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Case Report

An embryological outpouching of esophagus in the right thoracic cavity: a rare cadaveric case report

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ABSTRACT

The esophagus (EG) is a muscular tube, that connects the pharynx to the stomach. Stomach is the widest part of the alimentary tract and is located between EG and duodenum. Gatro-intestinal tract duplications are spherical or tubular prolongations extending from the oral cavity to the anal aperture. During the anatomical dissection for the medical students in a 70 years old male cadaver, a rare variation of esophageal dilatation (ED) was found in the posterior mediastinum. On removal of the anterior thoracic wall, EG showed an abnormal outpouching at the level of T2. ED passes distally through the oesophageal opening of the diaphragm and continues as the cardiac end of the stomach into the abdominal cavity. This is directly supplied by the branches of the descending thoracic aorta. Stomach appears to be normal and there were no associated anomalies in the rest of the regions. Abdominal duplications may remains asymptomatic and gets identified incidentally during routine investigations. Gastroenterologist, radiologists and thoracic surgeons should have a profound knowledge on these developmental variations.

Keywords: ED, Stomach, Embryological anomalies, Gastric mucosa

INTRODUCTION

The esophagus (EG) is a muscular tube, that connects the pharynx to the stomach. EG measures 25 cm in length for the adults, with a diameter of 1-2 cm. The EG divides into three parts: cervical, thoracic and abdominal part. Abdominal part of the EG measures 1-2.5 cm in length, that runs along the left aspect of the midline. It passes through the EG aperture of diaphragm, at the T10 vertebral level and ends at the cardiac orifice of the stomach.[1]

Stomach is the widest part of the alimentary tract and is located between EG and duodenum. It occupies the corresponding quadrants of the abdomen and is wedged between the upper abdominal viscera on either sides.[1]

In 1950, Ladd and Gross, defined the gastro intestinal tract (GIT) duplications as "spherical or tubular prolongations, extending from the oral cavity to the anal aperture, and maintaining the contact with the normal GIT". It can be communicative/blind end and histological possess a typical digestive mucosa. Arbona quotes Blasius (1711), stating that, incidence of esophageal cyst are unusual developmental anomalies of GIT.[2,3]

By the 4th week of embryological development, the respiratory bud protrudes from the ventral wall of the foregut. Development of the tracheo esophageal septum divides the foregut into ventral respiratory primordium and a dorsal EG. Simultaneously, in the primitive thoracic region, the stomach develops as a fusiform dilatation, adjacent to the respiratory diverticulum.[4]

Congenital hernias result due to herniation of abdominal contents through the diaphragm into thoracic cavity. Frequent sites of hernia can be found in posterolateral, oesophageal/ subcostosternal regions.[1]

Our case report shows a rare congenital variation of ED in the right thoracic region with the presence of a normal esophageal opening of the diaphragm and stomach in the abdominal region.

CASE REPORT

During routine dissection of a male cadaver aged 70 years, for teaching the medical students, in department of anatomy in our institution, we came across, a rare variation of ED located in the posterior mediastinum. A standard guideline was followed during dissection, with Cunningham's dissection manual of anatomy.[5]

Observations in the thoracic cavity

On removal of the anterior thoracic wall, the heart was observed to be in its anatomical position. EG showed an abnormal outpouching at the level of T2, extending into the right pulmonary space and size of the right lung appears to be small (Figure 1 A). On comparison with the normal left lung, the dimensions of the right lung appeared to be compromised (Figure 1 B). Bilateral lung dimensions were measured (Figure 1 C and D) and tabulated (Table 1).

Figure 1 (a-e): Right and left lung dimensions.

Displays the right and left lung. Mediastinal surface of right and left lung with measuring tool. Costosternal surface of right and left lung with measuring tool.

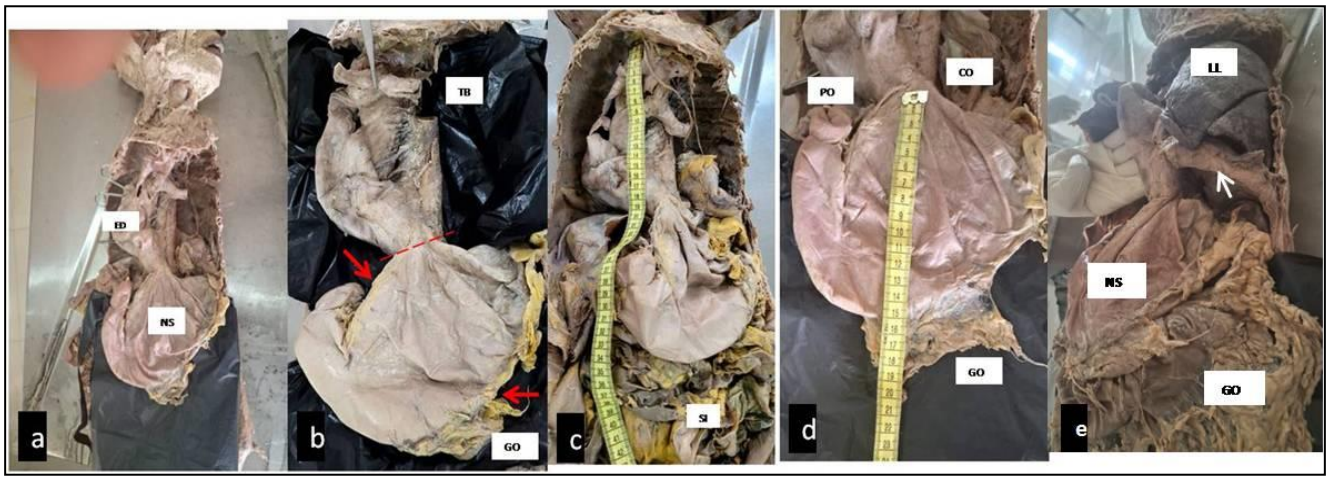

Figure 2 (a-e): Esophageal dilatation in right thoracic cavity and stomach in the abdominal region.

Displays esophageal dilatation (ED) in the right thoracic cavity and normal stomach (NS) in left upper quadrant of the abdomen. Tracheal bifurcation (TB) at level of T4 vertebral level greater omentum (GO), lesser and greater curvature (red solid arrows) and esophageal opening of the diaphragm (dotted lines). The total length of esophagus and stomach are measured in centimetres using measuring tape, small intestine (SI) is seen. Cardiac orifice (CO) and pyloric orifice (PO) are shown. Extent of stomach from cardiac orifice to maximum convexity of greater omentum is measured. LL-left lung resting on left dome of diaphragm (white solid arrows) and greater omentum (GO) from greater curvature of stomach.

On the right side of thoracic cavity, ED was found to be a continuation of the cervical part of the EG at the level of T2 (Figure 2 A). Tracheal bifurcation was at the level of T4 and the maximum convexity was appreciated below T4 (Figure 2 B).

ED showed maximum convexity at T6 vertebral level. Reduction in the diameter was observed as it descends towards the diaphragmatic opening at T10 level (Figure 2 B). ED dimensions were measured at different thoracic vertebral level and tabulated (Table 2).

ED passes distally through the oesophageal opening of the diaphragm and continues as the cardiac orifice of the stomach into the abdominal cavity (Figure 2 D). Along with the ED gastric nerves, esophageal branches of the left gastric vessels run through the esophageal opening.

On observation, ED is directly supplied by the branches of the descending thoracic aorta (DTA) (Figure 3 D) and also contributed by branches from posterior intercostal arteries from the dorsal side of ED. We also observed multiple arterial and venous ramification around ED.

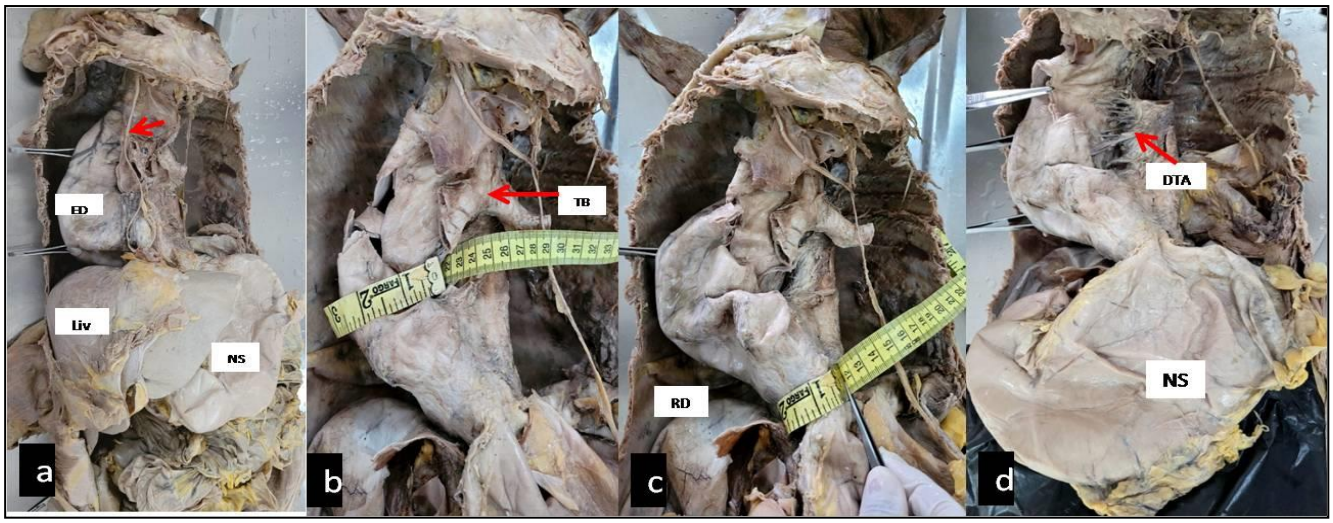

Figure 3 (a-d): Measurements of esophageal dilatation and its blood supply.

Esophageal dilatation (ED), Liver (Liv), Normal stomach (NS), Right phrenic nerve (red solid arrows). Tracheal bifurcation (TB) (red solid arrows), measurement of maximum convexity of ED. Right dome of diaphragm (RD) is shown and circumferential diameter of abdominal esophagus measurements are done. Descending thoracic aorta (DTA) and its branches supplying ED (red arrow) and normal stomach (NS).

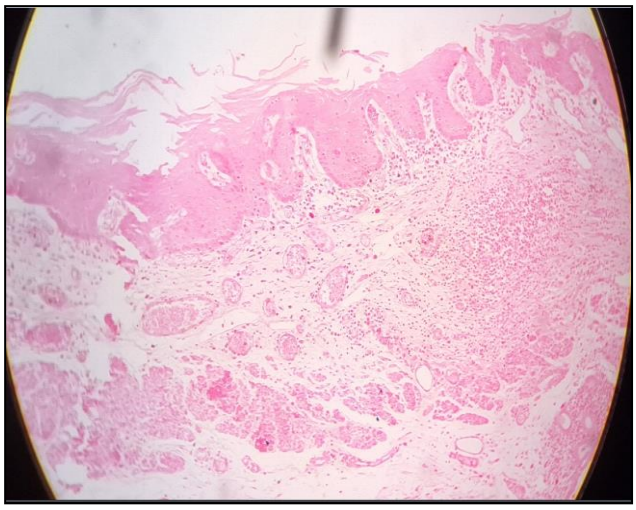

Figure 4: Histological image of ED in 10× resolution.

The mucosa was lined by stratified squamous non keratinised epithelium, lamina propria, muscularis mucosa with longitudinal muscle layer, submucous connective tissue with neurovascular bundles, circular and longitudinal muscle layers in the muscularis externa and adventitia.

The arch of azygos vein arches over the proximal part of ED in the superior mediastinum. The right phrenic nerve was passing in front of ED (Figure 3 A) and the right vagus nerve was passing on the right lateral aspect of ED. A longitudinal incision was made in the ED and exploration of the interior of ED revealed a hollow smooth cavity within. A small tissue section of ED was taken and subjected to histological examination.

Histological examination revealed, ED had the characteristic features that strongly resembled EG (Figure 4). The mucosa was lined by stratified squamous non keratinised epithelium, lamina propria, muscularis mucosa with longitudinal muscle layer, submucous connective tissue with neurovascular bundles, circular and longitudinal muscle layers in the muscularis externa and adventitia.

Observations in the abdominal cavity

On opening abdominal cavity, cardiac orifice and pyloric orifice appeared to be normal (Figure 2 D). Greater and lesser omentum was arising from greater and lesser curvature of stomach (Figure 2 B). All abdominal organs

located in their respective abdominal quadrants (Figure 2 and 3). Dimensions of stomach were measured and tabulated (Table 3). There were no associated congenital anomalies evident in this cadaver other than ED.

Table 1: Dimensions of right and left lung.

Parameters	Right lung (cm)	Left lung (cm)
Maximum height	16	20
Maximum width	22	21
Circumferential diameter (at max convexity)	46	40 (at cardiac notch)

Table 2: Measurements of ED into right side of thoracic region.

Variables	Width (cm)	Circumferential diameter (cm)
Vertebral levels		
C6	2.5	4.5
T2	6	12
T4	7.5	15
T6	10.5	21.5
T10	5.5	11
Length (cm)		
Total length of dilated part of EG from T2-diaphragmatic opening of EG	20	-
Total length of dilated part of EG from T4-diaphragmatic opening of EG	16	-
Normal EG length and diameter	25	3

Table 3: Measurements of stomach in abdominal region.

Parameters	Measurements (cm)	
	Length	
From cardiac end to max inferior border of greater curvature	18	-
	Width	**Circumference**
Maximum convexity of stomach-from lesser curvature to greater curvature	14.5	29
Cardiac end below diaphragmatic orifice		10

DISCUSSION

GIT duplications report for 2% of all the congenital anomalies with 60 % established communications. These duplications signify a developmental abnormality in notochord with vertebral malformations.[7,8] Alimentary tract duplications were found in 1 in 4500 live births. In Bremer's research, duplication occurs during the 6th-8th week of intra uterine life (IUL). ED are aberrations of the primitive foregut development.[9-11]

In American population incidence of ED was 1 in 8000 live births. ED is epithelial lined outpouching of the primitive gut. On examination it appears as cystic or tubular and they may communicate or non-communicable with EG lumen. ED appears as fluid filled structures attached to the EG or respiratory tree. It usually located in the right posterior inferior mediastinum.[11] Similar to findings of Casillas et al our findings showed, ED as a luminal dilatation enlarged from EG along the right posterio-inferior mediastinum.

Retrograde pouching of abdominal contents into the thoracic cavity was first reported by Valle and white in 1946. Oesophageal herniation can occur due to defect in the oesophageal opening of the diaphragm.[12] Our case findings also showed male predominance, similar to the studies done by Pokorny et al, Mazziotti et al and Sonoda et al.[13-15]

Our histological findings proved; the ED was lined by esophageal mucosa. Langston et al reported that on histological examination the esophageal duplications were lined by respiratory mucosa in case 1 and alimentary tract mucosa in case 2.[16] Histological findings of Johnston et al and Borrie et al, are gastric mucosa made of non ciliated columnar epithelium with chief and parietal cells with muscular layers mimicking the alimentary tract mucosa.[17,18]

Valle and white coined the term enteric cyst, for the duplications lined by ED. Lewis et al reviewed the literature in past 43 years.[12] He justified Bremer theory of intra-abdominal duplications. Out of 39 cases, 23 were of gastric mucosa, 3 was glandular epithelium, 3 were found to be columnar epithelium and 2 as atrophic epithelium. The lining mucosa was damaged in 3 cases and not disclosed in 5 cases.[19]

Radiological investigations like gastrointestinal contrast study and computerised tomographic scan of the thoracic cage helps in providing a differential diagnosis for ED. After clinical diagnosis, thoracoscopic surgery is recommended for an excellent outcome.[20,21]

CONCLUSION

Our case report will aid during diagnostic GIT instrumentation to avoid iatrogenic complications. According to the diverticular theory, these dilatations may project freely into the pleural or the peritoneal cavity. Abdominal duplications may remain asymptomatic and gets identified incidentally during routine investigations. Gastroenterologist, radiologists

and thoracic surgeons should have a profound knowledge on these developmental variations.

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