

often associated with typical fibrosis, and a red-green birefringence on polarized light microscopy of Congo-red stained tissue.⁴ Information on prognosis and treatment of amyloid in the lung is taken from testimonial reports. To date, no fully effective treatment is available.⁴ Nodular disease may progress slowly, with lesions increasing in size and number.¹ Some patients have an excellent prognosis,⁴ while in others, disease progresses until death from respiratory failure.¹

Although primary pulmonary amyloidosis is a very rare disorder, a definitive diagnosis may mean that the patient can be correctly monitored and unnecessary interventions and treatments can be avoided.

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Cecília Pacheco,* João Cruz, Lurdes Ferreira

Departamento de Neumología, Hospital de Braga, Braga, Portugal

* Corresponding author.

E-mail addresses: ceciliafpacheco@gmail.com (C. Pacheco), joaoffcruz@gmail.com (J. Cruz), lurdesferreira1@gmail.com (L. Ferreira).

Recurrent Tracheoesophageal Fistula in Children With Repaired Esophageal Atresia and the Usefulness of Flexible Bronchoscopy[☆]

Fístula traqueoesofágica recurrente en niños con atresia esofágica reparada y utilidad de la broncoscopia flexible

To the Editor:

Diagnosing recurrent tracheoesophageal fistula (TEF) in a child with congenital esophageal atresia repair during the neonatal period who has recurrent or persistent respiratory symptoms is a real challenge, since medium leakage in the gastrointestinal tract may be missed on standard contrast studies.^{1,2} We present a case that illustrates the importance of high-pressure administration of contrast medium in the esophagus via a catheter, for visualization of recurrent TEF, and the additional utility of flexible bronchoscopy for diagnosis.

A 3 ½-year-old boy was evaluated for recurrent lower respiratory tract infections. He had a history of congenital esophageal atresia with distal TEF repair in the neonatal period. At the age of 2 ½ years, the child was hospitalized for complicated pneumonia with pneumothorax and pneumomediastinum, requiring admission to the intensive care unit and chest tube placement. Chest CT showed diffuse ground glass opacities and areas of atelectasis in

both lungs. In view of the seriousness of the clinical signs, recurrent TEF was considered in the differential diagnosis. The patient had a history of anastomotic leakage, a known risk factor for fistula recurrence,³ yet there were no signs of contrast medium leak or esophageal stenosis in the gastroduodenal tract.

Nonetheless, the patient was admitted on 4 occasions in the following year for fever, cough and shortness of breath. Evaluation by a pediatric pulmonologist was requested by the attending physician. Chest auscultation revealed crepitations and wheezing. Chest X-rays showed areas of atelectasis in both lungs and mild gastroesophageal reflux was detected on pH-metry. The most important finding on flexible bronchoscopy was a pouch in the posterior tracheal wall, with air bubbling during bag valve mask ventilation administered by the anesthesiologist, suggesting a communication between the esophagus and the pouch (Fig. 1 and Supplementary video 1). Lipid-laden macrophages were found in bronchoalveolar lavage (BAL) fluid and *Pseudomonas aeruginosa* was isolated from BAL culture. Both of these data suggest chronic aspiration and lower respiratory tract infection. Since no diagnostic procedures had been performed in the gastroduodenal tract, contrast medium was injected via a catheter into the esophagus, confirming concurrent TEF (Fig. 1 and Supplementary video 2).²

Approximately 50% of children with history of esophageal atresia and TEF repair have recurrent or persistent respiratory

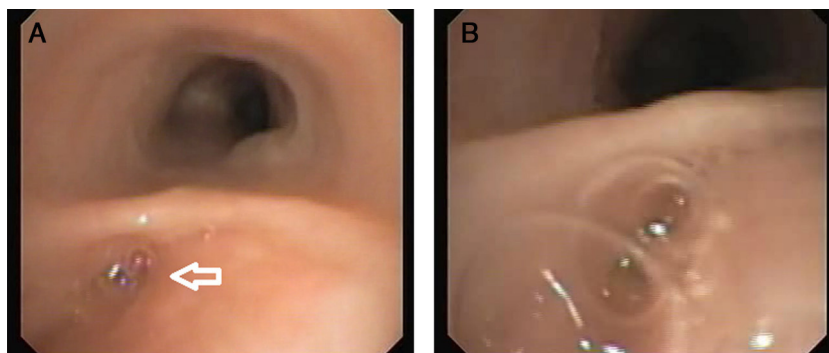


Fig. 1. (A) A tracheal pouch in the posterior tracheal wall was observed using flexible bronchoscopy (white arrow), near the carina. (B) Air bubbling was then seen during bag valve mask ventilation administered by the anesthesiologist, suggesting a communication between the esophagus and the pouch.

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symptoms due to bronchial hyperreactivity, tracheomalacia or aspiration due to esophageal dyskinesia, esophageal stenosis, gastroesophageal reflux or relapse of repaired TEF (in up to 10% of cases of esophageal atresia).^{3–5} Recurrence of the fistula in the gastrointestinal tract may be difficult to identify, so injection of contrast medium in the esophagus is the next diagnostic step.² Under cine-fluoroscopic guidance and with the patient prone, contrast medium is injected under pressure via a nasogastric tube placed in the stomach that is slowly withdrawn to the esophagus.

The identification of lipid-laden macrophages in the cytological examination of BAL fluid obtained by flexible bronchoscopy is a sign that aspiration may have occurred. Intubation of the fistula with the bronchoscope or injection of methylene blue with esophagoscopy are additional methods that can provide important information for the diagnosis of recurrent TEF.²

Conflict of Interest

None of the authors has any conflict of interests to declare.

Appendix A. Supplementary data

Supplementary data associated with this article can be found, in the online version, at <http://dx.doi.org/10.1016/j.arbr.2014.11.015>.

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Antonios Platnaris,^a Dimitra Lianou,^b Athanasios G. Kaditis^{a,*}

^a Pediatric Pulmonology Unit, First University Department of Pediatrics, University of Athens School of Medicine and Aghia Sophia Children's Hospital, Atenas, Greece

^b First Department of Pediatrics, National Health System, Aghia Sophia Children's Hospital, Atenas, Greece

* Corresponding author.

E-mail address: kaditia@hotmail.com (A.G. Kaditis).

Posterior Mediastinal Ganglioneuroma[☆]

Ganglioneuroma mediastínico posterior

To the Editor,

Ganglioneuromas are benign neurogenic tumors that can invade or adhere strongly to the mediastinal structures, producing varying clinical manifestations.¹ We present a case of brachial neuralgia due to compression of the brachial plexus by a mediastinal ganglioneuroma.

A previously healthy 39-year-old woman was hospitalized for a 3-month history of brachial neuralgia (BN) in the left arm. Physical examination was normal. Left laterotracheal opacity was detected on chest X-ray (Fig. 1A). A well-defined tumor in the left paravertebral region, 6 cm in diameter, with some areas of intravenous contrast medium uptake was found on chest computed tomography (CT) (Fig. 1B) and magnetic resonance imaging (MRI) (Fig. 1C). The mass was not compressing the spinal cord and there was no direct invasion of the vertebrae. Laboratory test results and bronchoscopy were completely normal. The apical mass and part of the sympathetic chain were extracted via a left posterolateral thoracotomy (Fig. 1D). The mass showed strong adhesions to the brachial plexus and the stellate ganglion, with widening of the intervertebral space. The pathology report confirmed the diagnosis of ganglioneuroma, in view of the mature ganglion cells contained in a stroma of collagen fibers with the presence of some Schwann cells. In the post-surgical period, the patient presented transient Claude Bernard–Horner syndrome that resolved fully

after 2 months. Clinical examination and CT at 3 and 6 months of follow-up were favorable.

Ganglioneuroma, originating in the neural crest cells, is the most mature form of neuroblastoma. It occurs most frequently in the mediastinum (70%)^{2,3} and age at presentation is generally young.⁴ Most cases are asymptomatic. In the series of Takeda, 67% of cases were asymptomatic, while only 1.4% had neurological symptoms and 2 cases of paresthesia of the arm. Some symptoms are related with the secretion of hormones, such as vasoactive intestinal peptide, causing diarrhea, or catecholamines that produce adrenergic symptoms (hypertension, palpitations or sweating), but our patient did not present any symptoms of this kind.⁵

On CT and MRI, ganglioneuroma is seen as a solid, well defined, encapsulated mass. Around 10% of neurogenic paravertebral tumors have an extension into the vertebral canal and thus are called 'dumbbell tumors', as was the case in our patient.

Spinal cord invasion by the tumor can be more precisely determined on MRI, so this evaluation must be performed before surgery in patients with suspected neurogenic tumor in order to rule out any intramedullary extension. Surgical resection is the standard treatment. The transmanubrial approach provides an excellent surgical field, with control of the subclavian vessels, the brachial plexus and exposure of the upper cervical and dorsal spine. Cardillo et al.¹ concluded that video-assisted surgery (VATS) was associated with lower morbidity and a shorter post-surgery hospital stay. The possibility of using VATS depends on tumor size and intramedullary extension. In our case, we decided to perform a thoracotomy, since surgery of the thoracic apex carries a greater risk of vascular or nervous injury, and also because greater control of the intraspinal extension of the tumor was required. Our case is unusual, in that the ganglioneuroma was exceptionally large and the first symptom leading to diagnosis was brachial neuralgia.

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