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CASE REPORT

Multiple Tiny Ectopic Sebaceous Glands Discovered Throughout Entire Esophageal Tract

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Abstract A 45-year-old woman complaining of abdominal fullness was referred for endoscopic examination. She was a non-smoker and non-drinker. An endoscopic examination revealed the presence of more than 100 tiny, rounded, elevated, yellowish lesions < 0.5 mm in diameter scattered throughout the upper and lower esophagus. Based on the endoscopic examination results, her stomach manifested symptoms of mildly superficial gastritis. Histopathologic examination of the esophagus biopsy specimen revealed that some of the lobules of the cells displayed typical sebaceous differentiation covered by a squamous epithelium. No evidence of inflammatory reaction, hair follicles, or malignancy was found. The patient's blood and serum findings were unremarkable. Our final diagnosis was multiple tiny ectopic sebaceous glands in the esophagus. This is an interesting and rare case of esophageal sebaceous glands distributed throughout the entire esophagus. Because there were no esophageal symptoms or/and eating problems, the patient did not require endoscopic surgery or other treatment. Follow-up examinations were recommended at intervals between 6 months and 1 year. At the 2-year follow-up, an endoscopic examination

revealed no change in the size or the number of the tiny ectopic esophageal sebaceous glands.

Keywords Ectopic sebaceous glands · Endoscopy · Esophagus · Multiple

Introduction

The occurrence of ectopic sebaceous glands is not unique to humans [1, 2] and has also been reported in rats [3, 4]. Ectopic sebaceous glands have been found in various tissues, such as the lips and mouth, external genitalia, parotid glands, eyelashes, and lacrimal caruncle, palms and soles, and tongue [5]. In humans, this lesion is mostly found in the buccal cavity [6], urogenital tract [7, 8], and various organs [9]. Lesions that are located on the lips and in the mouth are referred to as Fordyce's spots or Fordyce's disease [10].

The presence of ectopic sebaceous glands in the esophagus was first reported by De La Pava and Pickren in 1962 [11], but there is very little data on this condition [12, 13]. Only a few cases have been reported between 1995 and 2008 [14–16]. Here, we present an interesting and rare case involving tiny lesions scattered throughout the whole esophagus.

Case Report

A 45-year-old woman complaining of abdominal fullness was referred for endoscopic examination. She was a non-smoker and non-drinker. Her blood and serum test results were within normal limits.

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The endoscopic examination revealed more than 100 tiny, rounded, elevated, yellowish lesions <0.5 mm in diameter scattered throughout the whole esophagus (Fig. 1). Surprisingly, there were more lesions in the lower portion of the esophagus than in the middle portion (Fig. 1b–d), with the lowest number in the upper portion (Fig. 1a). Salt and pepper lesions of the duodenal bulb were also noted. Based on endoscopic findings, we diagnosed the stomach of this patient as manifesting slightly superficial gastritis. Additionally, the test for *Helicobacter pylori* was positive. Histopathologic examination [high magnification; hematoxylin and eosin (H&E) staining] of the esophagus biopsy specimens suggested that there were some lobules of benign sebaceous glands in the subepithelial region (Fig. 2). No evidence of an inflammatory reaction, hair follicles, or malignancy was noted.

Our final diagnosis was multiple tiny ectopic sebaceous glands throughout entire esophagus. Because there were no uncomfortable esophageal symptoms or/and eating discomfort, our patient did not require endoscopic surgery or other therapy. She was asked to return for follow-up observations at intervals of between 6 months and a year. An endoscopic examination 2 years later revealed no changes in the size or number of esophageal sebaceous glands. Her *H. pylori* infection was treated successfully.

Fig. 1 The results of endoscopic examination revealed more than 100 multiple, tiny, rounded, elevated, yellowish lesions <1 mm in size that were widely scattered from the upper esophagus through to the lower esophagus. **a** upper portion, **b** middle portion, **c** lower-mid portion, **d** lower portion

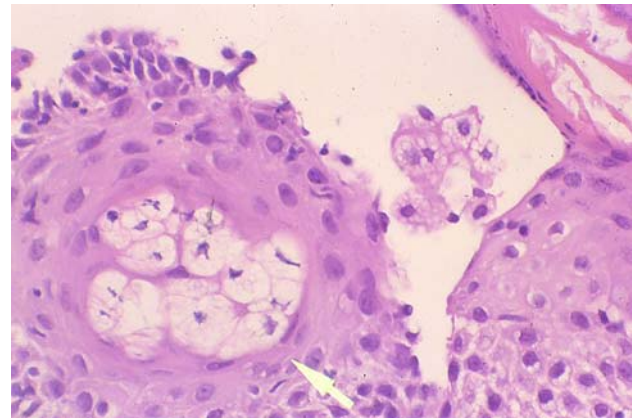
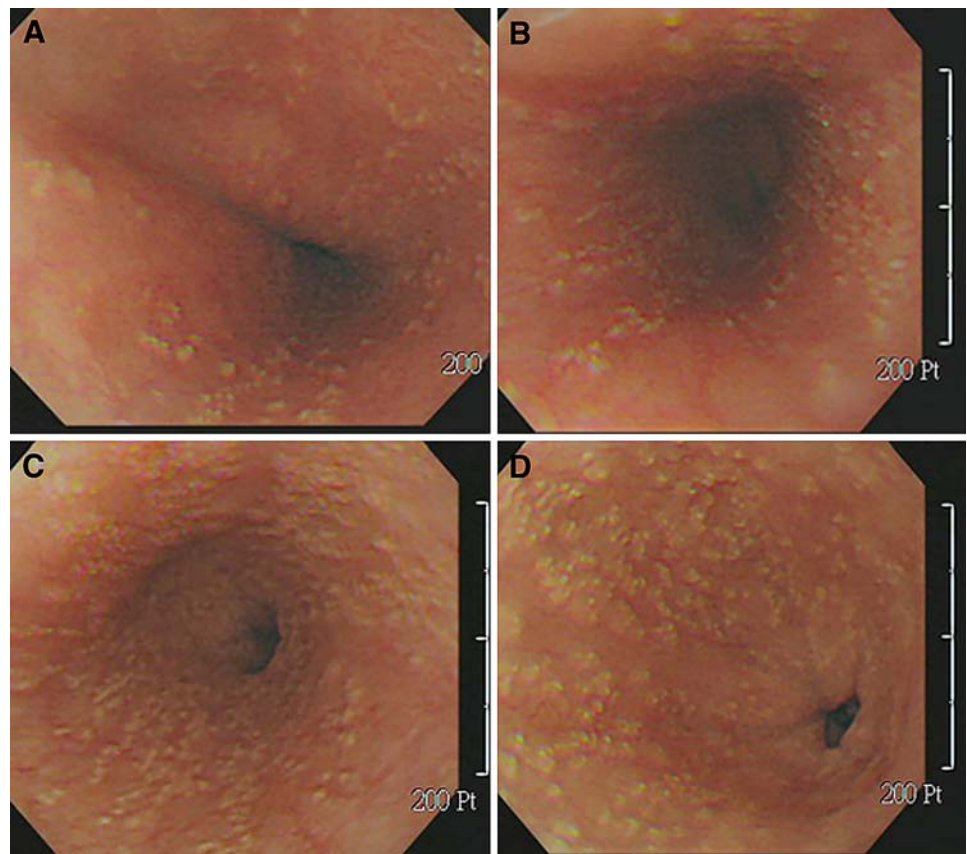


Fig. 2 The results of the histopathologic examination [high magnification ($\times 400$); hematoxylin and eosin stain] of the esophagus biopsy specimen revealed the presence of some lobules of benign sebaceous glands (arrow) covered by squamous epithelium region at high magnification ($\times 400$)

Discussion

Very few cases of ectopic sebaceous glands in the esophagus have been reported in the literature since 2000 [15, 16]. The apparent low incidence of this condition may be due to its lack of obvious clinical signs and symptoms [5, 17]. Most cases have been discovered incidentally by

endoscopy during a referral for alimentary tract examination or at an annual follow-up examination. As a result, it is rarely discovered in the esophagus. Our interesting case was discovered by chance during a routine annual examination. Our patient was a 45-year-old woman who did not smoke or drink. The patient complained of slight abdominal discomfort, but her esophagus was asymptomatic. She was subsequently referred for endoscopic examination. Wei et al. reported that the typical age of patients with ectopic sebaceous glands was nearly 50 years in both men and woman [16]. Our patient, however, was under 50 years of age, suggesting that this condition may be more prevalent in younger patients than has been previously thought.

Hoshika et al. reported on cases with a longitudinal distribution of lesions and postulated that the distribution of sebaceous glands in the esophagus may be meaningful; however, little data supports this suggestion to date [14]. In the case reported here, the lesions were distributed in a scattered pattern. The factors affecting the pattern and distribution of ectopic sebaceous glands in the esophagus require further study. Hoshika et al. [14] further showed that authors have typically reported numbers and sizes of sebaceous glands of <10 and more than 1 mm, respectively. Wei et al. [16] also indicated that the number of esophageal lesions varied from one to more than 100 in a patient, with a range in diameter of between 1 and 20 mm. Several studies have indicated that there are several different shapes of ectopic sebaceous gland lesions, such as the elevated form [16] and lobulated plaque form [17]. In general, plaque forms are larger than elevated ones. However, lesions of the elevated type tend to be more numerous than those with a plaque form. In our case, the lesions had an elevated form. The size and number of lesions found in our patient was < 0.5 mm and more than 100, respectively. In most of the cases reported to date lesions were found predominantly in the upper esophagus [5, 16] or mostly in the mid- and distal esophagus [14, 17]. Our patient is therefore remarkable in that the ectopic sebaceous glands lesions were located along the entire length of the esophageal tract (Fig. 1).

Although the underlying causative mechanisms of this lesion have yet to be clearly elucidated, many theories have been postulated [18]. There are two main hypotheses [16]. The first involves a developmental disorder of the esophagus when it is formed from endodermal tissue by means of an epithelium metaplastic process [19]. Bull et al. [20] postulated that c-Myc expression might be correlated with the enlargement of the sebaceous glands in skin. The findings of recent studies on transgenic mice suggest Wnt morphagenic signals may be associated with epithelial differentiation, and that this association may play a role in the development of lesions [21]. The second hypothesis

involves metaplasia of the sebaceous glands in the esophagus. However, Wei et al. [16] suggest that the latter hypothesis has been largely ignored. Therefore, further research is needed to shed light on mechanisms underlying the development of ectopic sebaceous glands in the esophagus.

Most patients with ectopic sebaceous glands do not need further treatment. Based on the H&E-stained histopathologic results on the biopsy specimen, there was no inflammatory reaction or evidence of malignancy in our case. Additionally, all blood and serum findings were unremarkable. Since there were no esophageal symptoms or/and eating troubles, our patient did not require endoscopic surgery or other therapy. Follow-up examinations were recommended at intervals of between 6 months and 1 year. At the 2-year follow-up, endoscopic examination revealed no change in the size or the number of esophageal sebaceous glands.

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