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BRIEF REPORT

Spontaneous self-expulsion of an oesophageal duplication cyst

Nara Lee,* Sanjeev Sabharwal,† Derrick Tee,‡ Deepak Dhatrak* and Andrew Ruszkiewicz*,§,¶

*Department of Anatomical Pathology, Royal Adelaide Hospital, SA Pathology, †Paragon Medical Centre, Solitaire Medical Group, ‡Department of Gastroenterology and Hepatology, Lyell McEwin Hospital, §School of Medicine, The University of Adelaide and ¶School of Pharmacy and Medical Science, University of South Australia, Adelaide, South Australia, Australia

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Correspondence
Andrew Ruszkiewicz, Department of Anatomical Pathology, Royal Adelaide Hospital, SA Pathology, Adelaide, SA, Australia.
Email: andrzej.ruszkiewicz@adelaide.edu.au

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Introduction

Oesophageal duplication cysts (ODCs) are a type of congenital foregut cyst and are the second most common duplication of the gastrointestinal tract, after those in the jejunum and ileum, accounting for 10%–15% of duplications. ODCs tend to be found incidentally in adults. When symptomatic, the most common presentations in adults are dysphagia and chest pain.[1] Cough is a presenting symptom that is implicated in only a minority of cases. Although rare, it is prudent to consider ODC in patients with chronic cough with no evident respiratory cause.

Case report

We present a case of a 65-year-old woman who was otherwise fit and well but presented to her general practitioner with a lump of tissue that she had "coughed out". She had a longstanding history of severe dry chronic cough, previously investigated with both chest and sinus CT scans, bronchoscopy and an infective screen with no abnormalities identified. The tissue was sent for histological evaluation.

The specimen consisted of two irregular grey brown portions of tissue. One appeared to be a sac-like structure measuring 25 × 15 × 10 mm with a thin wall and containing loose debris within the lumen. The other structure was thin and tubular, measuring 40 × 8 × 5 mm and resembling a stalk (Fig. 1a).

Microscopic evaluation revealed a cystic structure lined variably by specialised gastric-type mucosa with oxyntic-type glands, squamous epithelium, and colonic-type mucosa supported by thin muscularis mucosae and attenuated, double-layered smooth muscle containing myenteric and submucosal plexus structures resembling the muscularis propria of the normal gastrointestinal tract (Fig. 1b–e). No respiratory-type epithelial lining was seen, and there was no cartilage or seromucinous glands identified within the cyst wall. The cyst lumen contained degenerate vegetable matter, as well as numerous Enterobius vermicularis (pinworm) organisms, their eggs and fungal hyphae. The tubular stalk-like structure was lined by colonic-type mucosa with a thin underlying layer of smooth muscle. The appearances were consistent with an enteric-type oesophageal duplication cyst, with the cyst contents indicating a connection of the cyst lumen to the true lumen of the oesophagus.

Subsequently, the patient was referred for an upper gastrointestinal endoscopy and endoscopic ultrasound (EUS). This was performed several weeks following the expulsion of the cyst. No subepithelial lesions or mucosal defects suggestive of residual duplication cyst were identified; however, this was not unexpected given

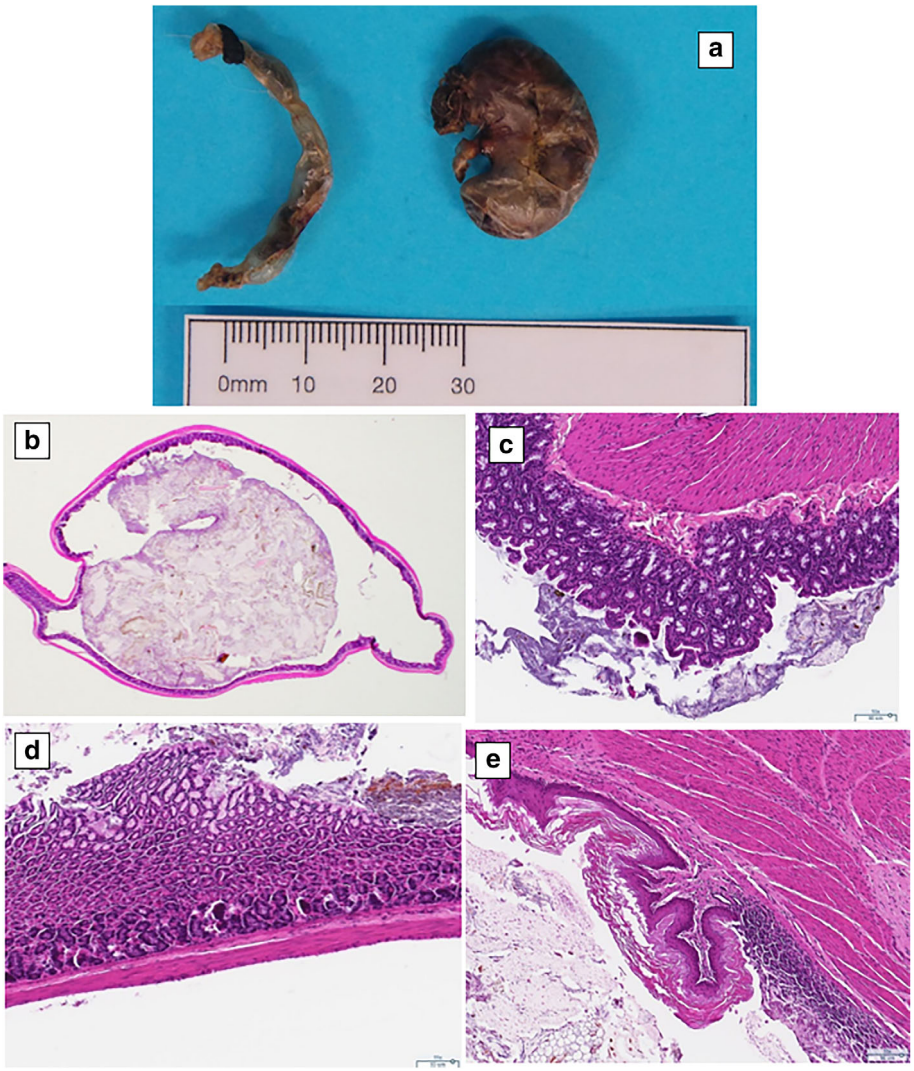

Figure 1 (a) Macroscopic images of the received tissue; a cystic structure measuring 25 mm and a stalk-like structure measuring 40 mm in length. (b–e) Microscopic images of the oesophageal duplication cyst. Despite significant delay in fixation, the structures were preserved enough for adequate histological examination. (b) Cystic structure lined by colonic-type mucosa with a double-layered smooth muscle wall (H&E, 20×). (c) Colonic mucosal lining with a layer of smooth muscle within the wall (H&E, 100×). (d) Specialised gastric mucosal lining with a layer of smooth muscle within the wall (H&E, 100×). (e) Squamous epithelial lining with a double layer of smooth muscle within the wall (H&E, 100×).

the small size of the stalk of the cyst and the interval time between expulsion and endoscopy allowing adequate time for healing. In addition, the previous CT images were reviewed, with no suggestion of any oesophageal pathology. At the time of this procedure, the patient reported complete resolution of her cough.

Discussion

Oesophageal duplication cysts (ODCs) are most commonly found in the lower third of the oesophagus and are generally mediastinal in location. The majority (75%) are diagnosed in early childhood, with presenting symptoms including dysphagia and nausea and vomiting, relating to compression of the oesophageal wall and respiratory distress, caused by compression of the respiratory tract structures. They can be associated with other congenital abnormalities including pulmonary bronchogenic cysts, neuroenteric cysts, vertebral defects, and annular and heterotopic pancreas.

ODCs are defined by the location within the oesophagus or attachment to the oesophageal wall, a double layer of smooth muscle within the wall and an epithelial lining compatible with those found in the embryonic oesophagus or a squamous epithelial lining.[2] ODCs may contain other heterotopic gastrointestinal epithelia including gastric and colonic mucosa. They can be distinguished from bronchogenic cysts by the lack of intramural cartilage or seromucinous glands.

Our case is unique, and to the best of our knowledge, this is the only case of self-expelled ODC that has been reported in the literature. Although a connection to the esophagus was not able to be demonstrated on imaging or endoscopic findings, the

presence of food matter and pinworm organisms within the lumen of the cyst is consistent with a direct connection to the lumen of the true gastrointestinal tract, an uncommon finding previously noted in only two reported cases of ODC in the literature.[3] In particular, the polypoid configuration of the cyst within the oesophageal lumen is unusual. We presume based on the symptom of cough and the self-expulsion that the duplication cyst was likely within the upper esophagus.

Surgical excision is the treatment of choice of most symptomatic ODCs; however, management of asymptomatic cases is more problematic as even biopsy of these lesions via EUS may result in the development of serious acute intrathoracic inflammation.[4] Despite the associated risks, endoscopic intervention may be necessary in asymptomatic cases when there is clinical suspicion of more ominous lesions including rare malignant transformation.[5]

Acknowledgment

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Ethics statement

This case report was prepared in accordance with the ethical standards as outlined in the Helsinki Declaration (as revised in 2013). Written informed consent was obtained for publication of this article, including clinical images.

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