Case Reports in Gastroenterology Case Rep Gastroenterol 2022;16:94–96

DOI: 10.1159/000522184 Received: November 29, 2021 Accepted: December 24, 2021 Published online: February 28, 2022 © 2022 The Author(s). Published by S. Karger AG, Basel www.karger.com/crg

OPEN ACCESS

This article is licensed under the Creative Commons Attribution-NonCommercial 4.0 International License (CC BY-NC) (http://www.karger.com/Services/OpenAccessLicense). Usage and distribution for commercial purposes requires written permission.

Letter to the Editor

Epidermoid Metaplasia in Diffuse Esophageal Intramural Pseudodiverticulosis

Florian Hentschel^a Christian Hirschmann^b Stefan Lüth^a

^aDepartment of Gastroenterology, Brandenburg Medical School (Theodor Fontane), Brandenburg, Germany; ^bDepartment of Pathology, Brandenburg Medical School (Theodor Fontane), Brandenburg, Germany

Dear Editor,

Diffuse esophageal intramural pseudodiverticulosis (DEIPD) is a rare disease. It is characterized by chronic fibrosing inflammation of the esophagus and the enlargement of its intramural glands that lead to the name-giving pseudodiverticula. Clinical symptoms include dysphagia and food impactions; endoscopic signs are multiple small diverticula openings, "frosted glass look," "faux uni pattern," and "trachealization" of the esophagus [1]. Risk factors for DEIPD are alcohol- and tobacco abuse, but since these are common and the disease is rare, an unknown additional pathomechanism must exist. Little is also known about the histopathologic signs of DEIPD. Often, if at all, they are reported just as "unspecific inflammation" [2, 3].

Now in a recent article, Shintaku et al. [4] present a patient suffering from DEIPD whose esophageal mucosal biopsies show unambiguous signs of epidermoid metaplasia (EM, synonymous epidermization). In this context, one of our own works was quoted as: "two cases showed moderate squamous cell hyperplasia and epidermization" [1], and it was criticized that we did not publish pictures of these conditions. First of all, we have to apologize for being unprecise here: in our own collective of 21 patients, we found 1 case of squamous cell hyperplasia and 1 case of epidermization. We have therefore re-examined the endoscopic photographs and histological slides of this 1 patient – a 58-year-old male European with a history of severe alcohol- and tobacco abuse. Results were as following. Compared to the findings of Shintaku et al. [4], the endoscopic/macroscopic aspect in our patient is not as suggestive of EM as theirs (shown in Fig. 1). Our mucosal biopsies, however, show an impressive similarity to theirs at 2.5 years after first visit (compare Fig. 2c, d in [4]): Multiple fine keratohyalin granules in the superficial layers of the epithelium with rather discrete hints of acanthosis and hyperkeratosis, typical for early stages of EM (shown in Fig. 2) [5, 6]. We did not find the acellular keratin layer they found in their follow-up biopsies, but since that layer is characteristic



Correspondence to: Florian Hentschel, f.hentschel@klinikum-brandenburg.de

Case Reports in Gastroenterology

Case Rep Gastroenterol 2022;16:94–96	
DOI: 10.1159/000522184	© 2022 The Author(s). Published by S. Karger AG, Basel
	www.karger.com/crg

Hentschel et al.: Epidermization in Esophageal Pseudodiverticulosis



Fig. 1. DEIPD, endoscopic view: frosted glass look (dull-white swelling of mucosa), discrete signs of trachealization (rigid appearance with reduced peristalsis, multiple rings), longitudinally aligned diverticle openings. Fujinon EG-600ZW, VP-4450HD.



Fig. 2. a–c DEIPD, histologic view of mucosal biopsies: Several fine keratohyalin granules in the superficial layers of the epithelium. Hematoxylin and eosin, ×400.

for later stages of EM, we suspect that, given enough time, it might develop in our patient too. So in general, we can confirm the results of our colleagues.

We have at the moment no explanation why some DEIPD patients develop epidermoid metaplasia and some do not. Still, this would be an interesting question to ask, especially against the background of DEIPD and EM sharing the same risk factors as esophageal squamous cell carcinoma [7–10]. Since our previous study centered on clinical and endoscopic characteristics of DEIPD, histologic data were only collected retrospectively from pathological routine examinations [1]. We are therefore preparing another study re-examining all archived biopsies to specifically look for early signs of epidermization, and we would like to thank Shintaku et al. [4] for pointing us to this topic.

Sincerely yours, Florian Hentschel, Christian Hirschmann, Stefan Lüth.

Statement of Ethics

All procedures reported in this letter and in the original publication were in accordance with the standards of the Institutional and/or National Research Committee and with the 1964 Helsinki Declaration and its later amendments or comparable ethical standards. Written informed consent or substitute for it was obtained from all patients for publication of the details of their medical information. The Ethics Committee's approval number for the follow-up study is E-01-20191028.



Case Rep Gastroenterol 2022;16:94–96
DOI: 10.1159/000522184 © 2022 The Author(s). Published by S. Karger AG, Basel
www.karger.com/crg

Hentschel et al.: Epidermization in Esophageal Pseudodiverticulosis

Conflict of Interest Statement

The authors have no conflicts of interest to declare.

Funding Sources

No funding was received.

Author Contributions

F.H. performed all endoscopies and wrote the letter. S.H. examined the original slides and re-examined the slides in preparation of the letter. S.L. cared for the patient and edited the draft of the letter.

References

- 1 Hentschel F, Lüth S. Clinical and endoscopic characteristics of diffuse esophageal intramural pseudodiverticulosis. Esophagus. 2020;17(4):492–501.
- 2 Medeiros LJ, Doos WG, Balogh K. Esophageal intramural pseudodiverticulosis: a report of two cases with analysis of similar, less extensive changes in "normal" autopsy esophagi. Hum Pathol. 1988;19(8):928–31.
- 3 Wightman AJ, Wright EA. Intramural oesophageal diverticulosis: a correlation of radiological and pathological findings. Br J Radiol. 1974;47(560):496–8.
- 4 Shintaku M, Shintaku M, Torii I. Development of epidermoid metaplasia of the mucosa in association with esophageal intramural pseudodiverticulosis and candidiasis. Case Rep Gastroenterol. 2021 May–Aug;15(2): 709–14.
- 5 Nakanishi Y, Ochiai A, Shimoda T, Yamaguchi H, Tachimori Y, Kato H, et al. Epidermization in the esophageal mucosa: unusual epithelial changes clearly detected by Lugol's staining. Am J Surg Pathol. 1997;21(5):605–9.
- 6 Ezoe Y, Fujii S, Muto M, Ochiai A, Ohtsu A. Epidermoid metaplasia of the esophagus: endoscopic feature and differential diagnosis. Hepatogastroenterology. 2011;58(107–108):809–13.
- 7 Taggart MW, Rashid A, Ross WA, Abraham SC. Oesophageal hyperkeratosis: clinicopathological associations. Histopathology. 2013;63(4):463–73.
- 8 Singhi AD, Arnold CA, Crowder CD, Lam-Himlin DM, Voltaggio L, Montgomery EA. Esophageal leukoplakia or epidermoid metaplasia: a clinicopathological study of 18 patients. Mod Pathol. 2014;27(1):38–43.
- 9 Plavsic BM, Chen MY, Gelfand DW, Drnovsek VH, Williams JP 3rd, Kogutt MS, et al. Intramural pseudodiverticulosis of the esophagus detected on barium esophagograms: increased prevalence in patients with esophageal carcinoma. AJR Am J Roentgenol. 1995;165(6):1381–5.
- 10 Khemani R, Shah P, Shah P. Intramural pseudodiverticulosis in squamous cell carcinoma of the esophagus. Indian J Gastroenterol. 1992;11(1):38.

