CASE REPORT

An unusual cause for halitosis

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SUMMARY

This report describes an unusual cause for halitosis and an unusual treatment for the underlying problem. Halitosis is a symptom which can result from a diverse range of underlying pathologies, most frequently those affecting the oral cavity or respiratory tract. Uncommonly, it arises due to pathology within the upper gastrointestinal tract. The case of a patient presenting with severe persistent halitosis attributable to mesh erosion occurring 8 years after redo laparoscopic hiatus hernia repair is described. Full external healing of the erosion tract was observed such that no symptomatic oesophageal perforation resulted. Mesh erosion is typically managed with surgical intervention. In this case, the infected mesh was successfully removed endoscopically.

ascertain the presence or absence of malodour. Other assessment methods include the use of portable halimeters, which detect sulfides present in exhaled breath (other volatile compounds associated with halitosis are not detected by these devices), and gas chromatography, which allows quantification of gases present in the breath and can detect the full range of volatile substances associated with halitosis.^{1–3}

We will describe the case of a patient seen in the General Surgery clinic who developed persistent and progressive halitosis attributable to a gastrointestinal cause that has been previously unreported as giving rise to this symptom in the literature. The management of the case is also noteworthy, and will be the focus of the discussion to follow.

BACKGROUND

Halitosis is a common symptom. Transient halitosis is usually physiological, typically resulting from a combination of intraoral microbial metabolism and other contributors including dietary factors, alcohol intake or smoking.¹ Pathological causes are more often present in cases of recurrent or persistent halitosis, approximately 85% due to disease within the oral cavity including gingivitis, periodontal disease, oral ulceration and rarely intraoral malignancy. The remainder are accounted for by extraoral causes, including respiratory disease, disease of the upper gastrointestinal tract and diverse systemic causes such as uraemia, hepatic failure, ketosis and others.^{2–3} Distinctions have been made between so-called 'genuine halitosis', which is objectively verifiable, and 'pseudo-halitosis', which describes the subjective experience of breath malodour without objective evidence for its presence. However, recent classification systems place greater emphasis on the pathophysiological origin of halitosis, without the focus on subjective versus objective symptomatology.⁴

Mechanistically, the production of odour-causing substances occurs due to the interaction of anaerobic intraoral bacteria with specific amino acid substrates within the mouth. Such substances include volatile sulfur compounds, diamines, indoles and short-chain fatty acids.^{1–2} Extraoral halitosis may be caused either by the direct passage of similar compounds from the respiratory or gastrointestinal tracts into the mouth, or odorous substances may reach the oral cavity as a result of haematogenous transmission from their site of production and alveolar gas exchange.^{1–2–4} The gold standard for clinical assessment of halitosis is organoleptic testing, which involves formal assessment of exhaled breath by an experienced examiner to

CASE PRESENTATION

A 47-year-old woman presented to the surgical outpatient clinic with progressively worsening halitosis. She had previously undergone two fundoplication procedures for repair of a giant sliding hiatus hernia, the latest 8 years ago.

The patient initially presented with severe reflux oesophagitis in her early 30s. Oesophagogastrroduodenoscopy had demonstrated a 5 cm hiatus hernia with columnar metaplasia noted in the lower oesophagus. Her medical history was significant for asthma and primary hypothyroidism. A laparoscopic Nissen's fundoplication was performed with cruroplasty to reinforce the hiatal defect and formation of a full posterior fundal wrap, sutured to the anterior oesophagus and right crus of the diaphragm.

Despite initial symptomatic improvement, recurrence of the hernia occurred with return of severe reflux symptoms, and a redo laparoscopic fundoplication was performed 4 years later. On this occasion, synthetic polytetrafluoroethylene (PTFE) mesh was used to reinforce the hiatal defect (see figure 1), anchored with sutures to the diaphragmatic crura, and a partial anterior fundal wrap was created.

Following this second procedure, the patient experienced a dramatic improvement in her symptoms, which remained well controlled for some 18 months. Antireflux medications were discontinued. However, at subsequent follow-up, she began to report progressively worsening dysphagia. Endoscopy revealed the formation of a fibrotic stricture around the mesh at the site of the repair. A series of endoscopic and radiologically guided balloon dilations were performed over the following 6 years, which led to transient improvement in swallowing after each attempt.

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Figure 1 Example of U-shaped fenestrated prosthetic polytetrafluoroethylene (PTFE) mesh.

The past 12 months saw the development of progressively worsening halitosis. The patient's subjective symptom was confirmed by close family members, and was noted on clinical assessment. At repeat endoscopy, the mesh was seen to have eroded through the full thickness of the oesophageal wall at the level of the oesophago-gastric junction.

INVESTIGATIONS

With respect to evaluation of halitosis, our clinic lacks familiarity with methods of organoleptic testing and halitometry, as well as the necessary equipment for the latter, and thus these were not performed. Suspecting her symptoms to be related to her hiatus hernia, the patient reported her symptoms to our clinic in the first instance, while patients with similar symptoms arising de novo without an apparent cause might present initially to dental services, from where they will be referred for specialist input if necessary. Their evaluation is more likely to include these assessment modalities as part of the diagnostic workup.

TREATMENT

The management options to remove the eroding mesh were discussed with the patient, and agreement was reached to attempt to endoscopically excise the mesh in the first instance. In case of failure, consideration would be given to attempting removal via

a laparoscopic approach, but it was highlighted that in either case the risks were considerable, and if perforation occurred, a more drastic procedure, potentially excision of the oesophago-gastric junction, may be required.

In fact, complete removal of the mesh was accomplished successfully at endoscopy (figure 2). A postprocedure gastrografin swallow study demonstrated no contrast leak and the patient was discharged the following day.

OUTCOME AND FOLLOW-UP

The patient reported complete resolution of her halitosis, with improvement in her ease of swallowing following the procedure and no symptoms to suggest significant recurrence of gastro-oesophageal reflux.

DISCUSSION

This report describes the occurrence of a serious surgical complication heralded chiefly by a symptom of halitosis. Mesh erosion following hiatus hernia repair accounting for halitosis has, to the best of our knowledge, not been described previously in the literature. The patient's insidious presentation contrasts with the clinical picture of free oesophageal perforation that can result from mesh erosion. From a different perspective, the case is also notable in chronicling the outcomes of serial surgical procedures to manage a giant hiatus hernia in a single patient, and prompts reflection on the different surgical approaches to hiatus hernia repair and their relative merits and shortcomings.

The patient initially underwent reduction of her hiatus hernia with simple cruroplasty and fundoplication in 2002 to repair a very large hiatal defect, and unfortunately experienced recurrence of the hiatus hernia. A redo procedure was carried out 4 years later, where repair of the defect was reinforced using PTFE mesh. This was complicated by fibrotic stricture formation and then some 8 years later by mesh erosion, which presented as progressive halitosis. Ultimately, the erosion was managed by means of endoscopic removal, without the need for more complex intervention.

While gastrointestinal causes for halitosis are uncommon, gastro-oesophageal reflux disease and *Helicobacter pylori* infection are established causes. Both are associated with production of volatile sulfur compounds which may reach the oral cavity by either direct or blood-borne transmission.^{1 2 4} Where these conditions give rise to breath malodour, treatment has been shown to lead to resolution of halitosis.^{1 2} Interestingly, our patient had suffered from severe gastro-oesophageal reflux in the past but had not reported halitosis prior to the development of mesh erosion. We may speculate that the mechanisms giving rise to malodour associated with mesh erosion may be similar to those causing halitosis due to other oesophageal or gastric pathology,

Figure 2 Photographs of 8×11 cm eroding mesh following endoscopic removal from level of gastro-oesophageal junction after 8 years in situ.

although we were not able to formally investigate this patient's halitosis within our clinic. In this case, the probable cause for the patient's symptom was apparent on endoscopy, and the high-risk nature of her problem warranted urgent intervention. Although the specific pathology reported here is clearly a highly unusual cause for halitosis, even among gastrointestinal causes, it highlights the importance of considering a wide range of differential diagnoses for this symptom in the context of the patient's background and medical history.

Mesh-associated stricture formation and erosion are increasingly recognised as uncommon but serious complications of prosthetic reinforcement of hiatus hernia repair.^{5–9} One study in the USA based on a survey of self-reported surgical practice reported a stricture rate of 0.2% and an erosion rate of 0.3%,⁵ but more robust estimates from formal prospective follow-up studies are lacking. Although mesh erosion typically presents as an emergency, there have been a small number of prior case reports of similar occurrences of gradual development of full thickness erosion without frank perforation which were managed successfully with endoscopic excision.^{5–9}

In determining the optimal approach to hiatus hernia repair, the evidence from high-quality comparative randomised trials and follow-up studies is limited, but suggests lower hernia recurrence with mesh repair, particularly where the hiatal defect is large. However, the risk of mesh-related complications must be borne in mind and the decision made by considering the likely risk and implications of both these adverse outcomes.^{10–11}

Learning points

- ▶ Surgical causes within the gastrointestinal tract should be considered among the causes of persistent halitosis.
- ▶ Stricture formation and erosion are rare but serious complications of mesh repair of hiatus hernia.
- ▶ Very unusually, mesh erosion through the oesophageal wall can occur with full external healing such that no visceral perforation results, which may be managed by endoscopic removal of the offending mesh.
- ▶ Studies evaluating the long-term outcomes of mesh reinforcement will aid decision-making regarding the best approach to hiatus hernia repair in the individual patient.

The latest guidelines issued by the Society of American Gastrointestinal and Endoscopic Surgeons (SAGES) in 2013 emphasise the evidence in favour of reduced early recurrence with mesh reinforcement for large hiatus hernias; however, lack of long-term data on mesh repair precluded a recommendation being issued for or against the use of mesh in this revision.¹²

For our patient, we anticipate that periprosthetic fibrosis while the mesh was in situ will have caused durable reinforcement of the defect closure and render further hernia recurrence unlikely. Further follow-up will be needed to determine whether symptoms resulting from the oesophageal stricture persist or improve following removal of the mesh.

Contributors GB was the primary author. HA, the supervising consultant surgeon, performed the procedure described and provided editing guidance. MS assisted during the procedure and contributed to the writing of the report. LP provided editing guidance. There were no other non-author contributors.

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REFERENCES

- 1 Scully C, Greenman J. Halitosis (breath odour). *Periodontology* 2008;48:66–75.
- 2 Bollen CML, Beikler T. Halitosis: the multidisciplinary approach. *Int J Oral Sci* 2012;4:55–63.
- 3 Delanghe G, Ghyselen J, Feenstra L, et al. Experiences of a Belgian multidisciplinary breath odour clinic. In: Van Steenberghe D, Rosenberg M, eds. *Bad breath: a multidisciplinary approach*. Leuven, Belgium: Leuven University Press, 1996:119–207.
- 4 Aydin M, Harvey-Woodworth CN. Halitosis: a new definition and classification. *Br Dent J* 2014;217:E1.
- 5 Frantzides CT, Carlson MA, Loizides S, et al. Hiatal hernia repair with mesh: a survey of SAGES members. *Surg Endosc* 2010;24:1017–24.
- 6 Stadlhuber RJ, Sherif AE, Mittal SK, et al. Mesh complications after prosthetic reinforcement of hiatal closure: a 28-case series. *Surg Endosc* 2009;23:1219–26.
- 7 Hazebroek EJ, Leibman S, Smith GS. Erosion of a composite PTFE/ePTFE mesh after hiatal hernia repair. *Surg Laparosc Endosc Percutan Tech* 2009;19:175–7.
- 8 Tatum RP, Shalhub S, Oelschlager BK, et al. Complications of PTFE mesh at the diaphragmatic hiatus. *J Gastrointest Surg* 2008;12:953–957.
- 9 Griffith PS, Valenti V, Qurashi K, et al. Rejection of Goretex mesh used in prosthetic cruroplasty: a case series. *Int J Surg* 2008;6:106–9.
- 10 Furnee E, Hazebroek E. Mesh in laparoscopic large hiatal hernia repair: a systematic review of the literature. *Surg Endosc* 2013;27:3998–4008.
- 11 Tam V, Winger DG, Nason KS. A systematic review and meta-analysis of mesh vs suture cruroplasty in laparoscopic large hiatal hernia repair. *Am J Surg* 2016;211:226–38.
- 12 Kohn GP, Price RR, DeMeester SR, et al., SAGES Guidelines Committee. Guidelines for the management of hiatal hernia. *Surg Endosc* 2013;27:4409–28.

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