Oesophageal intramural pseudodiverticulosis: a rare endoscopic finding

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SUMMARY
Oesophageal intramural pseudodiverticulosis is an uncommon condition, characterized by multiple small rounded cavities seen in the oesophageal wall during oesophagogastroduodenoscopy. It is often associated with gastro-oesophageal reflux disease, achalasia, oesophageal candidiasis and diabetes mellitus. We report a 40 year old Nigerian man who presented with recurrent dysphagia and endoscopic findings typical of oesophageal intramural pseudodiverticulosis. The patient was managed medically with resolution of the dysphagia. This report highlights the occurrence of this rare and benign cause of dysphagia in Nigeria.

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INTRODUCTION
Oesophageal intramural pseudodiverticulosis (OIP) is a rare condition that is characterized by multiple small flask-shaped outpouching lesions of the oesophageal wall.[1-3] Not more than 200 cases have been reported worldwide.[4] OIP is a benign disease of unknown cause and it is diagnosed by typical findings of multiple tiny orifices in the oesophageal mucosa on endoscopy, oesophageal barium contrast radiography, and pathologic findings of abnormal dilatation of the submucosal glands, usually from surgical specimen.

OIP is seen mainly in men between 50-60 years of age, though it can present at any age.[2] The most common symptom is dysphagia (seen in 80% of patients), often associated with oesophageal stricture as a sequelae.[5] Other symptoms are chest pain and vomiting seen in minority of cases. OIP can also be asymptomatic.

Though the cause is uncertain, it has been hypothesized that it may be due to chronic irritation of the oesophagus, and/or exaggerated (hypermotility) movement of the oesophagus. These factors cause obstruction or compression of the submucosal ducts of the oesophagus, leading to the formation of these pseudodiverticulae.[4] It is usually associated with diabetes mellitus (DM), oesophageal candidiasis, alcohol consumption and gastro-oesophageal reflux disease (GORD).[3]

We report a case of a young man with OIP who had typical endoscopic findings and was managed medically with resolution of his symptoms.

CASE REPORT
A 40-year old Nigerian man presented at the Gastroenterology clinic with a two-year history of recurrent dysphagia. The dysphagia was to solid foods and not to fluids. Solid foods were chewed into smaller bits before being swallowed, and aided with water. The patient`s symptoms had worsened in the last one month with sensation of food sticking to his upper throat. There was occasional retrosternal pain, described as burning in nature, non-radiating, and aggravated by meals. There was no history of food nor acid regurgitation. There was also no history of vomiting, haematemesis, weight loss, abdominal pain, abdominal swelling nor change in bowel movement.
He neither complained of joint pains nor body rash.

He was not a known patient with DM, systemic hypertension or GORD. His family history was not contributory or significant. He neither smoked cigarette nor ingested alcohol. Physical examination was unremarkable.

The platelet count was 179 x 10^9/L (Reference: 150-450x 10^9/L); packed cell volume (PCV), 39% (Reference: 35-45%); Haemoglobin count was 14g/dl(Reference: 13.5 - 17.5g/dl) and white blood cell count, 7.4 x 10^9/L (Reference: 4 -12x10^9/L).

Serum creatinine was 83 µmol/l (Reference: 60-100 µmol/l), urea was 5.7 mmol/l (Reference: 2.5-6.5 mmol/l), potassium was 4.1 mmol/l (Reference: 3.5-5.0 mmol/l), sodium was 138 mmol/l (Reference: 135-145 mmol/l).Serum alanine aminotransferase was 10iu/L (Reference: 4-36iu/L), Serum aspartate aminotransferase was 14iu/L(Reference: 4-36iu/L), Serum Alkaline Phosphatase was 88iu/l(Reference:45-146iu/L), Serum Bilirubin was 12mmol/l(Reference: 4-17mmol/l). Viral screening for hepatitis B, C and HIV were all negative.

The patient's chest x-ray appeared normal. Abdominal ultrasound scan and electrocardiography (ECG) findings were normal. Oesophageal barium contrast radiography was unremarkable.

Oesophagogastroduodenoscopy (OGD) examination, however, revealed multiple small openings in the oesophageal mucosa, measuring between 2 to 4 mm in diameter, extending from the upper one-third to the mid one-third of the oesophagus. There were whitish plaques attached to oesophageal mucosa (Figure 1). An endoscopic diagnosis of OIP with oesophageal candidiasis was therefore made.

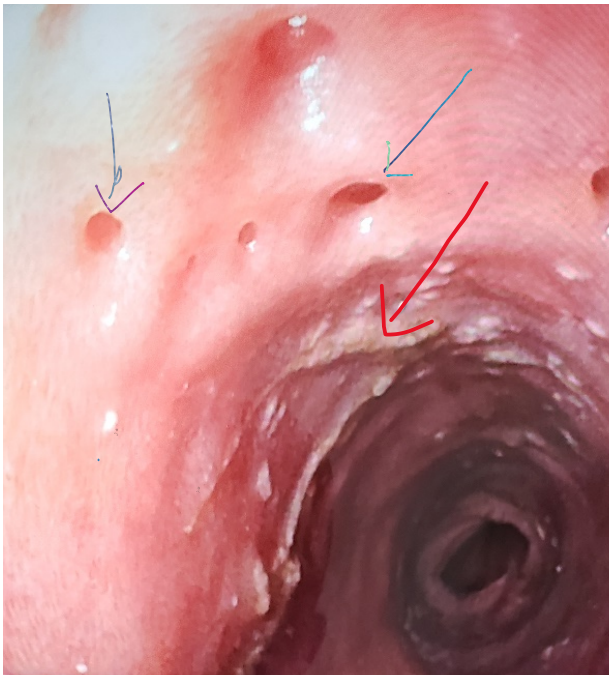

Figure 1 Oesophagogastroduodenoscopy showing small oval cavities with smooth edges in oesophageal wall (blue arrow) and whitish plaques (red arrow) attached to the oesophageal mucosa.

Histology report of the oesophageal mucosa biopsy showed non-specific inflammation with fungal infection. The patient was placed on the antifungal agent fluconazole, proton pump inhibitor (PPI) and antibiotics.
The patient`s symptoms resolved two weeks after starting medical therapy. He is currently being followed up in the clinic. A repeat OGD was planned for six months post treatment, the patient however refused to consent for a repeat procedure.

DISCUSSION

Oesophgeal intramural pseudodiverticulosis is a rare benign oesophageal disease with typical endoscopic findings of small orifices in oesophageal lumen.[6] Mendl *et al*, first reported OIP in 1960, and about 200 OIP reports have been published world-wide since then.[6] Though the aetiology is unknown, studies have postulated that obstruction of the submucosa glands of the oesophageal mucosa may have led to subsequent dilation of these excretory ducts as a result of inflammatory cells, epithelial desquamation, submucosa fibrosis, or a combination of these factors. OIP is also seen in GORD and infectious oesophagitis.[7] It has also been associated with DM, oesophageal candidiasis, achalasia and other motor disorder of the oesophageal wall.[8, 9]

To the best of our knowledge, our case will be the first to be reported in Nigeria, as most cases reported were seen in the western countries. Our patient was 40 years of age, which is below the predominant age range for OIP,[2] however it has been reported to be seen in any age group. This index case presented with dysphagia and retrosternal pain, the former being the commonest symptom reported in patients with OIP,[7] while the retrosternal pain is less common in patients with OIP. The retrosternal pain in our patient could be as a result of the associated candida esophagitis or possibly due to GORD, though findings suggestive of GORD were not present at endoscopy, it cannot be completely ruled out.

The endoscopic findings of our patient were multiple small orifices which was seen in the upper and middle one-third of the oesophagus and oesophageal candidiasis. These findings are typical of OIP with common associations mentioned in several literatures.[8] Though this patient presented with dysphagia, no stricture was seen both on OGD and oesophageal barium contrast radiography. The dysphagia could have been as a result of the oesophageal candidiasis, and the disappearance of this symptom following medical therapy further buttresses this point.

The treatment of OIP consists of the treatment of oesophageal inflammation and other associated conditions such as oesophageal candidiasis, strictures and perforations.

Treatment also includes but is not limited to antibiotic therapy.[10,11] Endoscopic dilatation can be performed to reduce the symptoms of OIP in patient with concomitant oesophageal stenosis. It has been reported that long-term surveillance showed no regression of diverticulosis despite appropriate treatment and improvement in the general condition of the affected patients.[10, 11]

The patient was treated medically with proton pump inhibitors and antifungal for two weeks with resolution of symptoms. However, repeat endoscopy could not be done due to patient`s refusal.

In summary, Oesophageal intramural pseudodiverticulosis is a rare condition and rarer in this part of the world. Our patient was diagnosed with typical endoscopic findings following a longstanding history of dysphagia. The patient was managed medically with resultant significant improvement in his symptoms. We believe that our report of this rare condition will further raise awareness thus aiding its prompt diagnosis and treatment.

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