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Letters to the Editor

Intestinal-type adenocarcinoma arising from gastric heterotopia of the oral tongue

Upon reading the case report by Guo et al.[1] from our institution, describing a primary intestinal-type adenocarcinoma of the tongue, we felt compelled to point out that this is a rare case of intestinal-type adenocarcinoma arising from gastric heterotopia (GH).

Histological examination of the surgical specimen revealed a moderately differentiated adenocarcinoma morphologically resembling colorectal adenocarcinoma. Heterotopic gastric mucosa of oxyntic type was found adjacent to the adenocarcinoma component. This component was lined mainly by gastric foveolar epithelium with focal areas of normal stratified squamous epithelium. The gastric glands beneath the surface contained both parietal and chief cells (Fig. 1). These glands were organized in a branching and lobular fashion separated by distorted smooth muscle bundles, and showed a downward growth pattern. Glandular dilatations with cystically dilated glands were noted, especially in the deeper part. Notably, a focal area demonstrating a transition from intestinal metaplasia to dysplasia to adenocarcinoma component was identified after extensive sampling (Fig. 2). Immunohistochemically, the carcinoma cells were diffusely positive for CK20, CDX2, and SATB2, partially positive for CK7, and focally positive for MUC2 and MUC5AC.

GH, also called heterotopic gastric mucosa, is a benign condition, generally thought to be congenital in nature. GH has been described at various levels of the gastrointestinal tract, most commonly in the oesophagus and duodenum[2]. Its occurrence in the head and neck region is very rare, but has been reported in the tongue, floor of the mouth, oropharynx, hypopharynx, nasopharynx, and larynx[3]. The anterior aspects of the tongue and the floor of the mouth are the most commonly affected oral sites[3]. It is mostly discovered in infants and young children, but can remain undetected or untreated into adulthood. It usually presents as an asymptomatic solid mass. Although exceedingly rare, malignant transformation in GH has been described in the oesophagus, duodenum, jejunum, ileum, colon, and rectum, with most reported cases being adenocarcinomas[2]. To the best of our knowledge, there has been no previous report of a case of adenocarcinoma arising from GH in the head and neck region.

In this case, the adenocarcinoma component showed an intestinal immunophenotype (CK20, CDX2, and SATB2-positive), as well as typical morphology of intestinal-type adenocarcinoma. Adenocarcinomas with an intestinal phenotype have been described in a wide variety of anatomical sites. Intestinal-type adenocarcinoma of the head and neck is a group of uncommon neoplasms that mainly affect the sinonasal tract. Primary intestinal-type adenocarcinoma of the tongue and oral cavity (PIATOC) is a newly emerging entity, with only 10 cases (including the case presented here) reported in the literature to date[4]. However, the histogenetic nature of this neoplasm is controversial. There are two theories about the origin of PIATOC: (1) metaplasia of minor salivary duct epithelium, and (2) transformation of pre-existing embryonic endodermal epithelial rests. Most authors have favoured or suggested a minor salivary gland origin, although without obvious evidence of derivation from mature minor salivary gland epithelium[4]. Only recently, Berg et al.[5] presented for the first time a case of PIATOC with histological evidence of metaplasia within the salivary gland epithelium. On the other hand, two previously published cases have demonstrated unequivocal origin of PIATOC from teratoid cyst or foregut duplication cyst containing gastric and intestinal-type mucosa[4]. In addition, Ligthelm et al.[6] recently reported a primary mixed adenoneuroendocrine carcinoma of the tongue arising within a foregut duplication cyst, also supporting an origin from entrapped endodermal elements. Our case provides further evidence to sustain a derivation of PIATOC from a pre-existing structure, demonstrating for the first time a PIATOC arising from GH of the tongue. An awareness of the rare occurrence of primary adenocarcinoma from GH may help avoid misdiagnosis, overstaging, and an unnecessarily extensive clinical workup.

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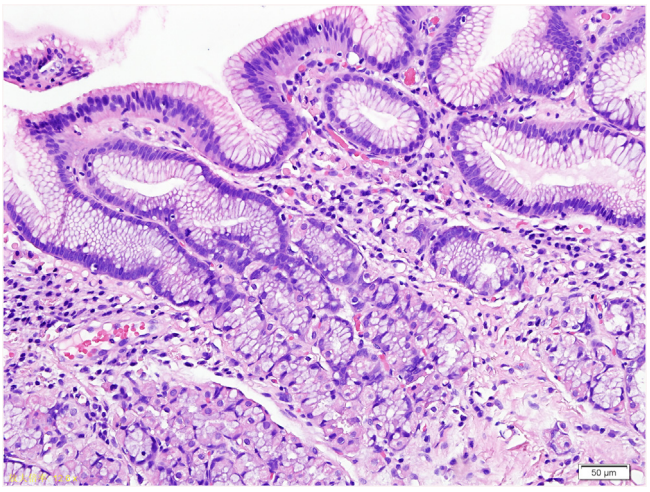

Fig. 1. High-power image showing the surface mucinous foveolar epithelium and underlying glands containing parietal and chief cells (original magnification, 200×; haematoxylin–eosin stain).

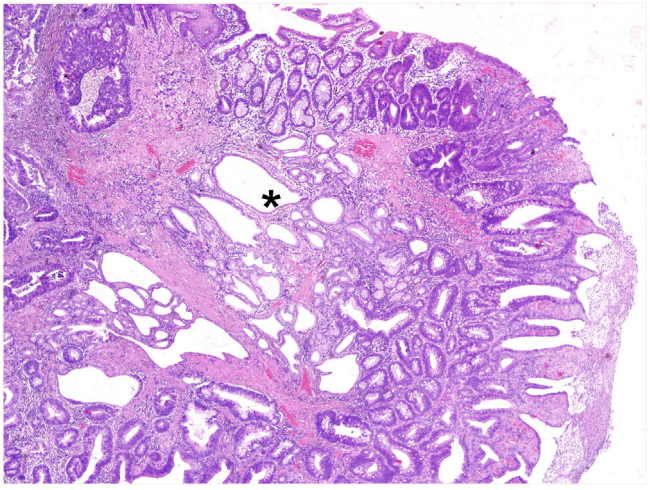

Fig. 2. Low-power image showing the heterotopic gastric mucosa with continuous progression from intestinal metaplasia to epithelial dysplasia to adenocarcinoma (original magnification, 40×; haematoxylin–eosin stain). Note the remnants of heterotopic gastric mucosa (*).

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None.

Ethical approval

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Patient consent

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Response to ‘‘Intestinal-type adenocarcinoma arising from gastric heterotopia of the oral tongue’’

We are pleased that our recently reported case of primary intestinal-type adenocarcinoma of the tongue aroused the interest of other pathologists at our institution[1]. Our article focused on the clinical characteristics and therapeutic methods applied in our patient’s case, as well as those applied in other cases of intestinal-type adenocarcinoma (ITAC) reported previously in the literature.

As stated in our article, several theories concerning the histological origin of benign gastrointestinal glandular epithelium at the base of the tongue have been offered. One theoretical source is salivary gland epithelium that undergoes metaplasia[2]. The theory that we favour – and that appears to be supported by immunohistochemistry in our patient’s case – is that the lingual glandular tissue is choristomatous as a result of its entrapment at the base