Tracheal Diverticulum: Report of Three Cases

Trakeal Divertikül: Üç Olgu Nedeniyle

Emine ARGÜDER, MD, Msc,[a]
Mükremin ER, MD, Msc,[a]
Ayşegül KARALEZLİ, MD, Msc,[a]
H. Canan HASANOĞLU, MD, Prof.,[a]
Hüseyin ÇETİN, MD, Msc[b]

Clinics of
[a]Chest Diseases,
[b]Radiology,
Ankara Atatürk Training and Research Hospital, Ankara

Geliş Tarihi/*Received:* 27.06.2011
Kabul Tarihi/*Accepted:* 12.09.2011

Yazışma Adresi/*Correspondence:*
Emine ARGÜDER, MD, Msc
Ankara Atatürk Training and Research Hospital,
Clinic of Chest Diseases, Ankara, TÜRKİYE/TURKEY
drgullu2000@gmail.com

ABSTRACT A right paratracheal air cyst in the thoracic inlet is a rare lesion. It generally originates from respiratory or gastrointestinal system disorders such as laryngocele, pharyngocele, Zenker's diverticule, tracheal diverticulum, apical hernia, lymphoephitelial cyst, or bronchogenic cyst. We present three cases of right paratracheal air cysts related to the trachea which were confirmed as tracheal diverticulum via three-dimensional reconstruction computed tomography and virtual bronchoscopic evaluation. These methods provide better demonstration of the lesions. These lesions may be congenital or acquired. Acquired form is more common. Tracheal diverticulum is generally overlooked, neither reported on chest computed tomography nor recognized unless it is complicated. We, therefore, aim to draw attention to this entity. In additional, a discussion of the evolution, importance, and complications of, as well as treatments for tracheal diverticulum is made in light of the currently available literature.

Key Words: Trachea; diverticulum; congenital; imaging, three-dimensional

ÖZET Toraks girişinde sağ paratrakeal hava kisti nadir rastlanılan bir durumdur. Genellikle solunum sistemi ya da gastrointestinal sistemden kaynaklanır. Bunların arasında laringosel, faringosel, Zenker divertikülü, trakeal divertikül, apikal herni, lenfoepitelyal kist ya da bronkojenik kist yer alır. Burada trakea ile ilişkili sağ paratrakeal hava kisti olan üç olgu sunulmuştur. Lezyonların bilgisayarlı tomografi ile üç boyutlu rekonstrüksiyonu ve sanal bronkoskopik değerlendirilmesi ile trakeal divertikül olduğu doğrulanmıştır. Bu yöntemler trakeal divertikülün daha iyi değerlendirilmesini sağlar. Trakeal divertikül konjenital ya da edinsel olabilir. Edinsel lezyon daha sık görülür. Trakeal divertikül genellikle gözden kaçan, komplike olmadıkça fark edilmeyen ve toraks bilgisayarlı tomografisi raporlarında genellikle bahsedilmeyen bir patolojidir. Bu nedenle bu makalede bu konuya dikkat çekmek istenilmiştir. Buna ek olarak trakeal divertikülün gelişimi, önemi, komplikasyonları ve tedavisi güncel bilgiler ışığında tartışılmıştır.

Anahtar Kelimeler: Trakea; divertikül; konjenital; görüntüleme, üç-boyutlu

Turkiye Klinikleri Arch Lung 2012;13(1):33-8

Paratracheal air cyst is rare and generally described as an incidental finding during chest computed tomography (CT).[1-3] Several lesions must be taken into consideration during differential diagnosis. While a few of them are related to the gastrointestinal system, the others are related to the respiratory system.[1] One of them is tracheal diverticulum. This entity can be either congenital or an acquired form.[2,3] We aim to present here three cases of acquired tracheal diverticulum as incidental at chest CT

finding accompanied by radiological reconstruction images and virtual bronchoscopic evaluation, and to review the literature on tracheal diverticulum.

CASE REPORTS

CASE 1

A 77-year-old woman presented with dyspnea and cough for one year. She was non-smoker and had been treated for lung tuberculosis 50 years previously. Clinical respiratory examination revealed inspiratory crackles at the lower of the left lung and upper of the right lung fields. Oxygen saturation was 75% without oxygen therapy. The chest CT showed two thin-walled air cysts in the right posterolateral and near side of the trachea and diffuse bronchiectasis. Fiberoptic bronchoscopy (FOB) was not applied due to respiratory failure. A three-dimensional reconstruction CT showed very small direct connections between the trachea and the paratracheal air cysts (Figures 1, 2). Also, virtual bronchoscopy demonstrated the mouth of a diverticulum (Figure 3).

CASE 2

A 34-year-old woman was referred for dyspnea and cough by the department of Neurology. Her symptoms had persisted for 7 years and had been progressive for a month. She had had hyperthyroidism for 7 years and had been investigated for ocular myasthenia gravis in the Neurology department. She had smoked 10 pack-years of cigarette. Expiration time was prolonged on respiratory examination. Pulmonary function tests revealed a normal pattern. The chest CT showed a thin-walled air cyst in the right posterolateral and near side of the trachea. The patient refused FOB. Three-dimensional reconstruction CT showed a very small direct connection between the trachea and the paratracheal air cyst (Figures 4, 5). Virtual bronchoscopy was

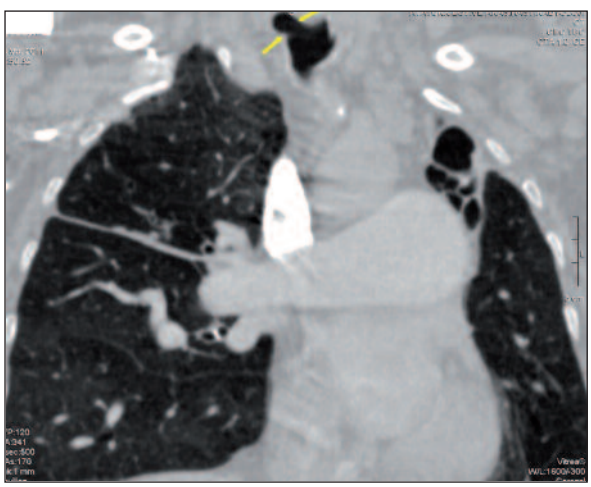

FIGURE 1: Chest CT scan in coronal plane. The tracheal diverticulum appears posterolaterally of the trachea.
(See for colored form http://akcigerarsivi.turkiyeklinikleri.com/)

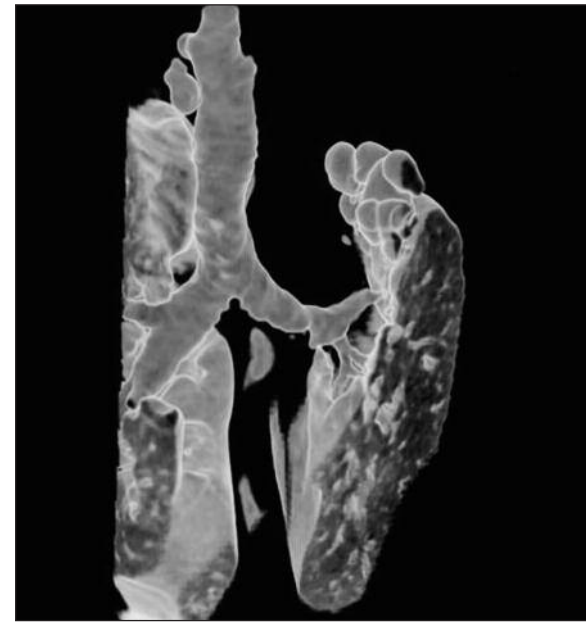

FIGURE 2: Anterolateral three-dimensional image of the trachea shows an air cyst communicating with the tracheal wall.

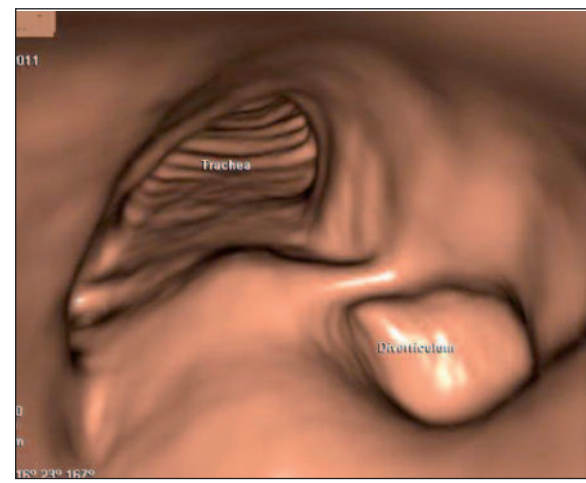

FIGURE 3: Virtual bronchoscopic view of the diverticulum mouth.
(See for colored form http://akcigerarsivi.turkiyeklinikleri.com/)

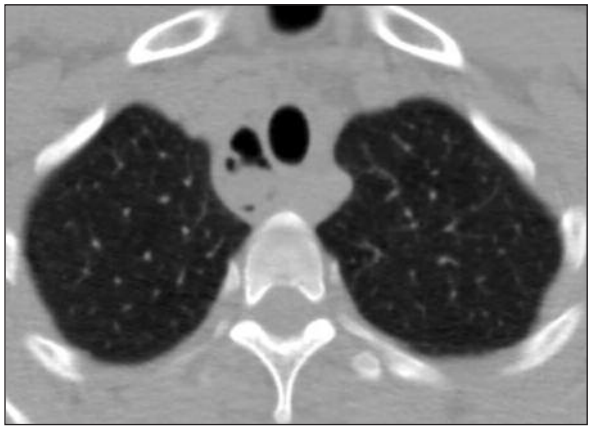

FIGURE 4: Chest CT scan shows paratracheal air cyst posterolaterally of the trachea.

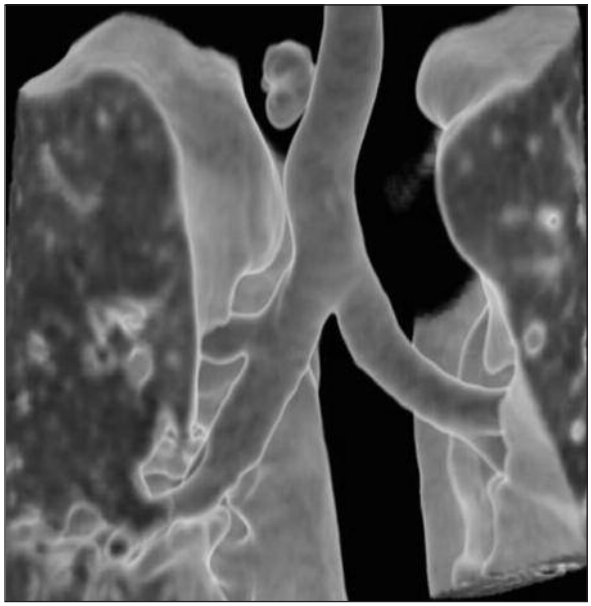

FIGURE 5: Anterolateral three-dimensional image of the trachea shows air cyst communicating with the tracheal wall.

applied; but did not reveal the mouth of the diverticulum possibly due to secretion. An intradiverticular view was obtained via virtual bronchoscopy (Figure 6).

CASE 3

A 54-year old male, ex-smoker, with a history of larynx carcinoma for 7 months consulted our department due to having dyspnea for a week. He had received both chemotherapy and radiotherapy for larynx carcinoma. His chest CT demonstrated air filled lesions originating from the trachea in the right paratracheal region and at the level of the thoracic inlet (Figures 7, 8). A connection with the tracheal lumen was also visible. No wall thickening or calcifications were found. In addition to these findings, multiple mediastinal lymph nodes and bilaterally pulmonary acinar nodules were detected on the CT. FOB could not be implemented due to a laryngeal lesion. Virtual bronchoscopy was applied; but, did not reveal the mouth of the diverticulum possibly due to a membrane. An intradiverticular view was obtained via virtual bronchoscopy (Figure 9).

An informed consent form was obtained from each patient.

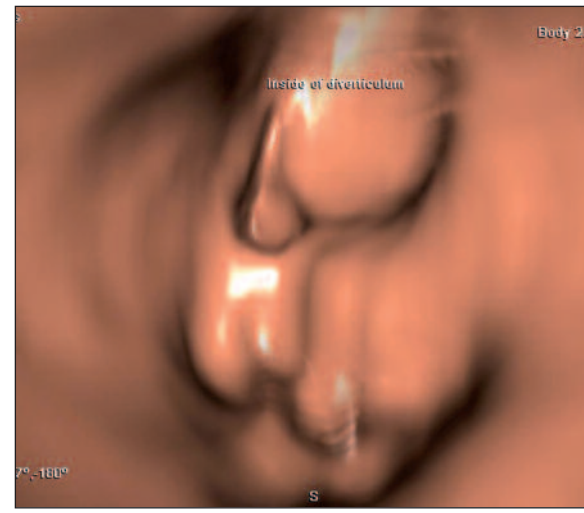

FIGURE 6: Virtual bronchoscopic view of inside the diverticulum.
(See for colored form http://akcigerarsivi.turkiyeklinikleri.com/)

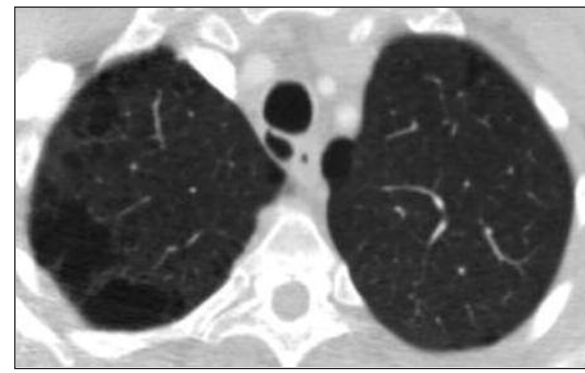

FIGURE 7: Chest CT scan shows paratracheal air cyst posterolaterally of the trachea.

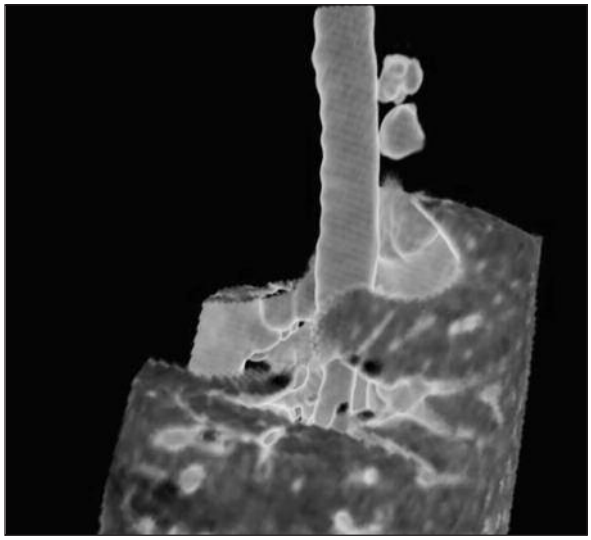

FIGURE 8: Lateral three-dimensional image of the trachea shows air cysts communicating with the tracheal wall.

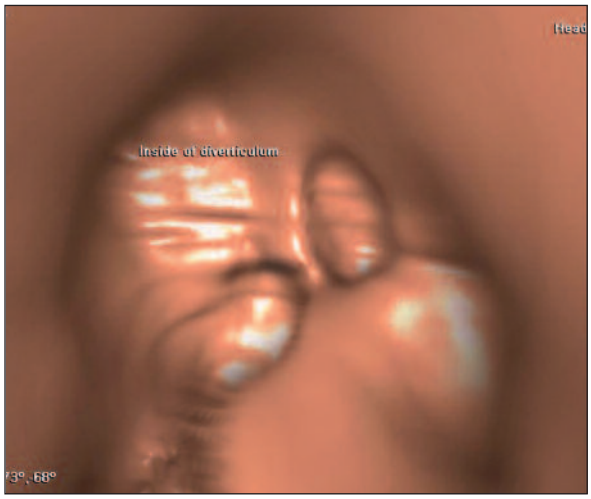

FIGURE 9: Virtual bronchoscopic view of inside the diverticulum.
(See for colored form http://akcigerarsivi.turkiyeklinikleri.com/)

DISCUSSION

Paratracheal air filled lesions originate from respiratory or gastrointestinal system disorders such as laryngocele, pharyngocele, Zenker's diverticule, tracheal diverticulum, apical hernia, lymphoephitelial cyst, or bronchogenic cyst.[1-5] Tracheal diverticulum is rarely described at chest CT and its frequency occurs at about 0.75-1%. It can be either congenital or acquired. The majority of these lesions are asymptomatic, being discovered as incidental findings on radiological imaging.[1-3,6]

The congenital form is smaller, located nearly 4-5 cm below the vocal cords, and is generally multiple. It is a small-mouthed opening of the tracheal mucosa with a predilection for males. The histological structure of this diverticulum resembles that of the trachea. It is hypothesized that it is developed as a result of a defect in endodermal differentiation during development of the tracheal pars membranacea or cartilage during embryonic life and is thought to be a supernumerary branch of the trachea.[7] The presence of cartilaginous rings in the wall of the diverticula strongly suggest the congenital form. It is not normally detected in infancy unless it is suggested by recurrent episodes of tracheobronchial infection or in association with other malformations.[2] An association between tracheal diverticulum and cystic adenomatoid malformation has been reported as a unique case report.[8]

The acquired form is sometimes referred to by the term tracheocele. It may arise from increased intraluminal pressure during repeated respiratory infection or chronic cough at any level of the trachea. As a result of this, it emerges at weak areas of the tracheal wall resulting in mucosal herniation. Additionally, esophageal or tracheal surgery, tracheostomy closure and orotracheal intubation are other etiologic factors.[1,2,9-13] They usually appear as a wide-mouthed pouch, with 98% of lesions located on the right posterior-lateral side of the trachea and one or more of them having a connection with the trachea. The histological structure of this diverticulum wall consists only of respiratory epithelium.[1,2] The esophagus and aortic arch protect the left side of the airway for development of diverticula.[1] The connection between the tracheal lumen and diverticulum may be shown by bronchoscopy or chest CT, which exploit thin sections or reconstructed images.[1-3,14,15] Furthermore, chest CT evaluates the peri-diverticular tissues, its exact location, size and wall thickness, and determines the nature of the tracheal diverticulum due to the presence of or absence of cartilaginous rings in the diverticulum wall.[1,6,16]

Most cases are asymptomatic, but when symptoms are present they are usually nonspecific. Dyspnea, stridor, dysphonia, hemoptysis, pneumomediastinum, and chronic chest infections may be

observed probably due to the tracheal diverticulum's propensity for acting as a reservoir for secretions. In addition, cases may be investigated for dysphagia, cervical swelling, or pharyngeal disorders.[1,10,16-21] There is no consensus on the treatment of tracheal diverticulum. In general, conservative measures, such as antibiotics, mucolytic agents, and chest physiotherapy are recommended.[2,3,10,16] When the diseases is symtomatic and medical therapy is insufficient, this disorder must be treated surgically. A variety of surgical methods have been reported, ranging from resection of the lesion via open neck or thoracic surgery to endoscopic repair, such as electrocauterization or application of fibrin glue by means of catheter.[9,22,23] Surgical endoscopic treatment is accepted as safe, less invasive, conservative, and reproducible.[19]

It was thought that our first and third cases had tracheal diverticula due to chronic lung disorder, and that the second case had tracheal diverticulum due to chronic cough. All our cases of diverticulum were connected to tracheal lumen. However, the presence of connections between the diverticulum and tracheal wall was not pathologically demonstrated in our cases, but rather demonstrated in 3-dimensional reconstruction chest CT, and in the case of the first case patient, the mouth of the divertiulum was revealed by virtual bronchoscopy. No symptoms were esteblished related to the tracheal diverticula's and no treatment was asministered by us to any of the patients. Resectioning of these tracheal diverticula was also not considered as a treatment option for the patients.

In conclusion, acquired tracheal diverticulum is rare entity which may be confused with several lesions. It is generally overlooked, not reported on chest CT, nor recognized unless it is complicated. Therefore, it should be kept in mind as a possible etiology for chronic cough, recurrent lung infection, hemoptysis, dysphagia and pneumomediastinum. In this article, the evolution, importance, complications and treatments of tracheal diverticulum were discussed in light of the current literature.

Acknowledgment

The authors wish to thank Ms Karin Marsden for her valuable evaluation of the manuscript in terms of English grammar.

REFERENCES

1. Goo JM, Im JG, Ahn JM, Moon WK, Chung JW, Park JH, et al. Right paratracheal air cysts in the thoracic inlet: clinical and radiologic significance. AJR Am J Roentgenol 1999;173 (1):65-70.
2. Soto-Hurtado EJ, Peñuela-Ruíz L, Rivera-Sánchez I, Torres-Jiménez J. Tracheal diverticulum: a review of the literature. Lung 2006;184(6):303-7.
3. Mineshita M, Tajima O, Kondo T. Paratracheal air cysts in middle-aged Japanese men. J Bronchol 2006;13(1):6-8.
4. Tanaka H, Igarashi T, Teramoto S, Yoshida Y, Abe S. Lymphoepithelial cysts in the mediastinum with an opening to the trachea. Respiration 1995;62(2):110-3.
5. Türkyılmaz A, Kurt A, Atlaş S, Eroğlu A. [Cervical bronchogenic cyst mimicking trachea diverticule]. Respiratory Diseases 2007;18(2): 126-9.
6. MacKinnon D. Tracheal diverticula. J Pathol Bacteriol 1953;65(2):513-7.
7. Doğan R. Konjenital akciğer anomalileri. Journal of Clinical and Analytical Medicine 2010 (Published online). doi: 10.4328/JCAM. 466.
8. Restrepo S, Villamil MA, Rojas IC, Lemos DF, Echeverri S, Triana G, et al. Association of two respiratory congenital anomalies: tracheal diverticulum and cystic adenomatoid malformation of the lung. Pediatr Radiol 2004;34(3): 263-6.
9. Bhatnagar V, Lal R, Agarwala S, Mitra DK. Endoscopic treatment of tracheal diverticulum after primary repair of esophageal atresia and tracheoesophageal fistula. J Pediatr Surg 1998;33(8):1323-4.
10. Teh BM, Hall C, Kleid S. Infected tracheocoele (acquired tracheal diverticulum): case report and literature review. J Laryngol Otol 2011; 125(5):540-5.
11. Gaissert HA, Grillo HC. Complications of the tracheal diverticulum after division of congenital tracheoesophageal fistula. J Pediatr Surg 2006;41(4):842-4.
12. Henderson CG, Harrington RL, Izenberg S, Dyess DL, Silver FM. Tracheocele after routine tracheostomy. Otolaryngol Head Neck Surg 1995;113(4):489-90.
13. Briganti V, Tavormina P, Testa A, Oriolo L. Giant tracheocele following primary tracheostomy closure in a 3 year old child. Interact Cardiovasc Thorac Surg 2004;3(2): 411-2.
14. Tanaka H, Mori Y, Kurokawa K, Abe S. Paratracheal air cysts communicating with the trachea: CT findings. J Thorac Imaging 1997; 12(1):38-40.
15. Sakarya ME, Ceran S, Koç O, Özbek O, Karabekmez LG, Ödev K. [Bronchocele: demonstration by 3D volume rendering imaging with multidetector computed tomography: case report]. Turkiye Klinikleri J Med Sci 2011;31(1):251-5.
16. Caversaccio MD, Becker M, Zbären P. Tracheal diverticulum presenting with recurrent laryngeal nerve paralysis. Ann Otol Rhinol Laryngol 1998;107(4):362-4.
17. Hernández Pérez L, Pac Ferrer J, Uribe-Etxebarría Lugariza-Aresti N, Jiménez Maestre U, Oleagoitia Cilaurre JM. [Tracheal diverticulum: a cause of dysphagia]. Cir Esp 2010;88 (3):197-8.

18. Pinot D, Breen D, Peloni JM, Gaubert JY, Dutau H, Vervloet D. An incidental finding in a 34-year-old male under investigation for haemoptysis. Diagnosis: The radiological and endoscopic images demonstrate a complex defect along the posterior tracheal wall consistent with acquired tracheal diverticulum. Eur Respir J 2009;33(5):1227-9.

19. Möller GM, ten Berge EJ, Stassen CM. Tracheocele: a rare cause of difficult endotracheal intubation and subsequent pneumomediastinum. Eur Respir J 1994;7(7):1376-7.

20. Sharma BG. Tracheal diverticulum: a report of 4 cases. Ear Nose Throat J 2009;88(1):E11.

21. Koffi-Aka V, Manceau A, Cottier JP, Renjard L, Beutter P. [Tracheocele: a rare cause of pharyngeal disorders]. Ann Otolaryngol Chir Cervicofac 2002;119(3):186-8.

22. Mathur NN, Sardana P, Singh VP, Bais AS. Adult tracheocele with large cervical presentation. J Laryngol Otol 1999;113(4):364-5.

23. Kıyan G. [Surgical approach in lower respiratory malformations]. Turkiye Klinikleri J Pediatr Sci 2006;2(3):76-84.