

Posterior mediastinal benign cystic teratoma in an infant – a rare cause of recurrent respiratory distress

Subhasree Beura¹, Baikuntha Narayan Mishra², Ranjit Kumar Joshi³, Prabin Prakash Pahi⁴, Meghmala Sadhukhan⁵

From ¹Senior Resident, Department of Pediatrics, ²Consultant, Department of Pediatric-Surgery, ³Consultant, ⁴Consultant, ⁵Junior Consultant, Department of Pediatrics, AMRI Hospital, Bhubaneswar, Odisha, India

Correspondence to: Subhasree Beura, Department of Pediatrics, AMRI Hospital, Bhubaneswar - 751 030, Odisha, India. E-mail: beurasubhasree@gmail.com

Received - 16 April 2020

Initial Review - 07 May 2020

Accepted - 01 June 2020

ABSTRACT

Mediastinal benign teratoma is a rare primary germ cell neoplasm composed of well-differentiated tissues derived from more than one of the three embryonic germ cell layers. Most of the mediastinal teratomas are found in the anterior mediastinum. The posterior mediastinal teratomas are rare. We describe a rare case of an infant with benign cystic teratoma in the posterior mediastinum presenting with recurrent respiratory distress, successfully treated by surgical excision with no recurrence in a 1-year follow-up.

Key words: *Infant, Posterior mediastinum, Respiratory distress, Teratoma*

Teratoma is a germ cell tumor, derived from pluripotent cells composed of complex differentiated tissues comprising elements of more than one embryonic cell layers with ectoderm, endoderm, and mesoderm [1-3]. The incidence of teratoma is around 1 in 4000 live births [2]. They can occur in any region of the body and maybe gonadal or extragonadal. Extragonadal teratomas are common in infancy and early childhood, whereas gonadal is seen in late childhood. The most common site of teratoma is the sacrococcygeal or pre-sacral region in about 40% cases. The other sites are ovary (25%), testis (12%), brain (5%), neck, and mediastinum (18%) [3]. About 1–3% of all germ cell neoplasms are located in mediastinum [2] with the majority in the anterior mediastinum.

The posterior mediastinal teratomas are very rare and comprise 3–8% of all mediastinal tumors [4]. Teratomas may be mature or immature. Mediastinal teratomas may be asymptomatic or may present with dyspnea, cough, or recurrent respiratory tract infections due to compression of adjacent structures, tumor rupture, or secretion of hormones and enzymes [5]. Asymptomatic cases may be detected incidentally with an X-ray chest. In symptomatic or incidentally detected asymptomatic cases, computed tomography (CT)/magnetic resonance imaging (MRI) may demonstrate the lesion more accurately [6]. Children, especially those with benign tumors, have a very high survival rate after surgical resection [3]. Herein, we describe our experience with a posterior mediastinal cystic teratoma in a 4-month-old baby.

CASE REPORT

A 4-month-old male baby weighing 5.4 kg was admitted to our hospital with respiratory distress for 3 weeks. He had a history of similar episodes 1 month back for which he was admitted twice in another hospital and treated as a case of pneumonia with intravenous (IV) ampicillin and gentamicin for 2 weeks initially, and with ceftriaxone and amikacin subsequently for another 7 days, before getting admitted to our hospital.

On admission, the baby was febrile (39.2°C), with a pulse rate of 140/min, tachypneic with a respiratory rate of 52/min with chest wall retractions, but was maintaining saturation in room air with SpO₂ of 98%. There were decreased breath sound over left hemithorax with bilateral crepitations and conducted sound on chest auscultation. All other systemic examinations were within normal limits.

Sepsis screen sent was negative, but injection piperacillin and tazobactam were started empirically in a dose of 300 mg/kg/day. The posteroanterior chest radiograph was normal, but MRI chest revealed a well-defined cystic lesion in posterior mediastinum which was compressing left main bronchus and esophagus with mild dilatation of the distal esophagus up to the gastroesophageal (GE) junction and consolidation of the right lower lobe in the apicoposterior segment (Fig. 1).

Esophagoscopy and upper gastrointestinal endoscopy were done which showed no evidence of compression internally, with intact GE junction and no evidence of reflux. An exploratory left

thoracotomy was performed. A cystic lesion of size 3.5 cm × 2.5 cm × 1.5 cm was found behind the arch of aorta, sitting over the left main bronchus and esophagus. The cyst was dissected free from surrounding structures and it was removed in toto (Fig. 2) and was sent for histopathological study which confirmed it to be a benign mature cystic teratoma.

Serum beta-human chorionic gonadotropin (HCG) and alpha-fetoprotein (AFP) levels were within normal limits. Postoperatively IV antibiotic was continued for the next 5 days, before switching to oral cefpodoxime at 10 mg/kg/day for another 5 days. He had an uneventful recovery and was discharged on the 10th post-operative day. No antibiotics were advised on discharge. He was doing well on follow-up visits.

DISCUSSION

Teratomas are embryonic neoplasms that arise from totipotent cells and contain elements from all the three germ layers (ectoderm, mesoderm, and endoderm) and are uncommon in children accounting for about 3% of all childhood neoplasms [6]. The primary mediastinal teratomas account for approximately 8–20%



Figure 1: Magnetic resonance imaging chest showing a cystic lesion in the posterior mediastinum



Figure 2: The picture showing the dissected cystic structure

of mediastinal neoplasms [3]. Approximately 8% of mediastinal tumors are benign teratomas, with 82% of these in the anterior, 4% in the posterior, and 14% in the middle mediastinum [3].

Mediastinal teratoma presenting with respiratory distress are more commonly located in the anterior mediastinum. Posterior mediastinal teratomas are reported in a wide age group, ranging from newborn to 18 years old. There is a definite male predominance. Most of the symptoms due to mediastinal teratoma result from the compression of adjacent structures [3]. Respiratory distress is the most common complaint, especially in the neonatal period and early infancy [7]. The other common symptoms are recurrent respiratory tract infections and cough.

Jaiswal *et al.* in 2014 described an incidentally detected posterior mediastinal teratoma [4]. Rupture and infection may produce complications, as reported by Sarin *et al.* in 2006 [5]. El Kalla *et al.* found an extension to the abdomen [7]. Vade and Nolan [8] described a case having the involvement of the esophagus. Recurrent respiratory distress with cyanosis has been reported by Eroglu *et al.* [9]. In our case, the presentation was recurrent respiratory tract infection and respiratory distress.

Neurogenic tumor accounts for 19–39% of the posterior mediastinal tumors. Enteric duplication cysts, pulmonary sequestration, lymphatic cysts, and lymphoma may occur in the posterior mediastinum and should be differentiated from teratoma. Posterior mediastinal teratoma is a rare entity and pre-operative clinical diagnosis is difficult and can only be suspected after radiological investigations [10].

X-ray chest is the initial radiological investigation in all cases of respiratory distress and may detect fluid levels, calcification, teeth, and bones in a case of teratoma [8,11] though no such evidence was found in X-ray chest in our case. Thoracic CT scan is the mainstay of evaluation of mediastinal masses, detected incidentally by radiography or by clinical presentation, but CT results are frequently indeterminate. MRI provides more information because of its superior tissue characterization and delineation. All cases of respiratory distress do not require MRI, but only those with mediastinal masses detected incidentally on X-ray or clinically having a recurrence of symptoms should undergo MRI for proper evaluation. As posterior mediastinal mass tends to involve adjacent structures such as aorta, esophagus, heart, pericardium, or chest wall, esophagram and angiogram are recommended along with thoracic MRI [8,11]. In our case, MRI confirmed the posterior mediastinal origin of the mass, provided additional information about its content, and revealed a relationship with surrounding structures. The esophagoscopy helped us in planning surgery.

Posterior mediastinal teratomas are managed by radical resection. The mass may be approached through either left or right thoracotomy, depending on its location. Complete excision is sometimes difficult because of dense adhesions to adjacent intrathoracic vital structures [8,11-13]. With early and complete removal, the prognosis is excellent. Those presenting with

unstable hemodynamics, cyanosis, and acute respiratory failure may require emergency surgery [12].

The common post-operative complications reported are Horner's syndrome, gastroesophageal reflux, and chylothorax [13]. However, we did not come across these complications in the present case. Histopathology is always confirmatory and differentiates mature and immature elements of teratoma. Serum AFP and beta-HCG are good indicators of prognosis and should be monitored, particularly in immature teratoma [14]. In our case, we did not encounter any problem during surgical removal and in the post-operative period. Histopathology confirmed the diagnosis of mature cystic teratoma and both serum AFP and beta-HCG were normal. The baby was on regular follow-up for 1 year and was doing well.

CONCLUSION

Posterior mediastinal teratoma can present with recurrent respiratory distress and infections in infancy, and the cases of suspected pneumonia not improving after an adequate course of IV antibiotics should be investigated further. Early diagnosis and prompt surgical intervention of such cases should be undertaken to reduce morbidity and mortality.

REFERENCES

1. Pattnaik MK, Majhi PC, Nayak AK, Senapati D. A rare presentation of a huge mature mediastinal teratoma with right lung cavitation. *BMJ Case Rep* 2014;2014:2014203835.
2. Dhond AP, Agrawal SO, Sirmukaddam SV, Srinath S, Roplekar P, Desai PR. Mediastinal teratoma: A case report with review of literature. *J Sci Soc*

- 2016;43:57-9.
3. Anand S, Longia S, Agarwal N, Maheshwari M, Apte A, Pathak K. Mature mediastinal teratoma-a rare cause of recurrent respiratory distress. *People's J Sci Res* 2010;3:33-5.
4. Jaiswal R, Rani P, Devenraj V. Asymptomatic posterior mediastinal teratoma diagnosed incidentally. *BMJ Case Rep* 2014;2014:bcr2013203228.
5. Sarin YK, Sengar M, Sinha A. Infected mature teratoma of lower posterior mediastinum. *Indian J Pediatr* 2006;73:369-70.
6. Roshanzamir F, Ghoroubi J, Mirhashemi A, Tabari AK, Rouzrokh M, Izadi M, *et al.* Teratoma in infants and children. *Iran J Pediatr Surg* 2015;1:76-9.
7. El Kalla S, Abdul-Hameed M, Al Ashbal A, Menon N. Posterior mediastinal teratoma with abdominal extension. *Thorax* 1990;45:773-4.
8. Vade A, Nolan J. Posterior mediastinal teratoma involving the esophagus. *Gastrointest Radiol* 1989;14:106-8.
9. Eroglu A, Kurkuoglu IC, Karaoglanolu N, Erdogan F, Gorguner M. Posterior mediastinal mature cystic teratoma. *Am J Case Rep* 2002;3:242-4.
10. Ogata S, Okusa Y, Ogawa T, Ogawa T, Inoue K, Ozeki Y. Mature cystic teratoma in the posterior mediastinum. *Can J Surg* 2009;52:E73-4.
11. Moeller KH, Rosado-de-Christenson ML, Templeton PA. Mediastinal mature teratoma Imaging features. *Am J Roentgenol* 1997;169:985-90.
12. Stajeric M, Dizdarevic I, Kronic I, Topic V. Mediastinal teratoma presenting with respiratory distress and cardiogenic shock in a neonate. *Interact Cardiovasc Thorac Surg* 2020;30:788-9.
13. Sayasathid J, Samboona N, Thopmaogkol S, Buddharadsa Y, Sukonpan K. Mediastinal teratoma in a neonate with acute respiratory failure. *Asian Biomed* 2011;5:123-9.
14. Upadhya M, Sajwany MJ, Tomas-Smiqura E. Recurrent immature mediastinal teratoma with life threatening respiratory distress. *Eur J Pediatr Surg* 2003;13:403-6.

Funding: None; Conflicts of Interest: None Stated.

How to cite this article: Beura S, Mishra BN, Joshi RK, Pahi PP, Sadhukhan M. Posterior mediastinal benign cystic teratoma in an infant – a rare cause of recurrent respiratory distress. *Indian J Child Health*. 2020; 7(6):277-279.

Doi: 10.32677/IJCH.2020.v07.i06.009