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Original Article / Özgün Makale

Thoracic surgery in newborns and infants

Yenidoğan ve bebeklerde göğüs cerrahisi

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ABSTRACT

Background: This study aims to evaluate the results of major thoracic surgical procedures performed in newborns (first 28 days of life) and infants (up to one year of life) in our clinic.

Methods: We retrospectively analyzed consecutive 27 newborns and infants (14 males, 13 females) who underwent thoracic surgical procedures between January 2000 and December 2014 in our clinic. Of the patients, 10 (37%) were newborns (mean age 98.4±103.1 days; range 2 to 340 days). We reviewed patients' age, gender, diagnosis, surgical procedure, postoperative hospitalization time, complications, and mortality rates. Patients who had chest tube for neonatal pneumothorax or bronchoscopy due to any reason were excluded from the study.

Results: Surgery indications were congenital cystic pulmonary malformations in 10 patients (37%) (six lobar emphysemas, four cystic adenomatoid malformations) mediastinal cysts in four patients (14.8%) (three gastroenteric cysts, one cystic teratoma), congenital diaphragmatic hernia in four patients (14.8%) (three Morgagni hernias, one Bochdalek hernia), diaphragmatic eventration in three patients (11.2%), mediastinal cavernous hemangioma in two patients (7.4%), Jeune syndrome in one (3.7%), cleft sternum in one, congenital chylothorax in one, and chest wall tumor (lipoblastoma) in one patient. Atelectasis was observed in two patients (7.4%) postoperatively. Hospital mortality was observed in one patient (3.7%) with Jeune syndrome. Mean postoperative hospitalization time was 6.9 days (range 1 to 30 days).

Conclusion: Although indications for thoracic surgery are rare in newborns and infants, this intervention may be lifesaving and performed successfully in majority of patients.

Keywords: Infant; newborn; thoracic surgery.

ÖZ

Amaç: Bu çalışmada kliniğimizde yenidoğan (yaşamın ilk 28 günü) ve bebeklerde (bir yaşına kadar olan dönem) uygulanan majör göğüs cerrahisi girişimlerinin sonuçları değerlendirildi.

Çalışma planı: Kliniğimizde Ocak 2000 - Aralık 2014 tarihleri arasında göğüs cerrahisi girişimleri uygulanan ardışık 27 yenidoğan ve bebek (14 erkek, 13 kız) retrospektif olarak incelendi. Hastaların 10'u (%37) yeni doğan (ort. yaş 98.4±103.1 gün; dağılım 2-340 gün) idi. Hastaların yaşı, cinsiyeti, tanısı, cerrahi işlemi, ameliyat sonrası yatış süresi, komplikasyonları ve mortalite oranları gözden geçirildi. Neonatal pnömotoraks nedeni ile tüp torakostomi uygulanan veya herhangi bir nedenden dolayı bronkoskopi uygulanan hastalar çalışmaya dahil edilmedi.

Bulgular: Cerrahi endikasyon 10 hastada (%37) doğuştan kistik pulmoner malformasyon (altı lober amfizem, dört kistik adenomatoid malformasyon), dört hastada (%14.8) mediastinal kist (üç gastroenterik kist, bir kistik teratom), dört hastada (%14.8) doğuştan diyafragma hernisi (üç Morgagni hernisi, bir Bochdalek hernisi), üç hastada (%11.2) diyafragma evantrasyonu, iki hastada (%7.4) mediastinal kavernöz hemanjiom ve birer hastada (%3.7) Jeune sendromu, kleft sternum, doğuştan şilotoraks ve göğüs duvarı tümörü (lipoblastom) idi. Ameliyat sonrası iki hastada (%7.4) atelektazi gözlendi. Jeune sendromlu bir hastada (%3.7) hastane mortalitesi gözlendi. Ortalama ameliyat sonrası yatış süresi 6.9 gün (dağılım 1-30 gün) idi.

Sonuç: Yenidoğan ve bebeklerde göğüs cerrahisi için endikasyonlar nadir olmasına rağmen, hastaların çoğunda bu girişim hayat kurtarıcı olup başarılı bir şekilde uygulanabilmektedir.

Anahtar sözcükler: Bebek; yenidoğan; göğüs cerrahisi.

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The majority of thoracic surgery patients are adults. Newborn (first 28 days of life) and infant (up to one year of life) patients are not treated frequently. Thoracic surgical procedures in newborns and infants are performed by surgeons from different specialties depending on the country. In United States and some European countries, pediatric surgeons perform those procedures. In some countries, thoracic surgeons and pediatric surgeons perform together. In Turkey, thoracic surgeons usually perform those procedures. There are several case reports and case series in the literature on thoracic surgery in such patients.[1-6] In this study, we aimed to evaluate the results of major thoracic surgical procedures performed in newborns and infants.

PATIENTS AND METHODS

The study included 27 consecutive newborn and infant patients (14 males, 13 females) who were operated with thoracic surgical procedures in Atatürk University Medical School Thoracic Surgery Department between January 2000 and December 2014. Exclusion criteria were: *(i)* rigid bronchoscopy (tracheobronchial foreign body aspiration or any other reason), *(ii)* rigid esophagoscopy, or *(iii)* tube thoracostomy performed cases due to neonatal pneumothorax or pleural effusion. Of the 27 patients, 10 (37%) were newborns (mean age 98.4±103.1 days; range 2 to 340 days).

Preoperative diagnoses were congenital cystic pulmonary malformations in 10 patients (37%) (six lobar emphysemas, four cystic adenomatoid malformations) mediastinal cysts in four patients (14.8%) (three gastroenteric cysts, one cystic teratoma), congenital diaphragmatic hernia in four patients (14.8%) (three Morgagni hernias, one Bochdalek hernia), diaphragmatic eventration in three patients (11.2%), mediastinal cavernous hemangioma in two patients (7.4%), Jeune syndrome in one (3.7%), cleft sternum in one (3.7%), congenital chylothorax in one (3.7%), and chest wall tumor (lipoblastoma) in one (3.7%) patient.

Age, gender, diagnosis, symptoms, surgical procedure, postoperative hospital stay, complications, and mortality rates of the patients were recorded and evaluated. The study protocol was approved by the Atatürk University Medical School Ethics Committee. The study was conducted in accordance with the principles of the Declaration of Helsinki.

RESULTS

The most common symptom was dyspnea in 25 patients (92.6%). There was severe dyspnea in eight patients. Three of them were diagnosed as congenital lobar emphysema, congenital cystic adenomatoid malformation, and enteric cyst and they were intubated. The other five patients were followed-up with oxygen. Other symptoms were swelling in the chest, a narrow rib cage, and sternum cleft. One patient was asymptomatic.

Patients diagnosed as congenital lobar emphysema underwent lobectomy (three middle lobectomies, two left upper lobectomies, and one superior bilobectomy) (Figure 1). Three patients with congenital cystic adenomatoid malformation

Table 1. Diagnosis of the cases

Diagnosis	Male	Female	n	%
Congenital cystic pulmonary malformations			10	37.0
Congenital lobar emphysema	3	3		
Congenital cystic pulmonary malformation	3	1		
Mediastinal cyst			4	14.8
Gastroenteric cyst	2	1		
Cystic teratoma		1		
Congenital diaphragmatic hernia			4	14.8
Morgagni hernia	3			
Bochdalek hernia		1		
Diaphragmatic eventration	1	2	3	11.2
Mediastinal cavernous hemangioma	1	1	2	7.4
Jeune syndrome		1	1	3.7
Cleft sternum		1	1	3.7
Congenital chylothorax		1	1	3.7
Chest wall tumor (lipoblastoma)	1		1	3.7
Total	14	13	27	100

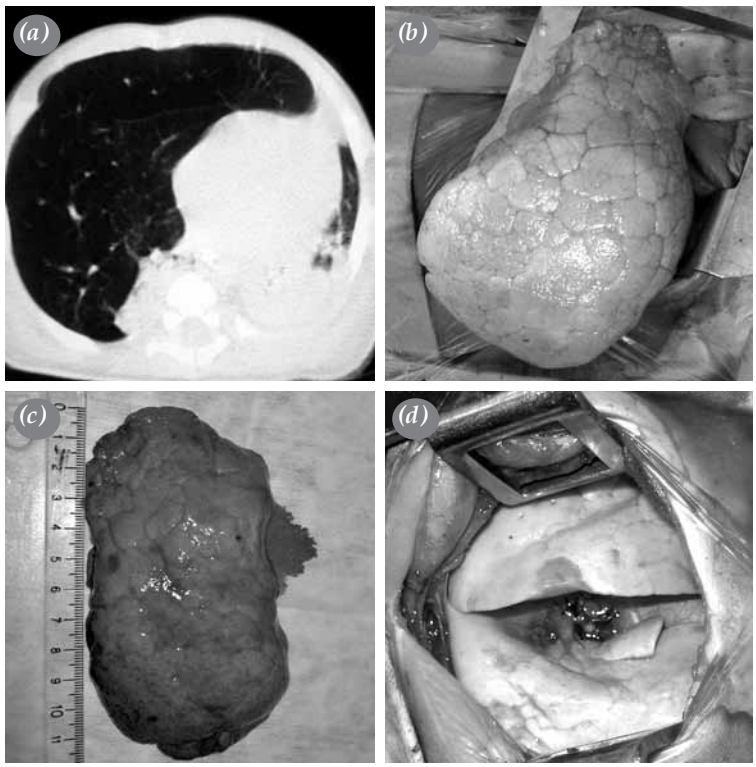

Figure 1. Thoracic computed tomography **(a)** image of 70-day-old female patient with congenital lobar emphysema and view of intraoperative appearance of middle lobe **(b)**, resection material **(c)** and upper and lower lobes after middle lobectomy **(d)**.

underwent wedge resection (right upper, middle and lower lobe in one patient, right upper and middle lobe in one patient, and right upper lobe in the other patient). Fourth patient with cystic congenital adenomatoid malformation underwent right upper lobe anterior segment and basilar segmentectomy (Figure 2). Four patients were operated for mediastinal cyst excision via thoracotomy (Figure 3). Four patients of diaphragmatic hernia were treated by laparotomy (n=2) or thoracotomy (n=2). Patients of diaphragmatic eventration were operated by plication with right (n=2) or left (n=2) thoracotomy (Figure 4). Mediastinal tumor resection was performed with left thoracotomy in two patients (Figure 5). Thoracic

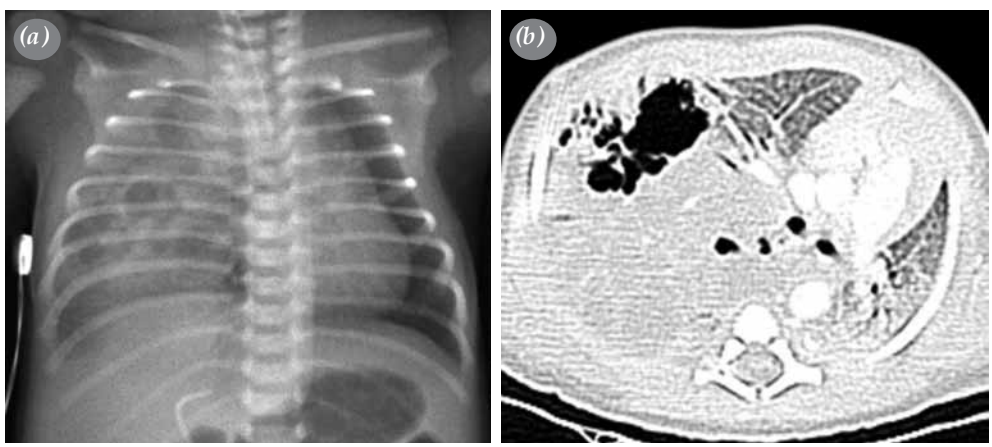

Figure 2. A two-day-old female patient with congenital cystic adenomatoid malformation; chest radiography **(a)** and axial images of thoracic computed tomography **(b)**.

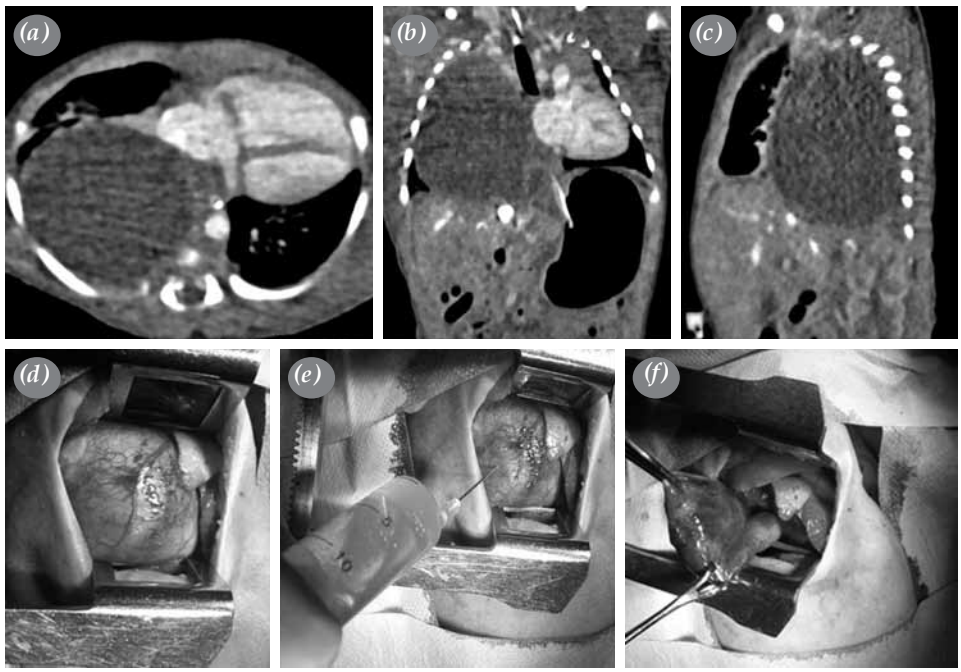

Figure 3. A two-day-old male patient with gastroenteric cyst; **(a)** axial and **(b)** coronal images of thoracic computed tomography. **(c)** Content of cyst and **(d-f)** contents have been emptied after cyst wall.

duct ligation was performed on the congenital chylothorax with a right thoracotomy. Chest wall tumor was resected and diagnosed as lipoblastoma by evaluation of resected specimen. Pericardium repair/sternum closure were performed on the cleft sternum. The Jeune syndrome patient underwent chest expansion procedure (Figure 6).

All patients were followed-up in the thoracic surgery clinic and intensive care unit postoperatively. In addition, all patients were consulted to a pediatrician daily.

In postoperative period, atelectasis was observed in two (7.4%) patients (one case of congenital lobar emphysema and one case of cavernous hemangioma). Postoperative hospital mortality was observed in one patient diagnosed as Jeune syndrome due to respiratory failure on 30th day. The mean postoperative hospital stay was 6.9 days (range 1-30 days).

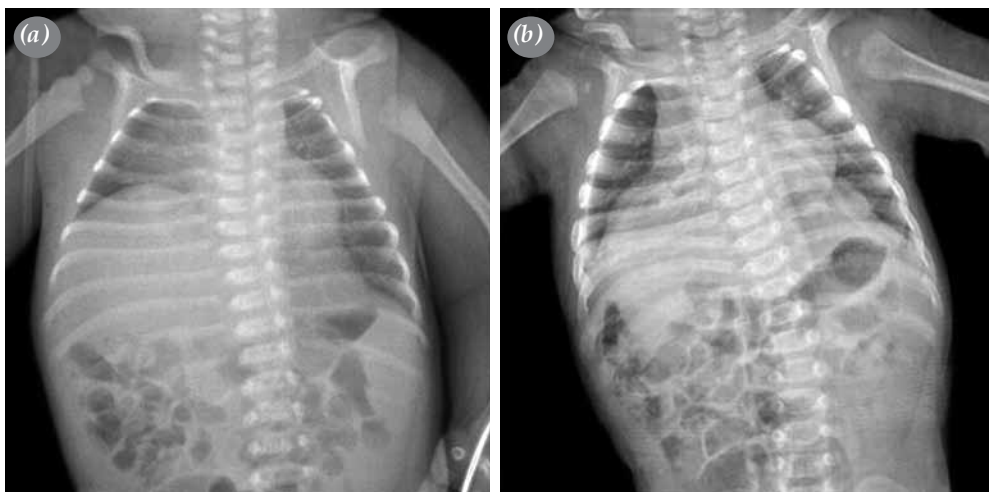

Figure 4. An 80-day-old male patient with congenital diaphragmatic eventration; **(a)** preoperative and **(b)** postoperative chest radiographs.

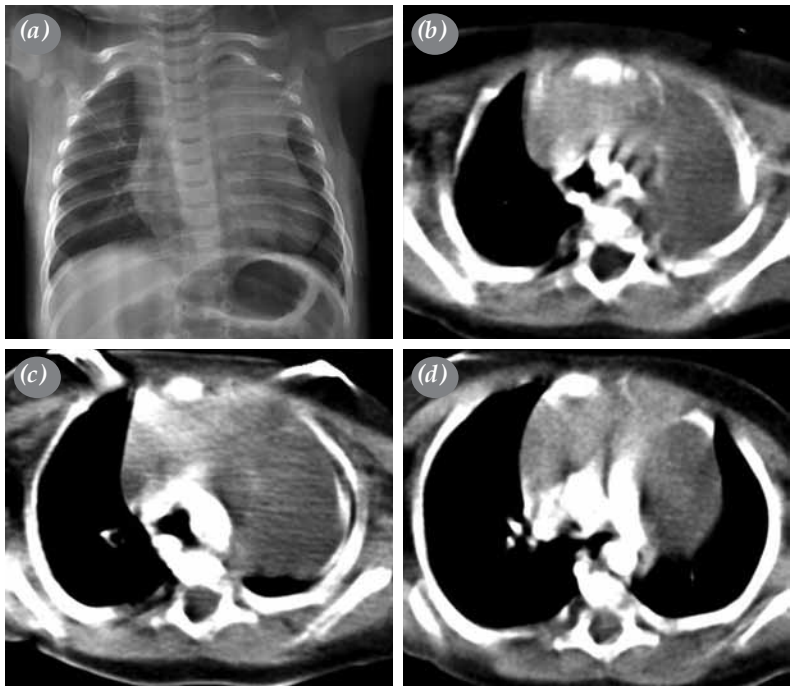

Figure 5. A 120-day-old male patient with mediastinal cavernous hemangioma; **(a)** chest radiograph and **(b-d)** axial computed tomography images.

DISCUSSION

Despite the small number of patients in our study, our results indicate that many different diseases may require thoracic surgical intervention. In newborns and infants, all thoracic surgical procedures could be performed for congenital pathologies except for one case (lipoblastoma).

In our study, majority of thoracic surgical procedures were performed for congenital cystic pulmonary malformations. The most common indication for surgery was congenital lobar emphysema. One-third of the congenital lobar emphysema cases are seen at birth and most cases are diagnosed in the first six months.[7] It is more common in males. Lesions are usually unilateral and involve a single lobe. The most important symptom is dyspnea, and dyspnea may be very severe in the some cases. Pulmonary resection should be performed in patients with persistent and progressive respiratory

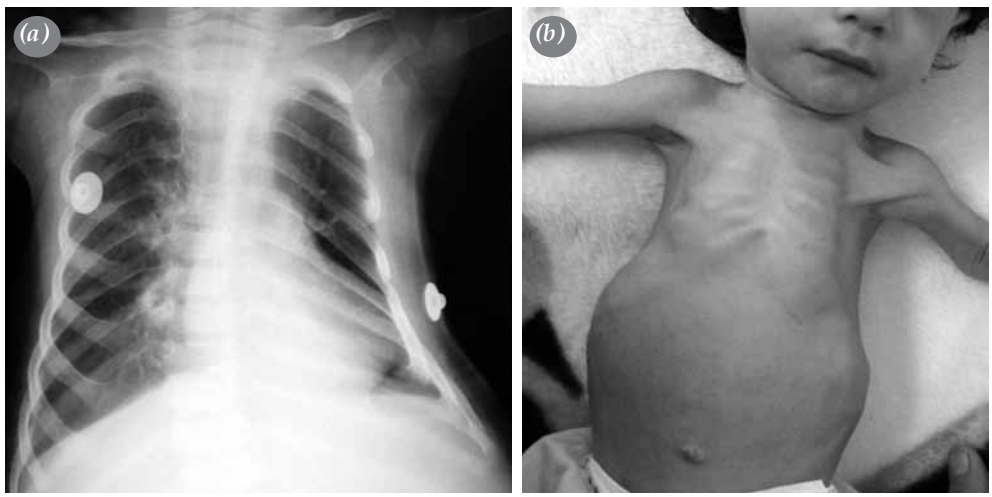

Figure 6. A 30-day-old female patient with Jeune syndrome; **(a)** chest radiography and **(b)** chest and abdominal morphology.

distress.[7] In our study, three of the patients were boys and three were girls. Pulmonary resection was performed for affected lobe in all patients. Middle lobectomy and left upper lobectomy were performed for three and two patients, respectively. However, both right upper and middle lobes were affected in one patient. Therefore, superior bilobectomy was performed in this patient.

Congenital cystic pulmonary malformation is a rare developmental anomaly of the lower respiratory tract and the most common congenital pulmonary lesion. It may originate from trachea, bronchi, bronchioles or alveolar tissue and be caused by cystic adenomatous elements including hamartomas. Both lungs may be affected equally and it can occur in all lobes. Lesions are usually limited to one lobe; it is rarely seen in multiple lobes. There are four types that are clinically and pathologically defined. Type 1 is the most common form (60%-70%).[8,9] Typical findings include tachypnea and respiratory distress. In symptomatic cases, surgical treatment is preferably lobectomy. Wedge resection of the lesion is not recommended because it has clearly indistinguishable boundaries. Surgery is performed in the newborn period with severe respiratory distress. However, it may be performed in less symptomatic patients electively in late childhood.[8] In our study, four patients underwent surgical treatment for congenital cystic pulmonary malformations. All of our patients were type 1. In three of them, there was multiple lobe involvement. Therefore, extensive pulmonary resection was avoided to save the anatomical and physical development of patient's body. Wedge resection of the affected lobes were performed in those three patients. Upper and lower lobe segmentectomy were performed in the other patient.

Mediastinal cysts are usually asymptomatic and detected after two decades despite being congenital lesions.[10] Our four patients had three gastroenteric cysts and one cystic teratoma. Gastroenteric cysts can be seen in almost every age group but are usually detected in childhood. These are gastric cysts which are located in the mediastinum. As with other cystic lesions in the mediastinum, they pressure on adjacent tissues. Furthermore, a peptic complication of these cysts is an important feature. Many of them are localized in the right. Complete excision of the cyst is performed for treatment.[11] One of our three cases was localized in the left side and other two cases were localized in the right side. There was no connection with the gastrointestinal tract.

Benign cystic teratoma is a subset of teratoma. It may cause life-threatening respiratory distress depending on the compression of the mass within a few days after birth, and surgical intervention may be required. After the treatment, the prognosis is excellent.[12] Our patient was 70 days old and underwent excision with a right thoracotomy.

The most common type of congenital diaphragmatic hernia is Bochdalek hernia; other types include Morgagni hernia, diaphragm eventration, and central tendon defects of the diaphragm. Eighty five percent of Bochdalek hernia is located in the left posterolateral region.[2] Pulmonary hypoplasia is seen in some patients. Symptoms usually occur with respiratory distress in the first few hours after birth. Sometimes, symptoms may be seen in a later period, depending on the size of the defect. Morgagni hernia is usually seen in the right anterior location. Unlike Bochdalek hernia, Morgagni hernia cases are often detected in adulthood. The treatment of diaphragmatic hernia is surgery. Abdominal or thoracic approach can be performed.[2,13] In this study, one of four patients was diagnosed with Bochdalek hernia. Patient was not in the neonatal period and underwent surgical treatment without the need for extracorporeal membrane oxygenation because he/she did not have respiratory distress. In contrast to the literature, Morgagni hernia was more frequent in our study.

Diaphragmatic eventration, which is development of primary pathology in utero insufficient muscular membranous diaphragm, is a rare anomaly. In newborns with severe and unilateral eventration, serious cardiovascular symptoms may be seen secondary to pulmonary hypoplasia at birth. Respiratory distress may be mild to severe forms even requiring ventilatory treatment. In the diaphragmatic eventration surgery, hemidiaphragm is moved inferiorly approximately two intercostal distances.[3,14] Three of our patients had diaphragmatic eventration and were operated with diaphragmatic plication.

Mediastinal cavernous hemangioma constitutes less than 0.5% of all mediastinal tumors.[15] It has been reported very rarely in newborns and infants. Mediastinal hemangioma may be easily confused with malignities due to solid growth pattern, large size, and infiltrative enlargement. For these reasons, surgical excision is a reliable method for diagnosis and treatment.[15] In our study, two patients underwent surgical treatment for mediastinal cavernous hemangioma.

Thoracic surgical indications are rare in newborns and infants, but include a broad spectrum of diseases.

In our study, surgical procedures were performed due to Jeune syndrome, cleft sternum, chylothorax, and congenital chest wall tumor.

In newborns and infants, thoracic surgical procedures can be performed by different surgical departments depending on geographical regions. We think thoracic surgeons should perform necessary surgery in these patients due to their experience in thoracic structures and complications. Patients can be followed-up in thoracic surgery clinics with consultation to pediatricians.

This study was unavoidably limited by its single-institution, retrospective analysis, small sample size and the among different diagnosis.

In conclusion, although indications for thoracic surgery are rare in newborns and infants, this intervention may be lifesaving and performed successfully in majority of patients. Moreover, diagnosing and treating newborns and infants require a multidisciplinary team work.

Declaration of conflicting interests

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