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Original Research Paper

Pathology

A RARE ENCOUNTER: SEBACEOUS GLAND HETEROTOPIA IN ESOPHAGEAL MUCOSA

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ABSTRACT Ectopic sebaceous glands represent an exceedingly rare finding within the esophageal mucosa. We present the case of a 40-year-old male who presented with a prolonged history of intermittent abdominal discomfort and recurrent emesis spanning two years, compounded by ten days of persistent heartburn. Upper gastrointestinal endoscopy disclosed a remarkable presentation: innumerable, minute, yellowish-white, glistening macules were distributed across the entirety of the esophageal lining. Select lesions were judiciously biopsied. Microscopic examination along with histochemical stains definitively confirmed the presence of sebaceous heterotopia within the esophageal submucosa. The plausible histogenesis of this anatomical anomaly is explored and meticulously discussed within this report.

KEYWORDS : Sebaceous gland heterotopia, Ectopic sebaceous gland, Choristoma, Esophageal mucosa.

INTRODUCTION:

Sebaceous heterotopia, or ectopic sebaceous glands, constitutes an uncommon finding, frequently unveiled incidentally owing to its typical asymptomatic nature. First described by Fordyce in 1896, these adnexal structures are most characteristically observed as Fordyce spots across the vermilion border of the lips, as well as within the salivary glands, buccal mucosa, parotid gland, tongue, larynx, ocular adnexa, external genitalia, uterine cervix, and rarely, the acral surfaces. (1) Early post-mortem investigations suggested a low prevalence, identifying these glands in only 2% of cadaveric specimens. However, the novel advancements in endoscopic technology have facilitated increased detection, with contemporary reports suggesting an incidence of approximately 0.05% in asymptomatic individuals undergoing routine esophageal evaluation. (2,3,4)

Case Presentation:

A 40-year-old male was referred to the gastroenterology outpatient clinic, presenting with two-year history of episodic abdominal pain and, more recently, a ten-day exacerbation of vomiting and intractable heartburn. The routine physical examination and laboratory investigations yielded unequivocal, unremarkable results.

Subsequent upper gastrointestinal endoscopy provided striking visual evidence: a multitude of distinct, yellowish-white, glistening papules were observed to stud the mucosal surface throughout the entire length of the esophagus (Figure 1). Furthermore, a Grade I hiatal laxity of the lower esophageal sphincter (LES) was noted. The gastric fundus, corpus, and antrum displayed evidence of superficial erosions. Multiple lesions from the esophageal mucosa, characterized by the yellowish-white appearance, were sampled for biopsy and sent for histopathological assessment. The initial clinical pre-biopsy impression favored glycogenic acanthosis coexisting with gastritis.

Macroscopic assessment of the collected biopsy material revealed three individual, pale grey-white tissue fragments, measuring approximately 0.2cm.

Microscopically, the esophageal mucosa showed distinct, well-circumscribed nodules comprising of aggregates of polygonal cells. These cells have small, uniform, round nuclei and copious amounts of clear, finely vacuolated cytoplasm, characteristic of sebaceous lineage. Ancillary special histochemical stains, including Periodic Acid-Schiff (PAS) and PAS with Diastase digestion (PASD), were negative within the cellular aggregates, confirming the absence of glycogen and mucin respectively, while the PAS stain appropriately highlighted the epithelial glycogen (Figure 2-4). Consequently, a definitive histological diagnosis of ectopic sebaceous glands (sebaceous heterotopia) in the esophagus was rendered. The patient was subsequently managed conservatively with an empiric course of proton pump inhibitors and antacids, over a two-week period.

IMAGES

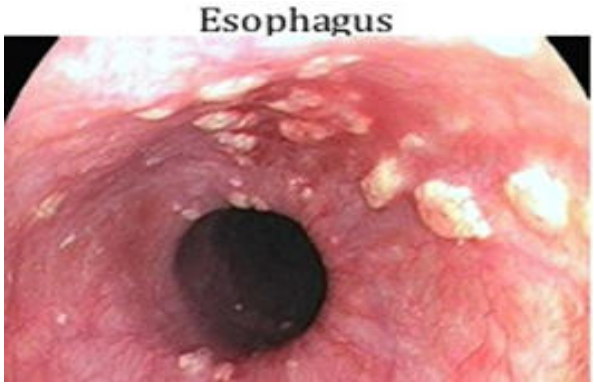

Fig.1: Upper gastrointestinal endoscopy revealed multiple yellowish-white shiny spots.

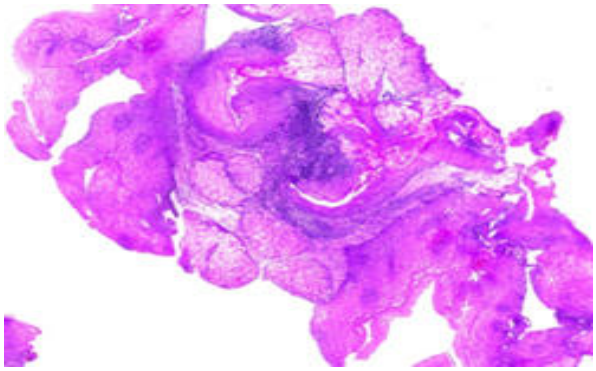

Fig.2: Esophageal mucosa with ectopic sebaceous glands. (40x)

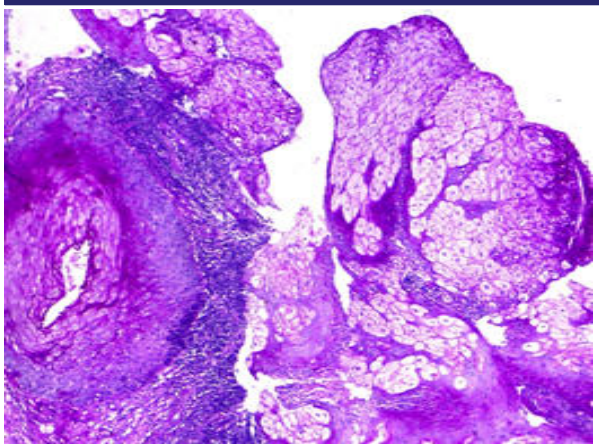

Fig.3: PAS negative sebaceous glands. (100x)

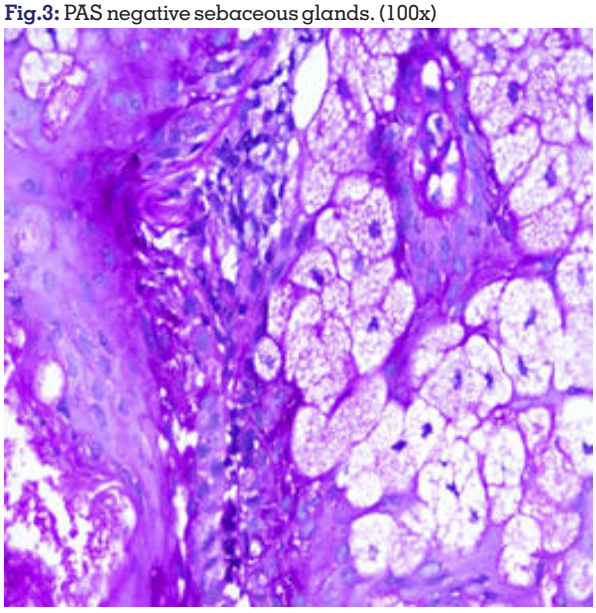

Fig.4: PASD negative sebaceous glands. (400x)

DISCUSSION:

Sebaceous glands are mesodermally derived adnexal structures which are primarily distributed in pilosebaceous units across the scalp, face, and other hair-bearing cutaneous regions. Their anomalous location in non-integumentary sites is denoted as "ectopic sebaceous glands" or "sebaceous heterotopia." While these glands are a common incidental finding in the oral cavity (e.g., Fordyce spots), external genitalia, and the parotid gland, their manifestation in the esophagus, an organ predominantly of endodermal lineage, is an exceptionally rare phenomenon. The inaugural documented instance was chronicled in 1962 by De Lava and Pickren. (7) Crucially, extended long-term surveillance data for such lesions has consistently failed to demonstrate any propensity for malignant transformation, thereby classifying these entities as unequivocally benign. (8)

Although the epidemiological landscape remains poorly defined, the reported mean age at the time of discovery is approximately 52 years, with no pronounced gender bias. These lesions most frequently colonize the middle and distal segments of the esophagus. (9,10) The histogenesis remains a subject of ongoing debate, with two principal hypotheses: a congenital developmental misplacement (choristoma) or an acquired metaplastic transformation. The increased frequency of detection in elderly patients lends greater credence to the theory of metaplasia, likely arising from esophageal submucosal glands, rather than an embryological anomaly. (5,6)

Endoscopic phenomenology typically involves the visualization of multiple lesions, often clustered in small, discrete foci (reported range of 25 to 100 lesions). The pathognomonic appearance is that of minute, yellow, slightly raised papules of variable size, interspersed amongst the unremarkable surrounding mucosa. The gold standard for definitive diagnosis rests upon meticulous histopathological assessment, which characteristically unveils lobules of sebaceous glands featuring the hallmark rounded nuclei and finely vacuolated cytoplasm. An associated excretory duct may occasionally be visualized.

Differential diagnostic considerations must include glycogenic acanthosis, mucosal xanthoma, squamous papillomas, and granular cell tumors. Given the inherently benign biological character of ectopic sebaceous gland endoscopic follow-up examinations are typically not recommended. (11)

CONCLUSION:

Sebaceous gland heterotopia in the esophagus represents an extreme rarity, likely constituting a manifestation of metaplastic conversion within the indigenous esophageal submucosal glands. Patients often present with vague, non-specific gastrointestinal complaints, necessitating clinical vigilance. The diagnosis is ultimately established through a combination of highly suggestive endoscopic findings and conclusive histopathological confirmation. This condition is unequivocally benign, devoid of any malignant potential, and thus neither necessitates active therapeutic intervention nor prolonged, intensive surveillance. Clinicians and endoscopists must maintain a high index of suspicion for this rare entity, as its accurate recognition is paramount to preclude unnecessary diagnostic protocols and therapeutic overreach.

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