Esophageal Xanthoma – Report of Two New Cases and Review of the Literature

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Abstract

Background: Esophageal xanthoma is a very rare lesion which can be incidentally discovered during endoscopy. Only eleven cases have been reported, including ours. Case reports: We present two new cases of esophageal xanthoma localized in the lower esophagus in a 56-year-old woman and a 62-year-old man. Endoscopically, esophageal xanthoma appears as yellowish granular spots or a slightly elevated lesion. Microscopically, it consists of fat accumulation in foamy histiocytes beneath the squamous epithelium. Conclusions: The clinical and pathological importance of these lesions and what they mean in patients is discussed, along with a review of the literature.

Key words


Introduction

Xanthomas are rare, smooth, yellowish tumor–like lesions which can be incidentally discovered in the upper gastrointestinal tract during gastrointestinal endoscopy [1]. Considering their benign nature and the endoscopic resemblance to ectopic sebaceous glands, xanthomas have not been reported frequently in the literature.

The reported incidence in the upper gastrointestinal tract varies among endoscopy series, the most frequent location being the stomach [1], followed by the duodenum and esophagus. Xanthomas can develop as solitary or multiple nodules or plaques with a diameter of less than 10 mm, usually less than 5 mm [1-5].

Esophageal xanthomas, like all upper gastrointestinal tract xanthomas, are asymptomatic, the patients being usually investigated for other conditions. Histologically, the lesions appear as clusters of large foamy xanthoma cells within the connective tissue in the lamina propria [1, 6]. In the esophageal mucosa only nine cases have been described in the literature, prior to our two cases, occurring in a 62 year old man and a 56 year old woman.

A review of the published literature on esophageal xanthoma is presented; a search was conducted to find all articles on esophageal xanthoma, from 1984 to 2010. Medical subject headings used to search the PubMed database (National Institutes of Health, United States) included upper gastrointestinal xantomatosis, as well as a keyword search using “esophageal xanthoma”, “esophageal xanthelasma”, “lipid islands in the esophagus”. A total number of 12 articles were originally identified. Limitation of the findings to human series and English language reduced the number of publications to 7.

Case reports

Case 1

The patient was a 56-year-old woman presenting with pain in the upper abdomen, upper abdominal fullness, accompanied by bloating, nausea and heartburn. The physical examination revealed only mild epigastric tenderness. An endoscopic examination was performed, which revealed a 4 mm sessile polyp close to the gastroesophageal junction. There were no other abnormalities in the stomach or esophagus, and a polypectomy was accomplished.

Case 2

The patient was a 62-year-old man. His past medical history was significant for Biermer anemia, chronic viral C hepatitis and a polyp in the gastric antrum. He did not complain of any gastrointestinal symptoms. He had a gastrointestinal endoscopy for evaluation of Biermer anemia and control of the previously diagnosed polyp in the gastric antrum. In addition, a small esophageal lesion was identified in the lower esophagus, which measured 3 mm in dimension.

Gastric biopsies revealed an antral hyperplastic polyp with focal adenocarcinoma, severe atrophic fundic gastritis and extensive intestinal metaplasia – compatible with type A gastritis.
Pathological examination

The biopsy specimens obtained from the esophageal mucosal polyp in each case were stained with hematoxylin and eosin (H&E), periodic acid-Schiff (PAS), and Alcian blue, and were evaluated by the pathologist.

Histologically, on H&E stained sections, both esophageal lesions were composed of medium to large islets of foamy macrophages in the lamina propria, under the squamous epithelium, with small nuclei, centrally or eccentrically located (Fig. 1). The covering squamous epithelium did not show any atypical cells, and in the first case showed some degree of parakeratosis. The cells were negative for PAS and Alcian blue reaction.

Immunohistochemical tests were performed on formalin-fixed paraffin embedded sections.

The immunohistochemical staining revealed the same findings in both cases. The cells were strongly positive for CD68 (Figs. 2, 4), and no reactivity was present for cytokeratin/AE1/AE3 (Fig. 3), S-100 protein and CD1a.

Discussion

Esophageal xanthoma is a benign asymptomatic lesion, always diagnosed by histological examination. The most common location within the esophagus seems to be distal. Endoscopically, esophageal xanthoma appears as yellowish granular spots or a slightly elevated lesion, measuring 2 to 10 mm. Microscopically, it consists of foamy histiocytes containing lipids, located beneath the squamous epithelium [1-3, 6]. They may be solitary or multiple.

The first reported case occurred in the upper esophagus and was defined as “lipid islands” in 1984 by Remmele and Engelsing [7]. Since 1984 to present, only 11 cases (the present ones included) have been reported [7-14], summarized in Table I.

The patients diagnosed with esophageal xanthoma were 6 males and 3 females (two of them not specified), with an average age of 54 years. The reported incidence of xanthoma in the upper gastrointestinal tract for endoscopy series was 0.23%, 12% of the cases occurring in the esophagus. Most of the lesions occurred in the lower esophagus, two in the middle and one in the upper (two of them not specified). The size of the xanthomas varied between 2 and 10 mm. Eight cases appeared as a solitary lesion. Endoscopic findings showed yellow-white, granular nodules or slightly elevated lesions, and one sessile appearance was reported.

There is no report of malignant esophageal xanthoma. The etiopathogeny and the significance of esophageal xanthoma remain unknown, and in contrast to cutaneous xanthoma, no correlation between esophageal xanthoma and hyperlipemia has been shown. The association of inflammatory cells suggests a response to a focal mucosal damage [15-17].

This theory would explain why gastric xanthomas appear to be more frequent than esophageal xanthomas, as traumatism and inflammation may be better tolerated by esophageal squamous epithelium than by gastric columnar epithelium.
Some studies also suggested a disturbance of carbohydrate or fat metabolism involved in the pathogenesis of gastrointestinal xanthomas. But there is no evidence yet indicating this kind of pathogenesis for esophageal xanthoma.

Conclusions

Esophageal xanthoma seems to be a very rare lesion, as only eleven cases, included our two cases, are reported in the literature. It has to be distinguished on endoscopy from carcinoid tumor, granular cell tumour, and ectopic sebaceous glands, and histologically a differential diagnosis of signet ring cell carcinoma must be performed, as in other locations in the gastrointestinal tract.

Acknowledgement

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References


Table I. Esophageal xanthomas in the literature from 1984 to present

<table>
<thead>
<tr>
<th>Authors/year</th>
<th>Age/sex</th>
<th>Site</th>
<th>Number</th>
<th>Size (mm)</th>
<th>Endoscopic findings</th>
<th>Other conditions</th>
</tr>
</thead>
<tbody>
<tr>
<td>1 Remmele and Engelsing (1984)</td>
<td>54/M</td>
<td>Upper</td>
<td>Solitary</td>
<td>10</td>
<td>Yellow spot</td>
<td>Gastrectomy</td>
</tr>
<tr>
<td>2 Stole and Seifert (1985)</td>
<td>45/M</td>
<td>Middle</td>
<td>Three</td>
<td>Not specified</td>
<td>Yellow flat elevation</td>
<td>Hyperlipidemia, diabetes mellitus</td>
</tr>
<tr>
<td>3 Vimala et al (2000)</td>
<td>37/F</td>
<td>Lower</td>
<td>Multiple</td>
<td>2-5</td>
<td>Yellowish nodular (lymphoma)</td>
<td>Gastric xanthoma</td>
</tr>
<tr>
<td>5 Hirokawa et al (2003)</td>
<td>67/M</td>
<td>Lower</td>
<td>Solitary</td>
<td>2</td>
<td>Yellow spots (sebaceous gland)</td>
<td>Hepatocellular carcinoma, hypertension, cholelithiasis</td>
</tr>
<tr>
<td>7 Gencosmanoglu et al (2004)</td>
<td>Not specified</td>
<td>Not specified</td>
<td>Multiple</td>
<td>Less than 5 mm in diameter</td>
<td>Yellow-white colored plaques</td>
<td>Not specified</td>
</tr>
<tr>
<td>8 Gencosmanoglu et al (2004)</td>
<td>Not specified</td>
<td>Not specified</td>
<td>Solitary</td>
<td>Less than 5 mm in diameter</td>
<td>Yellow-white colored plaques</td>
<td>Not specified</td>
</tr>
<tr>
<td>9 Dong-Sik Cho et al (2008)</td>
<td>49/M</td>
<td>Lower</td>
<td>Solitary</td>
<td>3</td>
<td>Yellowish elevated granular lesion</td>
<td>Atrophic gastritis</td>
</tr>
<tr>
<td>10 Becheanu et al (2011)</td>
<td>62/M</td>
<td>Lower</td>
<td>Solitary</td>
<td>3</td>
<td>Yellowish elevated lesion</td>
<td>Biermer anemia, HCV, antral hyperplastic polyp with focal adenocarcinoma, atrophic gastritis</td>
</tr>
<tr>
<td>11 Becheanu et al (2011)</td>
<td>56/F</td>
<td>Lower</td>
<td>Solitary</td>
<td>4</td>
<td>Sessile polyp</td>
<td>-</td>
</tr>
</tbody>
</table>