

Thoracoabdominal Duplication Symptomatic on Both Sides

A Case Report

S Moralioglu¹, OZ Pektaş¹, AC Celayir¹, O Bosnali¹, E Kaygusuz²

ABSTRACT

Thoracoabdominal foregut duplications account for less than 2% of all gastrointestinal duplications. Here, we report a case of thoracoabdominal duplication cyst in an eight-month old boy who presented with both respiratory and abdominal complaints. Excision of both thoracic and abdominal extensions of the thoracoabdominal duplications in the same session is an appropriate and safe option to prevent possible complications.

Keywords: Child, cyst, duplication, thoracoabdominal

Duplicación Tóraco-abdominal Sintomática en Ambos Lados

Un Reporte de Caso

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RESUMEN

La duplicación intestinal tóraco-abdominal representa menos del 2% de todas las duplicaciones gastrointestinales. Aquí reportamos el caso de un quiste de duplicación tóraco-abdominal en un niño de ocho meses que presentaba síntomas respiratorios y abdominales. La extirpación de las extensiones tanto torácicas como abdominales de las duplicaciones tóraco-abdominales en la misma sesión es una opción apropiada y segura para evitar posibles complicaciones.

Palabras claves: Niño, quiste, duplicación tóraco-abdominal

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INTRODUCTION

Gastrointestinal system duplications are uncommon congenital malformations. Thoracoabdominal foregut duplications account for less than 2% of all gastrointestinal duplications. Thoracoabdominal duplications are intrathoracic extensions of duplications that originate from the gastrointestinal system. Approximately 60% of the thoracoabdominal duplications communicate with the gastrointestinal tract, most often the jejunum and duodenum (1, 2). Symptoms and signs may be related to the thoracic or abdominal extension of the cyst. Here, we report a case of thoracoabdominal duplication that presented with both respiratory and abdominal complaints.

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CASE REPORT

An eight-month old boy was admitted to hospital with respiratory distress. Clinical history revealed intermittent colicky abdominal pain and hospitalization due to bronchiolitis five months ago. Respiratory sounds were decreased in the right hemithorax, and abdominal examination was normal. The posterior-anterior and lateral chest X-rays showed opacity on the right posterior mediastinum (Fig. 1). Computed tomography (CT) scan of the chest showed a cystic lesion (6 x 6 x 8 cm) in the right posterior mediastinum extending to the abdomen through the left lobe of the liver and lesser curvature of the stomach. Magnetic resonance imaging (MRI) results also supported these findings and revealed that there was no connection between cyst and spinal canal (Fig. 2). The patient experienced increased colicky abdominal pain while having antibiotic, steroid and bronchodilator therapy for bronchiolitis treatment and was hospitalized to our clinic. Feeding was stopped. Routine laboratory investigations and

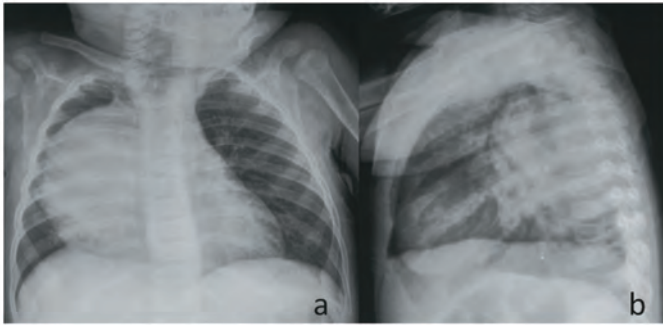


Fig. 1: (a) The posterior-anterior and (b) lateral chest X-rays show opacity on the right posterior mediastinum.



Fig. 2: Magnetic resonance imaging shows a cystic lesion (6 x 6 x 8 cm) in the right posterior mediastinum extending to the abdomen through the left lobe of the liver and lesser curvature of the stomach.

tumour markers were normal except for increased neuron-specific enolase (28 ng/mL; normal value 0–15 ng/mL).

In rigid oesophagoscopy, there was no luminal connection between the oesophagus and the mass, but indentation was seen in the middle 1/3 region of the oesophagus. Right-side thoracotomy revealed a posterior mediastinal cyst with its cranial ending attached to the second thoracic vertebrae, adjacent to the oesophagus without attachment, and extending to the abdomen through the diaphragm. The thoracic part of the cyst was excised. The abdominal part of the cyst which was noted to be perforated and adjacent to the lesser curvature of the stomach was resected during the operation by utilizing an upper midline



Fig. 3: Chest X-ray shows that the mass image in the thorax was decreased after treatment.

incision. Abdominal extension measured 3 x 2 x 4 cm and was not connected to the gastrointestinal tract. Chest X-ray showed that the mass image in the thorax was decreased (Fig. 3) and plain abdominal radiography was normal. The post-operative course was uneventful, and he was discharged on the sixth postoperative day. The pathological diagnosis of the lesion revealed a duplication cyst containing gastric and intestinal mucosa (Fig. 4).

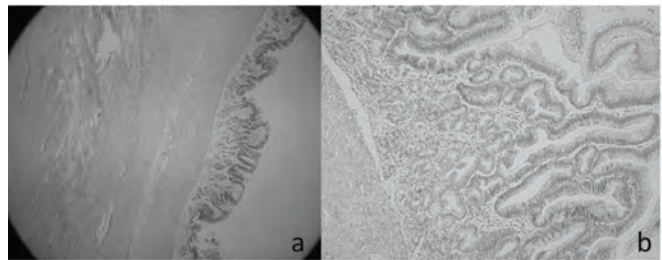


Fig. 4: (a) Mucosa is lined by intestinal-type epithelium and is composed of gastric-type glands (H&E stain x 100). (b) The cystic wall is similar to the intestinal wall, composed of mucosa, submucosa and muscularis propria (H&E stain x 10).

DISCUSSION

Multiple theories have been proposed regarding the aetiology of duplications. To date, no single unifying theory for their development has been described and no hypothesis has explained all types of malformations. All findings about aetiology of alimentary tract duplications suggest a multifactorial process for development and possibly different mechanisms depending on location (3).

Thoracoabdominal duplications are rare forms of the alimentary tract duplications; they arise in the abdomen and cross behind the oesophagus and aorta after passing through the right crus of the diaphragm. Thoracoabdominal duplications pass through the diaphragm through their own hiatus.

They tend to be long, often large, tubular lesions that are located in the posterior mediastinum, to the right of the midline (1). Generally, they are closed cranially. Pokorny and Goldstein (2) described one patient with communication to the cervical oesophagus. Approximately 60% of duplications communicate with the intestine below the diaphragm, most often to the normal duodenum or jejunum. The remainder end blindly in the abdomen. In our case, both ends of the duplication were blind.

The symptoms and signs may be related to the thoracic or abdominal extension of the duplication. Dyspnoea, severe respiratory distress or similar respiratory complaints are the most common symptoms in neonates. Almost half of the thoracoabdominal duplications are found during the neonatal period and 80% within the first year of life (2). Chest pain, abdominal pain, gastrointestinal bleeding and anaemia are common symptoms in older children. Macpherson (1) described that ectopic gastric mucosa was found in 29% of thoracoabdominal duplications. In the patients with ectopic gastric mucosa, gastrointestinal bleeding, anaemia and even perforation can occur as in the index case.

On the posterior-anterior and lateral chest radiographs, they appear as well-defined posterior mediastinal shadows. Also, thoracic vertebral anomalies can be seen on the X-rays. In the literature, the two largest series of thoracoabdominal duplications report an 88% incidence of vertebral anomalies in these patients (4, 5). Computed tomography or MRI is used to diagnose, as well as to demonstrate the relationship between duplication and adjacent organ, and evaluate for possible communication to the spinal canal (2). Tc-99m pertechnetate scintigraphy may demonstrate ectopic gastric mucosa within it (1). There was no vertebral anomaly or spinal canal communication in our case; however, neuron-specific enolase value was elevated.

Treatment of thoracoabdominal duplications is more difficult than isolated thoracic or abdominal cystic duplications. In cases with spinal involvement, a combined surgical approach with an experienced neurosurgical team should be employed (4). Complete excision of the thoracoabdominal duplications is necessary. Generally, it takes two discontinuous incisions and it may be performed in one or two stages. The portion of the cyst causing symptoms is usually excised first. If the abdominal portion

is excised first, thoracic procedure should be completed soon after first operation (6). It is important to note that following excision of the abdominal portion, the thoracic extension may increase in size due to lack of drainage. On the other hand, if the thoracic portion is excised first, abdominal extension may become complicated, especially in the presence of ectopic gastric mucosa. In our case, the abdominal part of the duplication became symptomatic soon after presentation of the thoracic part.

Serious complications related to the treatment of thoracoabdominal duplications have been reported, including incomplete excision of the thoracic or abdominal extension of the cyst, missed tracts from the chest to the abdomen, meningitis, haemo/pneumothorax, intestinal fistula and ileal necrosis after excision of the abdominal part of the duplications (5–7). There was no complication in our case during or after operation.

Symptoms and signs of thoracoabdominal duplications depend on the size of the thoracic and abdominal extensions, type of the contained ectopic mucosa, and the relationship between the cyst and adjacent organ. Excision of both thoracic and abdominal extensions of the thoracoabdominal duplications in the same session is an appropriate and safe option to prevent possible complications.

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