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Case report

Heterotopic salivary gland tissue in the gastro-oesophageal junction: a rare entity

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Abstract

Introduction: Gastric and pancreatic heterotopia are the two most common types of heterotopias seen in the gastrointestinal tract (GIT). These are the two most common types of heterotopias seen in the oesophagus as well. The occurrence of heterotopic salivary gland tissue (HSGT) in the GIT is rare, and most reported cases are colorectal and anal lesions with only a few cases involving the oesophagus.

Case report: A 69-year-old man presented with a seven-month history of gastro-oesophageal reflux disease (GORD). The upper gastrointestinal endoscopy showed a moderate hiatus hernia with a pale pink mucosal patch close to the gastro-oesophageal junction. The biopsy showed oesophageal tissue with morphological changes of mild reflux oesophagitis. The submucosal tissue contained organized glandular structures with mucinous glands and intercalated ducts resembling mature salivary gland tissue. There was no evidence of Barrett oesophagus, dysplasia or neoplasm.

Discussion and conclusion: HSGT is most commonly seen in the head and neck region of the body, and its occurrence in the GIT is rare. Occurrence of oesophageal HSGT is extremely rare and most cases have presented with symptoms of GORD. Although HSGT is usually benign and a rare phenomenon, clinicians and pathologists need to be aware of this condition to prevent a delay in diagnosis and inappropriate management.

Keywords : heterotopia, salivary gland tissue, oesophagus

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Introduction

Heterotopia is the presence of histologically normal, mature tissue at a site where it does not usually reside and where it lacks anatomical and vascular connections with the parent organ. When this heterotopic tissue forms a mass, it is known as a choristoma. The exact mechanism of heterotopia is not clearly understood, and embryonic maldevelopment is a suggested hypothesis (1). The most commonly reported types of heterotopias in the GIT are gastric and pancreatic heterotopias, and these are the two most common types of heterotopias seen in the oesophagus as well.

The occurrence of heterotopic salivary gland tissue (HSGT) in the GIT is rare and only a few cases were identified in the English literature (2). Most of these were colorectal and anal lesions. Only six cases were from the oesophagus and the gastro-oesophageal junction (GOJ) was the site of occurrence in five of these cases (2–4). We report a case of HSGT in GOJ in a patient who presented with gastro-oesophageal reflux disease (GORD), to highlight the importance of the awareness of this rare entity by clinicians and pathologists.

Case report

A 69-year-old man presented with a 7-month history of GORD. The upper gastrointestinal endoscopy showed a moderate hiatus hernia with a pale pink mucosal patch close to the GOJ (Figure 1).

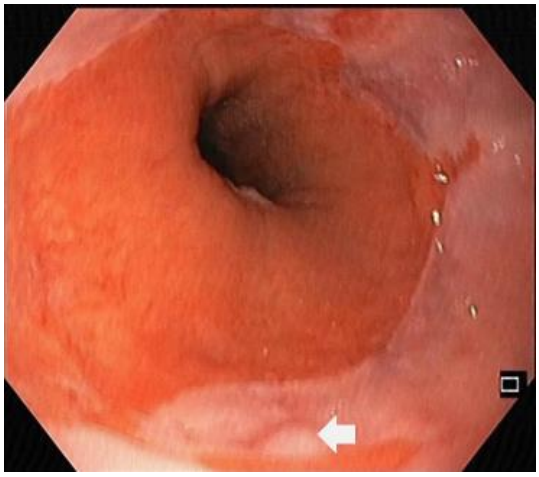

Figure 1. Endoscopic image showing a pale pink mucosal patch (white arrow)

Microscopy of the GOJ biopsies showed three mucosal tissue fragments, two of which showed stratified squamous epithelium with the morphological changes of mild reflux oesophagitis. The third fragment was non-specialized gastric mucosal tissue that was histologically unremarkable. There were organized glandular structures with mucinous glands and intercalated ducts, morphologically resembling mature salivary gland tissue, in the submucosa of one of these fragments. A mild diffuse chronic inflammatory cell infiltrate was identified within this tissue (Figure 2). These

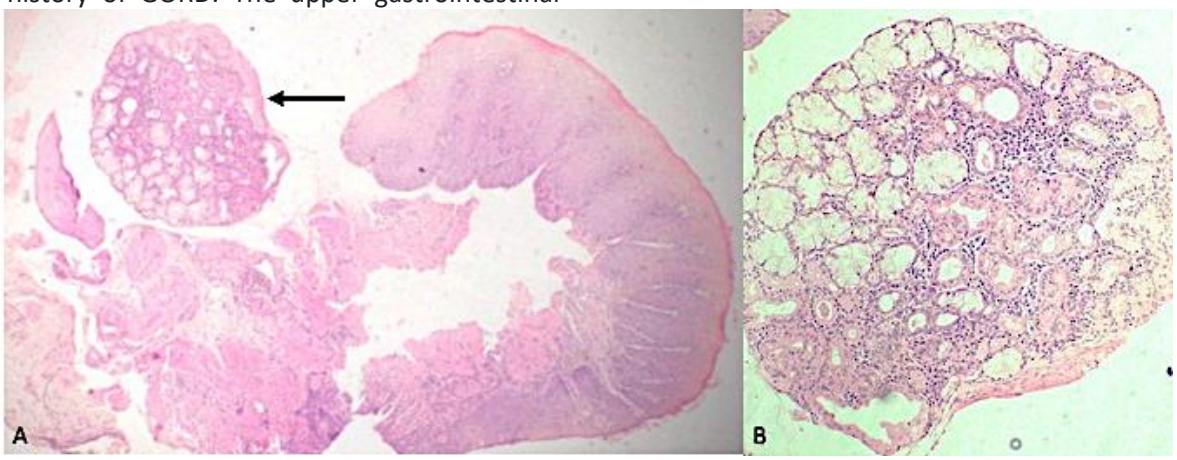

Figure 2. **A** GOJ biopsy showing an island of salivary gland type tissue. The stratified squamous epithelium shows morphological changes of reflux oesophagitis. (H&E x40) **B.** Salivary type tissue with mucinous glands and intercalated ducts. There is an infiltrate of chronic inflammatory cells around the glandular tissue. (H&Ex100)

features were in keeping with those of HSGT in GOJ. There was no evidence of Barrett oesophagus, dysplasia or a neoplasm.

The patient was reassured, and his symptoms were treated conservatively with proton pump inhibitors to which he responded well.

Discussion

HSGT is most commonly seen in the head and neck region of the body and its occurrence in the GIT is extremely rare (3). To the best of our knowledge, there are only 6 cases of HSGT in the oesophagus, reported in the English literature. (2–5). Four of these were choristomas at the GOJ, where heterotopic tissue was arranged into a tumour-like mass. In the other two, the endoscopy had shown a salmon-coloured mucosal patch just proximal to the GOJ and a hiatus hernia in one of the patients, and a segment suspicious of Barrett oesophagus and ulceration at GOJ in the other patient (2).

In five out of six patients, the presenting symptom was GORD and the fifth patient was on surveillance for Barrett oesophagus. Our patient also presented with GORD. In GORD, there is repeated exposure of the oesophageal mucosa to the gastric contents. Reflux of acid triggers protective mechanisms in the oesophagus. A decrease in pH associated with GORD causes an increase in the rate of mucin release from the salivary glands, oesophageal mucosal and submucosal glands into the mucus-bicarbonate layer (6,7). Intestinal columnar metaplasia at the GOJ, which occurs secondary to GORD, is a well-known phenomenon. However, the exact causative relationship between HSGT/ salivary metaplasia and GORD is obscure and needs further study.

HSGT is found to occur mainly in adults with one case reported in a child of five years (2). The incidence is more common in males than in females. The index case is also an adult male. At endoscopy, HSGT is seen as a pink mucosal patch, or a subserosal nodule or pedunculated mass, ranging from 1-2 cm in size, if it is a choristoma. Our patient had a hiatus hernia on endoscopy with a pale pink mucosal patch at the GOJ but no mass formation. Microscopically, HSGT is mostly seen as mixed serous and mucinous glands with salivary ducts (2). It may be entirely composed of mucinous glands in some of the cases, as in this index case (2).

Oesophageal submucosal glands are similar in morphology to salivary glands. However, these are not visualized in standard endoscopy and are not typically sampled by endoscopic biopsies (8). All reported cases of HSGT in the oesophagus, including our case, have shown a mucosal change on endoscopy. Salivary gland choristomas form a mass lesion and could raise the endoscopic differential diagnosis of gastrointestinal stromal tumour (GIST), schwannoma, leiomyoma, ectopic pancreas, lipoma, and carcinoid.

Although malignant transformation is a known occurrence in heterotopic salivary tissue in the head and neck region, there is only a single case reported in the GIT (2,3).

Conclusion

HSGT is extremely rare in the oesophagus and is usually harmless. However, clincians and pathologists need to be aware of this condition so that they will consider the possibility of this condition when dealing with similar cases. This will prevent delay in diagnosis and inappropriate management.

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