTFE: Polytetrafluoroethylene, PPC: Postoperative pulmonary complication, SCC: Squamous cell carcinoma

Only one (12.5%) patient necessitated a second injection of calcium hydroxyapatite, on postoperative month 5. Throughout our mean follow-up duration of 14.8 ± 16.2 months, the patients had stable vocal cord functions and no phonetic complaints.

Discussion

Intra-thoracic malignancies, especially bronchogenic and esophageal carcinomas are associated with a range of local complications caused by the primary tumor, lymph nodes, or the surgical resection itself [2,5,7]. One of the neighboring structures which is under risk of direct tumor invasion is the recurrent laryngeal branch of the Vagus nerve, particularly in malignancies of the left hemithorax, attributable to its anatomical location. During surgery, mechanical or thermal injury, or sacrifice of an invaded RLN may cause UVCP and alterations in laryngeal functions since it is the major motor nerve of the larynx [8-10].

UVCP is a known sequel of intra-thoracic malignancies in literature. In a group of eight pooled series of UVCPs consisting of 1019 patients, 36% of cases were caused by neoplasms, with more than half of these resulting from lung cancer [8]. Reversible vocal cord dysfunction incidence is reported as high as 31% in patients undergoing left-sided pulmonary resection for malignancy, but the true incidence is difficult to determine as few centers perform routine laryngeal examination on patients with postoperative dysphonia [9]. Consequently, patients with RLN damage suffer from postoperative phonetic (hoarseness and dyspnea during speaking) problems as well as swallowing, or respiratory (difficulty in coughing, atelectasis,

pneumonia or acute respiratory distress syndrome) complications [6,11]. Eleven patients in our cohort had new onset postoperative phonetic problems, of which 8 (0.3%) of them were diagnosed with UVCP, confirmed with indirect laryngoscopic examination, all have undergone pulmonary resection on the left side.

The thoracic surgery patients, in general, are at exceptionally elevated risk for postoperative respiratory complications, which are the major source of morbidity, due to the high prevalence of COPD, invasiveness of thoracotomy incision, postoperative pain, immobilization, insufficient respiratory exercise, impaired glottis function due to endotracheal intubation, and the nature of the primary surgical procedure directly diminishing pulmonary function and toilet. It's been shown that patients with UVCP injury demonstrate a 5-fold risk of contracting pneumonia (and a 20% death rate for those who contract pneumonia), a 5-fold risk of re-intubation or tracheostomy. Also, they have 40-60% longer hospital stays than patients without RLN injury because, vocal cord closure is considered to be the most important protective mechanism against aspiration [8-11]. In a study of patients with postoperative UVCP, rates of aspiration with swallowing and aspiration pneumonia were found to be 53% and 45%, respectively; and tracheostomy was necessary in 25% for the management of persistent aspiration [12]. In our series, postoperative pulmonary complication rate was 37.5% where one patient developed atelectasis necessitating bronchoscopy for bronchial toilet, and two patients had pneumonia.

Vocal cord medialization is the recommended

treatment of choice for unilateral vocal cord paralysis, in order to avoid repetitive aspiration which can lead to pulmonary complications with potentially life-threatening consequences [7,8,10]. Numerous different techniques have been described in current literature to close the glottic opening or to reposition the lateralized vocal cord. They may be performed under general or local anesthesia, using synthetic materials or allografts [11,13-16]. Intracordal injection of materials or buttressing the lateral side of the cord are the main techniques for medialization; all being used and recommended in achieving adequate closure of the glottic gap [8-11,13-15]. In our cohort, 6 patients underwent calcium hydroxyapatite injection under general anesthesia and 2 patients underwent PTFE graft implantation under local anesthesia, at the referred otolaryngology clinic, according to the surgeon's decision. No procedure-related complication occurred.

Timing of medialization is another essential issue. Early detection and surgical repositioning of vocal cords has been shown to significantly decrease the pneumonia rate, the requirement for postoperative bronchoscopy and the hospitalization time for patients developing UVCP after thoracic surgery, by preventing a possible aspiration [10,17-19]. Lack of a stable otolaryngology team at our institution and the need for scheduling external-consultation at another hospital may have caused some delay for diagnosis in our patients. Our median interval between the thoracotomy and the medialization was 4.3 days. There was a significant difference between the timing of medialization in patients with or without PPC in our group (5.3 days vs 3.6 days; $p = 0.011$).

As a conclusion, rapid intervention (≤ 4 days) for iatrogenic unilateral vocal cord paralysis by injection laryngoplasty appears to be a practical, safe, and effective method for minimizing pulmonary complications after pulmonary resection, possibly by reducing aspiration, improving cough and bronchial toilet; regardless of the surgical technique.

Declaration of conflicting interests

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Ethics approval

The study was approved by the institutional review board of Health Sciences University, Sıyirci Paşa Chest Diseases and Thoracic Surgery Training and Research Hospital (No: 14.01.21/179)

Authors' contributions

MED; Co-wrote the paper, conceived and designed analysis, collected data, ÇT; contributed data, VB; Conceived and designed the analysis. SE; Conceived and designed the analysis, co-wrote the paper

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Original Article

Minimally invasive pectus excavatum surgery: Nuss procedure

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ABSTRACT

Background: We reviewed pre-, intra-, and post-operative clinical data, including morbidity and mortality rates, for patients who underwent Nuss surgery to repair pectus excavatum.

Materials and Methods: Nuss procedure was performed in 140 patients at our clinic between 2012 and 2019 (males: 108; females: 32 females; mean age: 15.7 ± 7.8 years; range: 3-40 years). The Haller index was mild (2.5-3.2) in 72, moderate (3.2-3.5) in 40, and severe (3.6-6.0) in 28 patients.

Results: None of the patients died. Morbidity or bad blood loss was not observed. The mean duration of surgery was 59.5 ± 21 min (range: 30-120 min). The mean duration of postoperative hospitalization was 4.6 ± 2.9 days (range: 3-19 days). The procedure was performed using one ($n = 107$; 76.4%), two ($n = 31$; 22.2%), or three ($n = 2$; 1.4%) bars. There were five cases of early pneumothorax, four pleural effusions, five bar displacements, two wound infections, one hematoma, seven bar exposures due to allergies, and three costal fractures.

Conclusions: The Nuss operation is an effective, minimally-invasive treatment for pectus excavatum. It has the advantages of a short duration of surgery and low morbidity and mortality rates. Therefore, the Nuss operation may be considered the treatment of choice in cases of pectus excavatum.

Keywords: pectus excavatum, surgery, minimally invasive

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Introduction

Pectus excavatum (PE) is the most common congenital chest wall disorder, with an overall incidence of 0.1%. The male to female ratio for PE is 4:1 [1]. The traditional repair technique is open sternoplasty [2]. PE progresses during puberty [3]. In 1998, Donald Nuss first used minimally invasive surgery to repair 45 cases of PE [1]. In this procedure, a metal bar is inserted into the retrosternal area using a thoracoscope and later removed [2]. Connective tissue diseases, including Marfan and Ehler-Danlos syndrome, accompany PE in 6% of patients, while scoliosis is seen in 40-65% of cases. Patients may show diminished exercise capacity, fatigue, dyspnea on deep inspiration, and recurrent pulmonary infections. Surgery is important because the negative aesthetics reduce self-confidence, which can cause social problems [4]. Preoperatively, the transverse thorax diameter is divided by the sternovertebral distance on thoracic tomography to obtain the Haller index (HI). An HI > 3.25 indicates the need for surgery. Kelly considered the following as indications for surgery: HI > 3.25, major displacement of the heart and/or cardiac compression, pulmonary compression, restrictive ventilation, mitral valve prolapse, bundle branch block or another cardiac pathology related to cardiac compression, or a history of ineffective correction [5]. However, most patients are operated upon due to aesthetic concerns.

We analyzed the data of patients who underwent the Nuss procedure in our clinic between 2012 and 2019.

Materials and Methods

Written informed consent was obtained from all adult patients and the parents of young patients for the study. We analyzed a retrospective cohort study. Our institutional ethics committee approved the study (Approval no: 2020/01). We enrolled 140 patients (108 males and 32 females with a mean age of 15.7 ± 7.8 years (range: 3-40 years); one patient was 2.5-5 years of age, 28 were aged 6-12 years, 82 were aged 13-18 years, 19 were aged 19-25 years, and 11 were aged 25-40 years. One patient had previously undergone Abramson surgery; two others had undergone wedge resection to treat pulmonary sequestration and bronchiectasis, and one had undergone bullectomy. We obtained the patients' medical histories and performed electrocardiography, echog-

raphy, direct graphing, thoracic computed tomography, and pulmonary function tests prior to surgery. Preoperative clinical symptoms, including dyspnea on effort, chest pain, fatigue, and palpitations, were evident in 79 patients (56.4%); 108 (77.1%) patients had pulmonary function disorders, 31 (22.1%) had mildly restrictive ventilation dysfunction, 51 (36.4%) had moderately restrictive ventilation dysfunction, 6 (18.5%) had severely restrictive ventilation dysfunction, 27 (19.2%) had echocardiographic abnormalities, 10 (7.1%) had mitral valve prolapse, 4 (2.8%) had mild mitral insufficiency, 2 (1.4%) had patent foramen ovale (Table 1).

Patients were classified according to their condition using the HI, as mild (HI = 2.5-3.2; n = 72, 51.4%), moderate (HI = 3.2-3.5; n = 51, 36.4%), or severe (HI = 3.6-6.0; n = 28, 20%). Comorbidities were present in 18 (12.8%) patients, including eight with pterygoid chests, 12 with scoliosis, one with pulmonary sequestration, one with asthma, one with coarctation of aorta, two with Marfan syndrome, and one with growth retardation (Table 2).

Table 1. Demographic and clinical data of the pectus excavatum patients before surgery.

	n (%)
Gender	140
Male	108
Female	32
Age	15.7 (3-40 years)
Recurrence	3
With clinical symptoms	79
Dyspnea on effort	27 (19.2%)
Chest pain	40 (28.5%)
Fatigue	5 (3.5%)
Palpitation	4 (2.8%)
Other	3 (2.1%)
Pulmonary function disorder	108
Mild restrictive dysfunction	31 (22.1%)
Moderate restrictive dysfunction	51 (36.4%)
Severe restrictive dysfunction	26 (18.5%)
Echocardiographic abnormality	27
Mitral valve prolapse	10 (7.1%)
Mild mitral insufficiency	4 (2.8%)
Patent foramen ovale	2 (1.4%)
Pressure on heart	6 (4.2%)
Other	5 (3.5%)

Table 2. Clinical data of the pectus excavatum patients before surgery.

	n (%)
Severity of pectus excavatum	140
Mild (HI < 3.2)	72 (51.4%)
Moderate (HI:3.2-3.5)	40 (28.5%)
Severe (HI: 3.6-6.0)	28 (20%)
Comorbidity	18
Scoliosis	12 (8.5%)
Pulmonary sequestration	1 (0.7%)
Asthma	1 (0.7%)
Coarctation of aorta	1 (0.7%)
Marfan syndrome	2 (1.4%)
Growth retardation	1 (0.7%)

Surgical indications

The criteria for surgery were as follows: pectus index (PI) > 3.25, requirement for cosmetic defect correction, major displacement of the heart and/or cardiac compression, pulmonary compression, a restrictive ventilation pattern, mitral valve prolapse, a bundle branch block or other cardiac pathology related to cardiac compression, and a history of unsuccessful operations.

Surgical procedure

All patients were placed under general anesthesia via single-lumen endotracheal intubation. The preoperatively measured Nuss steel bars fitted the chest wall well. Bars were made of surgical steel and were 13 mm wide and 2 mm thick (Hipokrat®, Izmir, Turkey). Three-centimeter-long transverse incisions were created on both sides of the thorax, running from the beginning to the anterior axillary line to where it met the deepest point of the depression. After dissection of subcutaneous tissue and muscle, the tissue along the rib surface up to the highest hinge point was manually dissected. A camera port was inserted in the midaxillary line of the seventh intercostal area. Under videothoracoscopic guidance and carbon dioxide insufflation, an introducer was placed from the highest hinge point to the right thoracic cavity, and then passed from the right mediastinum to the left chest wall and then to the highest contralateral hinge point. The introducer was then grasped and pressed against the sternum to shape the chest contour. Nylon tape was adhered to the end of the introducer. After the introducer had been extracted from the left thoracic wall, the Nuss bar was bound to the end of the tape and the bar was pulled back to the contralateral side of the chest

wall using a rope. The bar was then inverted to lift the sternum. Two fixation stabilizers were placed on the tips of the bar. A feeding tube was placed in the right chest wall and the lung was inflated to remove the artificial pneumothorax. In patients with extremely severe PE, a small incision was created under the xiphoid to allow the surgeon's fingers to guide the introducer. Alternatively, a crane was used to elevate the sternum.

Postoperative treatment

Epidural analgesia was used to relieve pain, which is the most serious postoperative problem. If an epidural catheter could not be inserted, intravenous analgesia was used. Intravenous antibiotics were given early on the first postoperative day. Patients were rapidly mobilized and discharged after 3–7 days in hospital. They were instructed not to lift heavy objects or perform vigorous exercise for 2 months. They were followed-up regularly and the bars were extracted after 3 years.

Results

In all 140 patients, surgery was successful, and morbidity or blood loss was not observed (Figures 1,2). On computed tomography scans, the mean HI was 4.5 ± 1.4 (range: 2.8-6.2). The mean duration of surgery was 39.5 ± 21 min (range: 30-120 min). The postoperative mean duration of hospitalization was 4.6 ± 2.9 days (range: 3-19 days). No pericardial perforation was noted. Three patients underwent Abramson surgery using the sandwich technique, while one underwent thoracoscopic bullectomy and another pulmonary wedge resection. A total of 107(76.4%) patients received one bar, while 31(22.2%) received two bars, and 2(1.4%) received three bars. No patient received more than three bars. Two or three bars were placed for larger affected areas on the 93 chest wall (Figures 3,4).

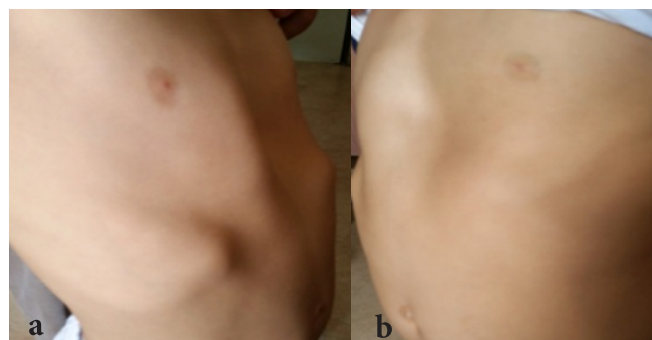


Figure 1. Preoperative view of a pectus excavatum patient (a), postoperative view of a pectus excavatum patient (b).



Figure 2. Preoperative view of a pectus excavatum patient (a), postoperative view of a pectus excavatum patient (b).

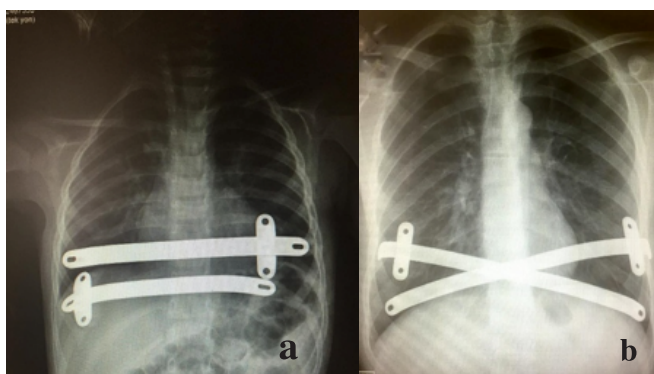


Figure 3. Postoperative view of a pectus excavatum patient with two parallel bars (a), postoperative view of a pectus excavatum patient with two transverse bars (b).

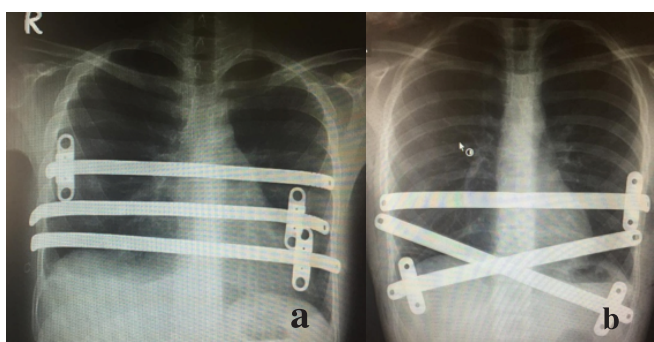


Figure 4. Postoperative view of a pectus excavatum patient with three parallel bars (a), postoperative view of a pectus excavatum patient with one parallel and two transverse bars (b).

In terms of postoperative complications, early pneumothorax was seen in five patients, of whom two underwent tube thoracostomy. Pleural effusion was evident in four patients, but treatment was not necessary. Postoperative bar displacement was evident in five patients, so the bars were removed 20-23 months early. The bars were also removed early in patients who complained of painful scoliosis. Wound infections were noted in two

patients, both of whom responded to antibiotics. A subcutaneous hematoma was observed in one patient, and debridement was performed. Bar exposure caused by allergy was noted in seven patients, and costal fractures were seen in three others (Table 3). Allergy developed to Nuss steel 101 bars with no. 14 and 15. Tip of the bars got out of the skin in the patients with bar allergy. Surgical wound infection was seen in 2 patients. Bars were removed in 3 patients due to allergy, 4 patients with bar allergy were treated with surgical wound revision and antibiotics. Vacuum-assisted wound closure was applied in one patient. Patients were followed up at 1, 3, 6, and 12 months. The mean follow-up time was 43 ± 26.3 months (range: 1-93 months). There were no cases of obvious recurrence.

Table 3. Clinical data of surgery and complications.

	n (%)
Duration of surgery	Median: 39.5 (30-120 min)
Blood loss during surgery	0
Duration of hospitalization	Median: 4.6 days (3-19 days)
Pericardial perforation	0
Reoperation due bar displacement	7 (5%)
Morbidity	0
Number of steel bars	140
1 bar	109 (77.8%)
2 bar	31 (22.2%)
3 bar	2 (1.4%)
Complications	15 (10.7%)
Bar exposure due to bar allergy	7 (5%)
Pneumothorax	5 (3.5%)
Pleural effusion	4 (2.8%)
Bar displacement	5 (3.5%)
Wound infection	2 (1.4%)
Hematoma	1 (0.7%)
Costal fracture	3 (2.1%)

Discussion

PE is a progressive genetic deformity of the cartilaginous regions of the ribs and sternum, characterized by some degree of depression of the anterior chest wall [2-5]. The male:female ratio of PE was reported at 4:1[3]; in our study, it was 3.3:1. Although females are less frequently affected, their deformities tend to be more severe. Marfan syndrome was evident in 15% of patients [3]. As the risk of PE recurrence is much higher in such patients, Marfan syndrome status requires careful evaluation [4]. Two of our patients had Marfan syndrome, but no postoperative recurrence was noted. Among all patients, 26% had scoliosis and 12(8.4%) had scoliosis-

like deformities. The patients were generally asymptomatic, although some complained of chest pain, fatigue, dyspnea, palpitations, and movement limitations. Seventy-nine patients (56.4%) were symptomatic. Congenital cardiac defects, especially mitral valve prolapse, may accompany PE [2]. Twenty-seven patients showed preoperative echocardiographic abnormalities and ten (7.1%) had mitral valve prolapse.

Ravitch sternoplasty is the traditional method for surgical correction of PE [6]. In 1949, Ravitch was the first to excise all deformed cartilage and perichondrium, decompose the xyphoid and intercostal packages from the sternum, and perform right sternal alignment accompanied by transverse sternal osteotomy. Kirschner wires were used for fixation; silk sutures were later preferred [2]. However, these techniques caused pain, and were associated with long operative and hospitalization times, bleeding, many postoperative complications, and poor cosmetic outcomes.

In 1987, Donald Nuss, a pediatric surgeon, described a minimally invasive method that did not involve osteotomy. The ideal patient age for the procedure was considered to be 6-12 years, where PE involves the costal cartilages [1]. Also, the psychological impact of PE is less in children. Our patients were aged between 3-40 years (mean age, 15.7 ± 7.8 years). Only one patient was aged under 6 years; that 3-year-old patient was operated upon because a severe deformity depressed the heart. Although the technique is more difficult to apply in older patients with stronger chest walls, selected patients may benefit.

The Nuss technique is associated with good cosmetic results, short surgery, no blood loss, and fast recovery. The mean duration of surgery in our study was 39.5 ± 21 min, which is markedly shorter than that for the traditional technique. Cardiac injury is the most important potential complication. We observed no postoperative bleeding or pericardial perforation in our patients, none of whom died. The thoracoscopic view was excellent in all patients and we were able to protect the heart. Carbon dioxide insufflation allowed us to proceed beyond the adhesive bands that lie behind the sternum; this further reduced the risk of cardiac damage. For severe PE cases, a small incision created below the xyphoid or sternum allows the surgeon to elevate, and deal with adhesions behind, the sternum.

The Nuss technique is valuable for patients who develop recurrence after open sternoplasty. However, these patients require careful evaluation because retrosternal adhesions may be present [2]. None of our cases had a history of open surgery.

The most common complications of Nuss surgery are pleural effusions, bar displacement, wound infection, pneumothorax, scoliosis, and bar allergy. Zhang et al reported a postoperative complication rate of 9.7% in 639 patients [1]. The rate in our study was similar, at 10.7%. Nuss reported that the most common complication among their 1,015 cases was pneumothorax (60.4%) that spontaneously resolved [7]. The most common complication among our patients was bar exposure caused by bar allergy (5%). Pneumothorax (3.5% of patients) was the second most common complication; however, this was diagnosed by chest X-ray only. Nuss reported that 3.6% of pneumothorax cases required chest drainage, compared to a rate of 1.4% in our study.

In a Canadian study, postoperative pleural effusion developed in 4.65% of patients; in our study, the rate was 2.8%, while the wound infection rate was 1.4%. Postoperative pneumothorax developed in 3% of patients in their study; in our study, 151 the rate was %3.5. Bar displacement was seen in %3.7 in their study; in our study, the rate was 152 %3.5. In patients with relatively large affected areas, we placed two or three bars (fixed with 153 stabilizers) on the right and left tips; the stabilizers usually prevent bar dislocation. In patients with relatively large affected areas, we placed two or three bars (fixed with stabilizers) on the right and left tips; the stabilizers usually prevent bar dislocation. Postoperative bar displacement was evident in five patients. We did not routinely place chest tubes after surgery; instead, we inflated the lungs and extracted all air from feeding tubes that were temporarily placed in the right hemithorax during surgery.

All steel bars were removed after 3 years. Two patients could not tolerate the bars due to pain. Seven patients were allergic to the bar material and thus exhibited delayed wound-healing. Postoperative bar displacement was evident in five patients, so the bars were removed 20-23 months early. The bars were also removed early in patients who complained of painful scoliosis. Overall, the results were satisfactory.

The Nuss technique requires only small incisions; moreover, the operation is quick, there is no bleeding, and the skeleton can develop in the absence of cartilage. Also, the complication rate is low and the outcome is usually good.

In conclusion, the Nuss technique is an advanced surgical method, although the postoperative outcomes in cases of bar removal require further study, as do the effects of the procedure on cardiopulmonary function [8-10]. A limitation of our study was its single-center, retrospective design. The surgical technique used should be determined on an individual patient basis, i.e., according to general condition and age; more research is still required.

Declaration of conflicting interests

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Ethics Approval

The study was approved by the institutional ethical board of Health Sciences University, Istanbul Mehmet Akif Ersoy Thoracic and Cardiovascular Surgery Training and Research Hospital (No:01/2020).

Authors' contributions

TA, MA; co-wrote the paper, collected the data, performed the analysis, contributed data/analysis tools, conceived and designed the analysis.

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Case Report

Late term broncho-esophageal fistula and endoscopic treatment: a case report

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ABSTRACT

Fistulas between the respiratory and digestive tract (trachea-esophageal, broncho-esophageal) can be congenital or acquired. Acquired fistulas due to malignancy can occur by direct invasion of the malignant tissue; or it may be a result of surgical treatment, chemotherapy or radiotherapy. Although such fistulas occur during or shortly after treatment, it is unlikely that they will emerge long years after treatment. We report a case of late term broncho-esophageal fistula and an endoscopic treatment method in a patient who underwent left pneumonectomy for bronchial carcinoma. Treatment of the patient was accomplished with bronchoscopic placement of polyglycolic acid patch with surgical tissue glue into the fistula tract. Fistulas between trachea-bronchus and digestive tract should be kept in mind, after surgical interventions of neighborhood structures. Bronchoscopic interventions may be a treatment alternative in appropriate patients.

Keywords: broncho-esophageal fistula, bronchoscopy, bronchial carcinoma, polyglycolic acid patch, surgical tissue glue

Introduction

Broncho-esophageal fistula in adults is a challenging problem for surgeons. Previously the initial treatment choice of this complication was surgery [1]. As a result of the development of endoscopic and bronchoscopic interventional methods and materials, such interventions can be used as the first choice in the treatment of broncho-esophageal fistula [2].

Case Report

A 68-year-old male patient underwent left pneumonectomy in 2004 due to poorly differentiated squamous cell bronchial carcinoma. During the procedure, partial esophagus wall resection was performed due to invasion of tumor into the outer muscle tissue of the esophagus wall which was approximately 4 cm long. Pleural patch was used to support to esophageal wall and secure the mediastinum. The patient with pathological stage IIIA received chemotherapy and radiotherapy after surgery. Follow-up revealed no signs of recurrence for 9 years. In 2013, the patient was diagnosed with pneumonia after a cough complaint. Since the symptoms of the patient did not regress with antibiotics, thoracic tomography was performed, which revealed clues of broncho-esophageal fistula. In his esophagography, enlarged esophagus part due to the partial wall resection and the contrast leakage from esophagus to the left bronchial stump was seen (Figure 1). During esophagoscopy procedure, fistula orifice was observed in the 25th centimeter of the esophagus with the polypropylene suture part which was partially closing the orifice (Figure 2). There was no connection between the fistula and the pneumonectomy space. After obtaining informed consent from the patient, endoscopic fistula closure was planned and rigid bronchoscopy was performed under general anesthesia. During bronchoscopy, one end of the polypropylene suture was observed in the bronchial system. (Picture 3). After removing the suture material from the fistula tract, about 1 cm of polyglycolic acid patch (NEOVEIL- Gunze Limited, Kyoto/Japan) was placed tightly on the fistula tract and surgical tissue glue (TISSEEL- Johnson & Johnson corp. /New Jersey/ USA) was applied on it. The procedure was ended after waiting for the drying time of the glue. In the follow-up, the patient had no symptoms and control bronchoscopy and esophagography (Figure 1) was performed 3 weeks later and there was no evidence of fistula. During the annual controls of the patient, there was no fistula signs and symptoms until the present day.

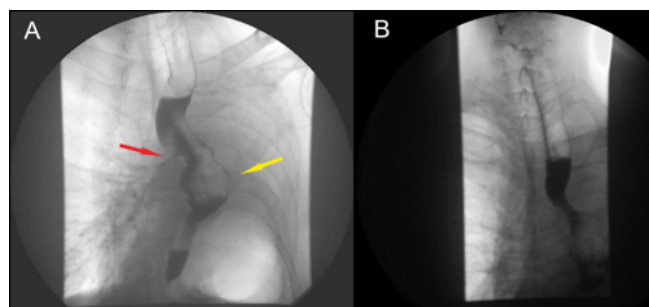


Figure 1. Esophagography before the closure of the broncho-esophageal fistula (Yellow arrow: dilated esophagus, Red arrow: the passage of the contrast through the fistula) (A), esophagography after the closure of the fistula without contrast leakage (B).

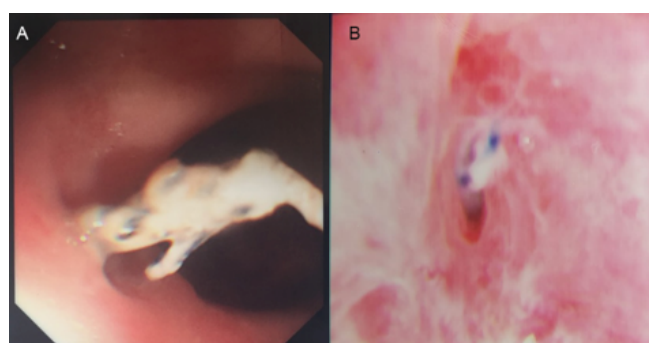


Figure 2. Fistula mouth and protruding polypropylene sutures in esophageal lumen (A), Bronchoscopic view of the fistula mouth and polypropylene fragment in the left main bronchial stump (B).

Written informed consent was obtained from the patient for publication of his data.

Discussion

Tracheo-broncho-esophageal fistulas can lead to severe lung complications and should be treated immediately after diagnosis. Fistulas can be seen immediately after surgery, or it may take years to appear. They are mostly seen in left side probably due to the relation with the esophagus and the left main bronchial stump after left pneumonectomy as in our patient [1]. Esophageal wall involvement and the suture material used to close the stump are probably responsible for fistula development. Contrast-enhanced esophagography is essential in diagnosis [2]. Treatment options vary from endoscopic procedures to surgery [2,3]. The goal of treatment is to prevent contamination of the bronchial system and to protect the patient from recurrent pneumonia and associated septic complications. The general condition of the patient and the localization of the fistula should be considered while planning the treatment. Considering that the majority of cancer patients have a high probability of surgical procedure complications, endoscopic

interventions should be the primary treatment option. Moreover, even if endoscopic interventions fail, they do not interrupt the open surgical interventions for the treatment [4]. Endoscopic interventions can be performed via the airway or esophagus [3-5]. Endoscopic stent applications with esophagoscopy or bronchoscopy or endoscopic clipping via esophageal way are some of the treatment options. Additionally tissue adhesive can be applied directly to the fistula area [5]. Usually esophageal route is preferred for endoscopic clip use and stent applications. However, in patients with esophageal dilatation, these methods have a low chance of success and may be difficult to perform. In recent years, some successful results have been published on closure of esophageal fistulas using polyglycolic acid patch and tissue glue [6,7]. However these applications were made via esophagoscopy, bronchial interventions can be a treatment choice especially in patients with dilated esophagus. And we think that polyglycolic acid patch application into the fistula tract with surgical tissue glue had an additive effect on tissue adhesion since there is no recurrence in our patient for 5 years.

In conclusion, the possibility of fistula in operated bronchial cancer patients with chronic cough complaints should be kept in mind even if they have passed through for many years. Endoscopic procedures should be preferred as initial therapy because they involve less risk compared to open surgical methods. Fistula closure with polyglycolic acid patch and surgical tissue glue via bronchoscopic way may be a treatment alternative.

Declaration of conflicting interests

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Authors' contributions

MCS, ED; Collected the data, performed the analysis, co-wrote the paper.

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Case Report

Pulmonary large cell neuroendocrine carcinoma: a diagnostic challenge

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ABSTRACT

Pulmonary large cell neuroendocrine carcinoma (LCNEC) is known by its highly aggressive behaviour. General presentation is early spread to both regional lymph nodes and distant sites. Here we present a totally asymptomatic patient with a huge fluid-filled cystic lesion detected incidentally in a chest radiograph.

Keywords: large cell neuroendocrine carcinoma, pulmonary, hydatid cyst

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Introduction

Pulmonary large cell neuroendocrine carcinoma (LCNEC) is a rare entity and accounts for 3% of all lung cancers [1]. In the latest 4th lung tumor classification of the World Health Organization (WHO, 2015), large cell neuroendocrine carcinomas were grouped with small cell carcinomas, and typical-atypical carcinoid tumors under a single heading and classified as the “neuroendocrine carcinoma group” of the lung [2]. Due to its aggressive behaviour, it is categorized as a high grade tumor. Most of the LCNECs present as an expanding mass located peripherally [1]. In this study, we are going to present a totally asymptomatic patient with a large fluid-filled pulmonary cystic lesion with a diagnosis of pulmonary LCNEC after resection.

Case Report

A 67-year-old male patient applied to the hospital with a huge lesion detected incidentally on a chest x-ray (Figure 1a). By profession, he was a butcher at a slaughterhouse. He was totally asymptomatic and routine blood examination was normal. The chest CT scan showed a homogeneously low-attenuation mass replacing the left lower lobe totally without any mediastinal or pleural involvement (Figure 1b).

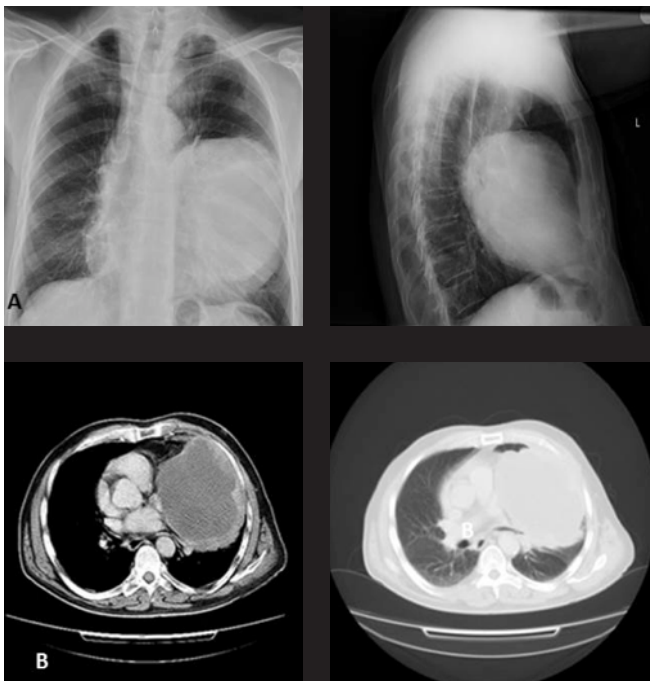


Figure 1. Chest x-ray at admission (A), thorax CT shows a homogeneously low attenuation lesion without enhancement (B).

Since the CT showed no hilar or mediastinal lymph node swelling and invasion, malignancy was not con-

sidered. Pulmonary echinococcosis was suspected, but no diagnostic intervention was conducted in order to avoid breaking the cyst. Immunodiagnostic tests were also positive and supported the clinical diagnosis of cystic echinococcosis, but a definitive diagnosis could not be made before surgery. Therefore, a muscle-sparing right thoracotomy was performed. At surgery, completely hemorrhagic and necrotic fragile tumoral tissue filling the entire lower lobe was observed (Figure 2). Intraoperative frozen section analysis was consistent with malignancy. Due to extensive hemorrhage from the tumor, lower lobectomy and mediastinal lymph node dissection were performed rapidly and bleeding was controlled. The post-operative course was uneventful.

Features of a neuroendocrine morphology such as peripheral palisading and rosette-like structures were present. The tumor cells were large (greater than the diameter of three lymphocytes) with moderate to abundant cytoplasm. The cells had nuclei with vesicular chromatin and the prominent nucleoli were remarkable. All of these features were useful in discriminating large cell neuroendocrine carcinoma from small cell carcinoma. More than 10% of the tumor cells were stained with CD56 and were negative with TTF-1 and p40. As a result of these staining patterns, adenocarcinoma and squamous cell carcinoma were not considered. The tumor was diagnosed as large cell neuroendocrine carcinoma with metastatic mediastinal lymph node. (Figure 3). After the completion of surgical treatment, PET/CT was performed and results were found to be consistent with metastasis in both the liver and adrenal gland. The patient was referred to the oncology department.

Written informed consent was obtained from the patient for publication of his data.



Figure 2. Macroscopically tumor appears to be composed of hemorrhage and necrosis.

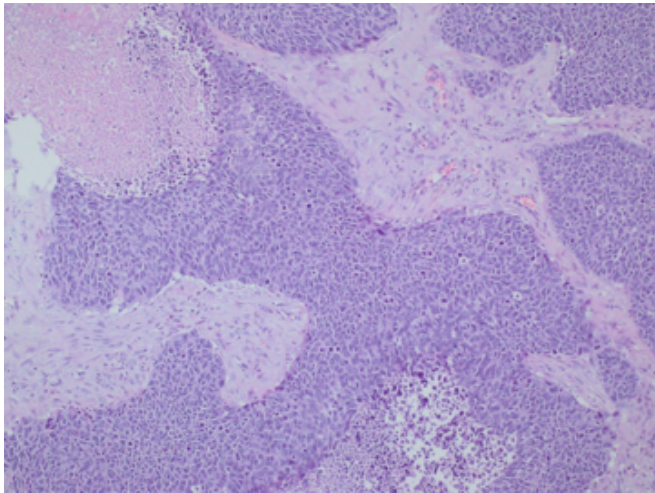


Figure 3. Large cell neuroendocrine carcinoma with peripheral palisading, rosette-like formation and large area of necrosis (H&E, X100).

Discussion

LCNEC generally presents with a peripheral pulmonary mass and patients with LCNEC are less likely to present with symptoms [1]. This is because the majority of reported LCNEC cases were under 5 cm in diameter. İncekara et al reported 25 LCNEC cases in 10 years and the average size of the tumors was about 3 cm. The tumor was large in only one patient (10x13x17 cm) [3]. The majority of LCNEC cases were also under 5 cm. in a series published by Gürsoy et al [4]. In another report by Lee et al, the mean tumor diameter was 3.8 ± 2.1 cm [5]. In our patient, the tumor replaced the entire lower lobe and the patient was fully asymptomatic. It occurred to us that pulmonary LCNEC can reach a very large size and remain in pleural reflection without local invasion. Since the aggressive clinical behaviour and poor prognosis of LCNEC are well documented, it was challenging.

It is reported that many types of lung cancer undergo necrosis and areas of haemorrhage [6]. In particular, squamous cell and large cell types are most frequently necrotic [7]. Rapid growth exceeding blood supply results with tumor necrosis and completely necrotic tumors may be indistinguishable from cysts [7]. Yoshida et al reported a 10 cm LCNEC that ruptured into the

pleural cavity with the extensive necrotic and hemorrhagic content causing hemorrhagic shock [6]. In our case, the fragile huge mass gave evidence of significant hemorrhage and necrosis at thoracotomy. Histopathological evaluation revealed the tumor mostly consisted of hemorrhage and extensive necrosis. Oshiro et al analyzed the CT findings and pathological specimens of the 38 pulmonary LCNEC cases and reported that in large-diameter large cell neuroendocrine carcinoma, the tumors contained extensive necrosis and the necrotic areas seemed to be confluent and more extensive [8]. Necrotic lung cancers sometimes also mimic an abscess or other cystic lesions when cavitated [7].

This case demonstrates a rare presentation and a diagnostic challenge. Firstly, the disease was advanced and the patient did not manifest any symptoms. Secondly, despite being so large, it didn't invade the pericardium, chest wall or diaphragm. Thirdly, the patient was a butcher by profession. ELISA was positive and presented with a large fluid-filled cystic mass in an endemic country. So malignancy was not suspected. Although a transthoracic biopsy would have broken the lesion, the possibility of a cystic echinococcosis could not be excluded pre-operatively.

In conclusion, although large cell neuroendocrine carcinoma is known as an aggressive neoplasm, it can reach giant sizes and can feature massive necrosis without any symptoms. This case presents an atypical presentation of an asymptomatic patient with pulmonary large cell neuroendocrine carcinoma.

Declaration of conflicting interests

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Authors' contributions

DY, MSY, AT; Collected the data, performed the analysis, co-wrote the paper.

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Case Report

A case of mediastinitis causing massive hemothorax

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ABSTRACT

Acute mediastinitis is an infection of the mediastinal tissue, which is a rare condition with high mortality and morbidity usually requiring intensive care follow-up. The most common reported causes of this condition are postoperative mediastinitis and descending necrotizing mediastinitis. The patients of the condition usually present with nonspecific symptoms such as fever, dysphagia, and chest pain. Laboratory findings observed are nonspecific, and also leukocytosis and high C reactive protein (CRP) values are encountered. Heterogeneity in the mediastinal tissues, air densities in the mediastinum and loculated fluid collections are radiologically observed. The treatment of the condition is antibiotherapy and surgery. Hemothorax is not reported as an expected complication in the follow-up of mediastinitis. In this case report, we present a patient with hemothorax hospitalized with the diagnosis of mediastinitis and empyema after an extraction of a tooth.

Keywords: abscess, empyema, hemothorax, mediastinitis

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Introduction

Acute mediastinitis is an infection of the mediastinal and surrounding connective tissues with an incidence rate of 0.3-5% causing high mortality as it affects such vital organs as the heart, large vessels, trachea and bronchi. Postoperative mediastinitis, descending necrotizing mediastinitis (DNM) and fibrous mediastinitis are the leading causes in etiology, of these, postoperative mediastinitis and DNM often have an acute and fulminant course, whereas fibrous mediastinitis follows a chronic and asymptomatic course. *Staphylococcus aureus* is the most common cause of postoperative mediastinitis with a rate of 60-80%. DNM is generally polymicrobial as it develops as a result of the spread of pharyngeal or odontogenic infection [1].

Hemothorax is the presence of blood in the pleural space caused mostly by trauma. Non-traumatic hemothorax causes are less common which can be listed as malignancy, anticoagulants, vascular ruptures (aortic dissection, arteriovenous malformation), endometriosis, and vascular adhesions in spontaneous pneumothorax [2].

In this study, a case report of a descending necrotizing mediastinitis developing after tooth extraction and resulting in hemothorax is presented.

Case Report

A 35-year-old male patient, who had a history of tooth extraction about two months ago, was admitted to the emergency department with the complaints of dyspnea, rash on the neck and chest skin accompanied by pain. The patient was present with no comorbidities except for diabetes mellitus. In his physical examination, respiratory sounds could not be heard in the basal lung zones. Blood pressure was 130/80 mmHg, pulse 92/min, body temperature was 38.3°C, and respiratory rate was 14/min. In the first laboratory results, leukocytosis and increased CRP were detected. Pleural effusion in bilateral lower zones was detected in the chest radiography of the patient and the mediastinum was wider than normal. Computed tomography (CT) revealed abscess formation in the retropharyngeal area, heterogeneous opacities in the mediastinum and bilateral pleural effusion. Bilateral chest tube was inserted, and bilateral empyema fluid was drained. Ceftriaxone and metronidazole were started as empirical treatment. No bacterial signal was detected in pleural fluid microbiological examination. In the follow-up of the patient, the pulse rate increased to 130/min, respiratory rate to 20/min,

while blood pressure decreased to 80/50 mmHg. Due to worsening clinical findings, he was taken to the intensive care unit with a pre-diagnosis of systemic inflammatory response syndrome (SIRS). Antibiotherapy was changed to piperacillin-tazobactam and teicoplanin and 1 g/kg/day intravenous immunoglobulin was added to the treatment. *Staphylococcus hominis* and *Staphylococcus haemolyticus* were detected in blood culture. No evidence of immunodeficiency was found in the patient's tests. It was thought that the retropharyngeal abscess progressed to mediastinitis due to poorly controlled diabetes. On the 10th day of the treatment of the patient whose SIRS findings regressed, postprandial hemoptysis and hypovolemic shock developed. Hemorrhagic drainage was started from the thoracic drain. There was hemothorax in the right hemithorax on thoracic tomography. When the images were examined in detail, an intercostal artery aneurysm (Figures 1a,b) showing active extravasation originating from the descending aorta and crossing the esophagus towards the right hemithorax was detected. Aneurysmatic intercostal artery was embolized (Figures 2a,b). The thoracic drain of the patient, whose infection regressed and hemorrhagic drainage was ended, was removed after embolization. The patient was discharged on the 32nd day of the treatment. Pleural decortication was planned when the pulmonary capacity was found to be decreased in the polyclinic control. Pleural decortication was applied to right hemithorax on the 30th day and to left hemithorax on the 120th day following the discharge. The patient was evaluated with chest radiography (Figure 2c) and respiratory function test on the 180th day. The forced expiratory volume (FEV1), which was 1620 mL in the preoperative pulmonary function test, increased to 2290 mL in the postoperative 6th month.

Written informed consent was obtained from the patient for publication of his data.

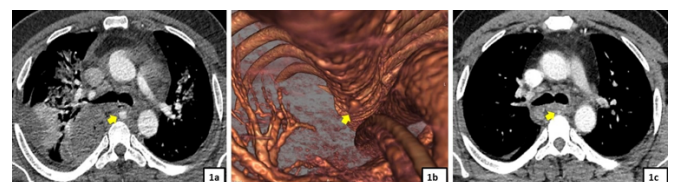


Figure 1a. Aneurysmatic dilatation in the intercostal artery at the carina level (arrow) (Horizontal plane tomography section) (a), 3-D view of aneurysmatic dilatation in the intercostal artery (arrow) (b), tomography section at the carina level and normal width intercostal artery view in the patient's admission tomography (arrow) (c).

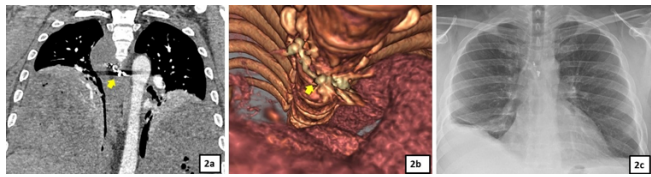


Figure 2. The coil in embolized intercostal aneurysmatic dilatation arising from the descending aorta (arrow) (Coronal plane tomography section) (a), 3-D view of coil in the embolized aneurysmatic dilatation (arrow) (b), chest radiography on the 180th day after discharge (c).

Discussion

Descending mediastinitis is a rare disease that can be progressive and life threatening. It usually develops after dental infections (36-47%). The other causes are pharyngeal infections and head and neck infections. Patients usually present with symptoms such as fever, skin rash and warmth and pain. In advanced cases, crepitation may develop in the skin of the affected area [3]. Our patient in this case presented with chest pain, rash on the neck and anterior chest wall skin in compliance with the literature. When the patient with poor oral hygiene was questioned, it was learned that he had a history of tooth extraction 2 months ago.

CT may be preferred for detailed imaging in patients with suspected mediastinitis learning the patient's history and conducting the physical examination. On CT scans, loculated mediastinal fluid, mediastinal air densities, decreased adipose tissue density, pleural-pericardial effusion, and mediastinal lymphadenopathies are significant findings in terms of mediastinitis [4]. When our case was evaluated with CT, heterogeneity in the mediastinal tissues, bilateral pleural effusion and retropharyngeal abscess were detected. For this reason, the patient who underwent bilateral tube thoracostomy was diagnosed with empyema secondary to descending mediastinum. He was taken to the intensive care unit due to worsening vital signs and development of SIRS.

Despite the drainage, use of antibiotics and advances in intensive care treatment, the mortality rate caused by oropharyngeal and dental deep neck infections is still high. Delay in diagnosis and treatment of deep neck infection may end up with descending necrotizing mediastinitis. According to the literature, the mortality rate associated with descending mediastinitis before the introduction of aggressive surgical debridement techniques was 40-50%. The reason for this high mortality

rate was associated with the fact that descending mediastinitis continues the infective process, causing empyema, pleural and pericardial effusion, pericarditis and blood vessel erosion. In severe mediastinitis, the mortality rate can rise up to 67%. There are many surgical approaches such as cervicotomy, thoracotomy, mediastinotomy or lavage through the thoracic drain [5]. We preferred to insert bilateral chest drain and daily pleural lavage because the purulent fluid was drained into both hemithorax in our case.

The patient who developed acute hemorrhagic drainage from the thoracic drain was evaluated with CT in terms of possible causes of hemorrhage. Interestingly, an aneurysmal dilatation was observed in intercostal artery originating from the aorta and extending towards the right hemithorax at the level of the tracheal bifurcation. When the previous tomography sections of the patient were examined, it was found that this aneurysm was not present on his admission imaging and that the intercostal arteries at the bifurcation level were normal. Intercostal artery aneurysm was urgently embolized. Hemorrhagic drainage from the thoracic drain ended in approximately 1 hour. The patient, who survived the acute empyema period, was applied pleural decortication in the chronic period because of empyema-related pleural thickening and decreased respiratory capacity.

In conclusion, this mediastinitis case which seems to be classical in terms of etiology, symptoms and laboratory results has drawn an interesting path due to the aneurysmatic dilatation of the intercostal artery. This case showed us that hemothorax should be kept in mind as an important cause of mortality in patients with mediastinitis. We are of the opinion that our report will contribute to the literature in terms of both mediastinitis resulting in postprandial hemothorax and the management of this emergency.

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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Authors' contributions

SA, ŞMKB, TİA, DB; Collected the data, performed the analysis, co-wrote the paper.

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Letter to the Editor

Skin reaction after pectus surgery

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Dear Editor,

I have read the article entitled “Minimally invasive pectus excavatum surgery: Nuss procedure” by Apaydin T and Akkuş M published in Current Thoracic Surgery (2021; 6(2): 69-74) with a great interest [1]. I want to congratulate the authors for this successful article, and make some contributions.

One of the most common complications in patients who have undergone the Nuss procedure is skin reactions defined as bar allergy and consequent bar exposures, as stated by the authors. The problem encountered on the skin often occurs in the surround of the bar ends and progresses as a dermatitis with non-itching erythema. If appropriate follow-up and treatment is not performed, it can progress to open wound formation and bar exposure. However, the point to be emphasized here is that this skin reaction is not entirely the result of metal bar allergy. Although bars containing metals such as nickel and chromium, which may cause allergic reactions, are used in surgery, it should be kept in mind that the contact of the metal bar with the skin and skin tension may also cause skin reaction. Although it has been

reported in the literature that metal allergy is 10% in the general population, the complication rate defined as bar allergy in pectus excavatum series has been reported to be between 0.5% and 6.4% [2]. While this rate was 5% in the authors’ study, it was 3.1% for pectus excavatum and 8% for pectus carinatum according to our experience [1, 3]. Compared to the normal population, metal allergy is less common in pectus patients. In the literature, it was found that nickel, molybdenum, manganese release did not change in patients with bar, chromium and iron release increased, and it was reported that there was no difference in metal release in patient groups using single bar and more than one bar [4]. It has also been shown that preoperative allergy testing on the skin is not an appropriate method to screen for the risk of post-operative reactions [5]. In fact, there is no consensus on whether this complication is due to allergy, metal release, or surgical technique. It would be more appropriate to use the terminology of “skin reaction” or “metal reaction” instead of “bar allergy” or “metal allergy” in the definition of that complication. The reasons for this reaction may be metal release, as well as direct contact of the bar ends with the dermis, pressure, and tension

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on the skin. Therefore, after the bar is implanted, the complication rate can be reduced by preparing a muscle flap to cover the bar ends and bending the bar ends to fit the thoracic arch.

Keywords: complication, pectus excavatum, skin reaction.

Declaration of conflicting interests

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CURRENT THORACIC SURGERY

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on the skin. Therefore, after the bar is implanted, the complication rate can be reduced by preparing a muscle flap to cover the bar ends and bending the bar ends to fit the thoracic arch.

Keywords: complication, pectus excavatum, skin reaction.

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