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A rare cause of respiratory distress in the neonatal period: posterior mediastinal gastroenteric cyst

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Summary

Presentation of mediastinal enteric cysts is observed rarely during neonatal period. They are mostly asymptomatic. The commonest mode of presentation in neonatal period is respiratory distress. Basically, diagnosis of enteric cyst is made with histologic findings. In this article, a case of mediastinal gastroenteric cyst who developed respiratory distress on the fourth day after birth and who was diagnosed with cystic adenoid malformation in the lung on prenatal ultrasonographical examination is presented. Investigations revealed a well defined gastroenteric cyst in the posterior mediastinum with vertebral anomalies. The purpose of this case report is to highlight the clinical diagnosis and management of posterior mediastinal enteric cyst in the neonatal period. *(Turk Arch Ped 2011; 46: 87-9)*

Key words: Enteric cyst, newborn, respiratory distress

Introduction

The patient was born at term by cesarean section as the fourth living baby from the seventh pregnancy of a preeclamptic mother of 35 years old and a healthy father of 43 years old who had no consanguinity. Apgar score was found to be 5 in the first minute and 8 in the fifth minute. The male baby who had a prediagosis of cystic adenoid malformation in the left lung on ultrasonographical examination performed in the 34th gestational week prenatally was hospitalized in the Neonatal Intensive Care Unit after birth. Physical examination findings were as follows: birth weight 3400 g (50-75% p), head circumference 35 cm (50-75% p), heigth 49 cm (50-75% p), pulse rate 152/min, blood pressure 78/32 mmHg. Slight prolongation of expirium in the left lung areas compared to the rigth lung areas was found on lung oscultation. Heart sounds were normal, peripheral pulses were palpated simultaneously with heart sounds. No organomegaly was found on abdominal examination and orogastric catheter was inserted into the stomach. Baseline arterial blood gases were as follows: pH: 7.35, $PaCO_2$: 43 mmHg, PaO2:71 mmHg, HCO3: 19 mmol/l and oxygen saturation: 95%. Complete blood count and serum biochemical values including sodium, potassium, urea, creatinine, calcium and blood glucose were within normal limits. Right and left paracardial consolidated mass with regular contours was found on thorax graphy and corpus defect was noted in T3, T4 and T5 vertebrae (Picture 1). On thoracic computarized tomography, a large mass lesion pushing the trachea, esophagus and aorta mainly in the right side starting from the upper part of the diaphragm and extending to the upper part of tracheal bifurcation was observed. The cyst was found to be consisted of two parts (Picture 2). On esophagography, 1/3 lower part of the esophagus was dislocated anteriorly and slightly to the left and was compressing the trachea. On echocardiographic examination a cystic formation with dimensions of 3.5x5 cm was found behind the heart adjacent to the left atrium. The patient who had no problem until the 4th day of life developed respiratory distress on the 4th day. Blood gases measured simultaneously were as fol-

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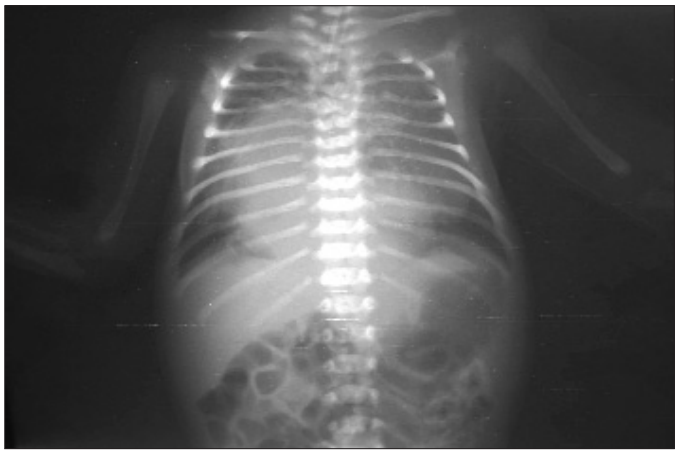
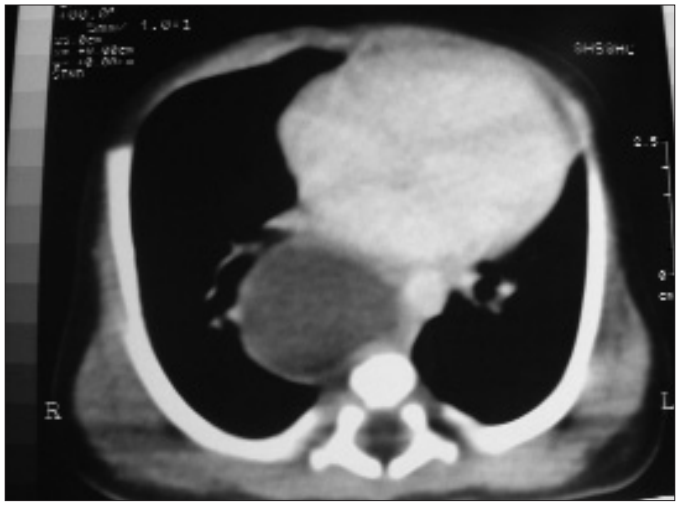
lows: pH: 7.18, $PaCO_2$: 86 mmHg, PaO_2: 47 mmHg, HCO_3: 14.9, oxygen saturation 76%. The patient was intubated and supported by mechanical ventilation. He was extubated on the seventh day. On the 8th day of life he was operated. A cystic formation with a thick wall extending from the right posterior mediastinum fully towards the side was found. The pleura superior to the csyt was partially thickened. The mass was adherent to the surrounding tissues because of inflammation. Thick, slightly mucoid, dirty yellow liquid of 10 cc was evacuated from the mass. Lung areas previously collapsed because of the cyst started to get ventilated. The cyst was observed to be adherent to the vertebral body with a fibrotic band in the middle line. Pathologic examination revealed that the cystic formation was lined with gastric epithelium and intestinal mucosa. Smooth muscle tissue was observed in the cyst wall. Ulcerative inflammation was noted in the epithelium. Consequently, it was found to be compatible with enteric cyst.



Picture 1. Consolidated, well-defined mass localized in the right and left paracardiac regions on chest graphy and corpus defects in T3, T4 and T5 vertebrae



Picture 2. Cystic mass pushing the trachea, esophagus and aorta anteriorly in the right side extending to the upper part of tracheal bifurcation on computarized tomography of the chest

Respiratory distress rapidly improved postoperatively and the patient was discharged 10 days later. No complication or problem was experienced during the 1-year follow-up period after discharge.

Discussion

Mediastinal cyst is seen rarely in the neonatal period and early childhood. The incidence of posterior mediastinal cyst is reported to be 7% among all mediastinal cysts (4). Enteric cysts in the posterior mediastinum are generally asypmtomatic. If the cyst is related to the respiratory tract, respiratory distress may develop. The cyst may be infected and may cause respiratory complications by showing characteristics of sepsis. Observation of inflammatory tissue cells in the cystic fluid in our case was compatible with the literature (5).

Enteric cyst is a developmental disorder of the foregut containing esophageal duplication and neuroenteric cysts (6). The term of “neuroenteric cyst” is used to define the loss of association of notachord connection secondary to vertebral or neural anomalies to the foregut. The cyst can be observed to be connected to the vertebrae with a fibrous cord (1). The cyst was bound to the vertebral corpus also in our case and corpus defect was present in T3, T4 and T5 vertebrae.

Neuroenteric cysts generally contain enteric tissue with both neural and gastric mucosa. Approximately 90% of neuroenteric cysts are observed in the posterior mediastinum. In 50% of the cases, vertebral anomalies including scoliosis, anterior spina bifida, hemivertebrae and butterfly vertebrae accompany (7). The wall of esophageal duplication cysts contains gastrointestinal epithelium and 50-60% of this contains gastric mucosa and pancreatic tissue. Mediastinal enteric cyst content is different from esophageal cysts. Enteric cyst is generally behind the mediastium on the right side and behind the heart and frequently extends to the right hemithorax (7). Clinically, it causes respiratory distress because of mass effect. In our case, the patient who had no sign during the first 4 days after birth developed respiratory distress after the fourth day. Examinations revealed that the cyst extended to the right hemithorax.

Very few number of posterior mediastinal enteric cysts have been reported in the neonatal period and early childhood (8-10). It is difficult to differentiate cyst type preoperatively. Chest graphy usually reveals a round soft tissue cyst localized typically in the posterior or middle mediastinum. The wall of the cyst is thicker than the wall of a bronchogenic cyst. Radioisotope examination performed with technetium 99 is useful in the diagnosis of enteric cyst. Computarized tomography screening can be used to determine the anatomic localization of the cyst (11). Surgical excision is the main treatment (12,13).

Consequently, mediastinal enteric cysts may display a severe prognosis and be life-threatening because of mass effect on the adjacent organs, although they are benign. Posterior mediastinal enteric cyst should be kept in mind in subjects in whom a suspicious mass was found in the lungs on prenatal ultrasonogrphic examination and delivery of these babies should be performed at sites where neonatal intensive care units are present. It should be kept in mind that appropriate intervention performed early in collaboration with the surgical team is life-saving.

Conflict of interest: None declared

References

1. Birmole BJ, Kulkarni BK, Vaidya AS, Borwankar SS. Intrathoracic enteric foregut duplication cyst. J Postgrad Med 1994; 40: 228-30. (Abstract)
2. Kumar R, Jain R, Rao KM, Hussain N. Intraspinal neurenteric cysts. Report of three paediatric cases. Childs Nerv Syst 2001; 7: 584-8. (Abstract) / (Full Text) / (PDF)
3. Azzie G, Beasly S. Diagnosis and treatment of foregut duplications. Semin Pediatr Surg 2003; 12: 46-54. (Abstract) / (PDF)
4. Sethi GK, Marsden J, Johnson D. Duplication cysts of the esophagus. South Med J 1974; 67: 616-8. (Abstract) / (PDF)
5. Yadav TP, Bhardwaj M, Aggarwal S, Mohan B, Ndeswari A. Mediastinal enteric cyst infected with Salmonella typhi. Indian Pediatr 2000; 37: 667-70. (Abstract)
6. Nawaz A, Matta H, Jacobsz A, Ansari F, Al-Khouder G, Al-Salem A. İntrathoracic foregut duplication cysts in neonates. Annals of Saudi Medicine 2001; 21: 206-9. (PDF)
7. King RM, Telander RL, Smithson WA, Banks PM, Han MT. Primary mediastinal tumors in children. J Pediatr Surg 1982; 17: 512-20. (Abstract) / (PDF)
8. Mam MK, Mathew S, Prabhakar BR, Paul R, Jacob S. Mediastinal enterogenic cyst presenting as paraplegia--a case report. Indian J Med Sci 1996; 50: 337-9. (Abstract)
9. Alrabeeah A, Gillis DA, Giacomantonio M, Lau H. Neurenteric cysts-a spectrum. J Pediatr Surg 1988; 23: 752-4. (Abstract) / (PDF)
10. Rammurty DV, Soans B, Mohan M, Gupta HO, Baijal VN, Kulshrestha R. Enterogenic cyst in mediastinum. Indian Pediatr 1990; 27: 1109-11. (Abstract)
11. Fernandes ET, Custer MD, Burton EM, et al. Neurenteric cyst: surgery and diagnostic imaging. J Pediatr Surg 1991; 26: 108-10. (Abstract)
12. Newnham JP, Crues JV, Vinstein AL, Medearis AL. Sonographic diagnosis of thoracic gastroenteric cyst in utero. Prenat Diagn 1984; 4: 467-71. (Abstract)
13. Perek A, Perek S, Kapan M, Goksoy E. Gastric duplication cyst. Dig Surg 2000; 17: 634-6. (Abstract) / (Full Text) / (PDF)