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Case report

Intramural oesophageal dissection as an unusual presentation of chest pain: A case report

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HIGHLIGHTS

- This is the first known case of IOD resulting from the use of bisphosphonates.
- Bisphosphonates have a well documented adverse effects profile.
- There is a paucity of information regarding the appropriate management of IOD.
- Our findings, diagnosis and management are described in this case report.

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ABSTRACT

Introduction: Intramural oesophageal dissection (IOD) is a rare clinical condition and there is a paucity of information regarding the appropriate diagnosis and management. It is described as bleeding in the submucosal plane of the oesophagus, and has various documented causes.
Presentation of case: We report a case of a 73 year old female who developed IOD. She presented with severe chest pain. Subsequent imaging revealed IOD and haematoma formation. This was confirmed on oesophagogastroduodenoscopy (OGD). She was on a bisphosphonate for her osteoporosis, as well as having age-related dysmotility of her oesophagus on manometric studies. She was also taking fish oil. Treatment was conservative and the patient was discharged with proton pump inhibitors and follow up.
Discussion: Spontaneous haematoma formation and IOD resulted likely from a combination of the anticoagulant effect of fish oil and oesophageal dysmotility. Bisphosphonates also have some well documented gastrointestinal side effects involving mucosal damage. The possibility that the concurrent use of bisphosphonate led to a pre-existing ulcer which could have contributed to the development of IOD in this patient should be considered.
Conclusion: spontaneous IOD can occur in elderly patients who are anticoagulated. Fish oil has not been previously reported as having an association with IOD. This is the first known reported case of spontaneous IOD occurring in association with concurrent use of a bisphosphonate and fish oil. IOD is a rare disorder, and any anticoagulated patients presenting with severe chest pain may need careful investigation prior to definitive management.

1. Introduction

Intramural oesophageal dissection (IOD) is an uncommon clinical condition which is not readily described in most surgical textbooks [1]. It occurs as a result of bleeding into the submucosal plane of the oesophagus, with tearing of the mucosa, most often in association with anticoagulation or in medical conditions that predispose to bleeding. It has been noted that oesophagitis may also play a part in facilitating rupture [2–4]. Although oesophagitis and oesophageal ulceration has been commonly reported, IOD remains a scarcely reported pathological entity. There have been several case reports describing this entity since it was first published by Williams in 1957 and again by Marks and Keet 11 years later, which they had termed 'intramural rupture of the oesophagus' [2,5]. Most of the cases that have been reported have occurred in female patients in their seventh or eighth decades [3]. Presented here is a case of IOD in an anticoagulated patient taking regular bisphosphonate, and potential risk factors and causes are explored.

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2. Case report

A 73-year-old female presented to the emergency department May 2014, 9 h after developing a sudden onset of severe central chest pain radiating into her back and into her neck. She was able to swallow liquids but had some difficulty swallowing solid food. No reported haematemesis or malaena. There was a 4 month history of dysphagia prior to this admission. Her past medical history includes severe osteoporosis and diverticulosis. Her last OGD was normal a year ago. She had been taking Fosamax 70 mg for three years, and reports adherence to product instructions of sitting upright for 30 min and taking with water. Her other pre-admission medications include Premarin 0.3 mg (conjugated oestrogen as part of hormonal replacement therapy, 20 years duration), Vitamin B12, Zinc, Fish oil 1000 mg once daily. She is a non-smoker. Her initial blood profile revealed a haemoglobin of 120 g/L, platelets 198 × 10^9/L, coagulation profile normal with an INR 1.0. A normal electrocardiogram and cardiac biochemical profile made the diagnosis of acute coronary syndrome less likely. A subsequent CT

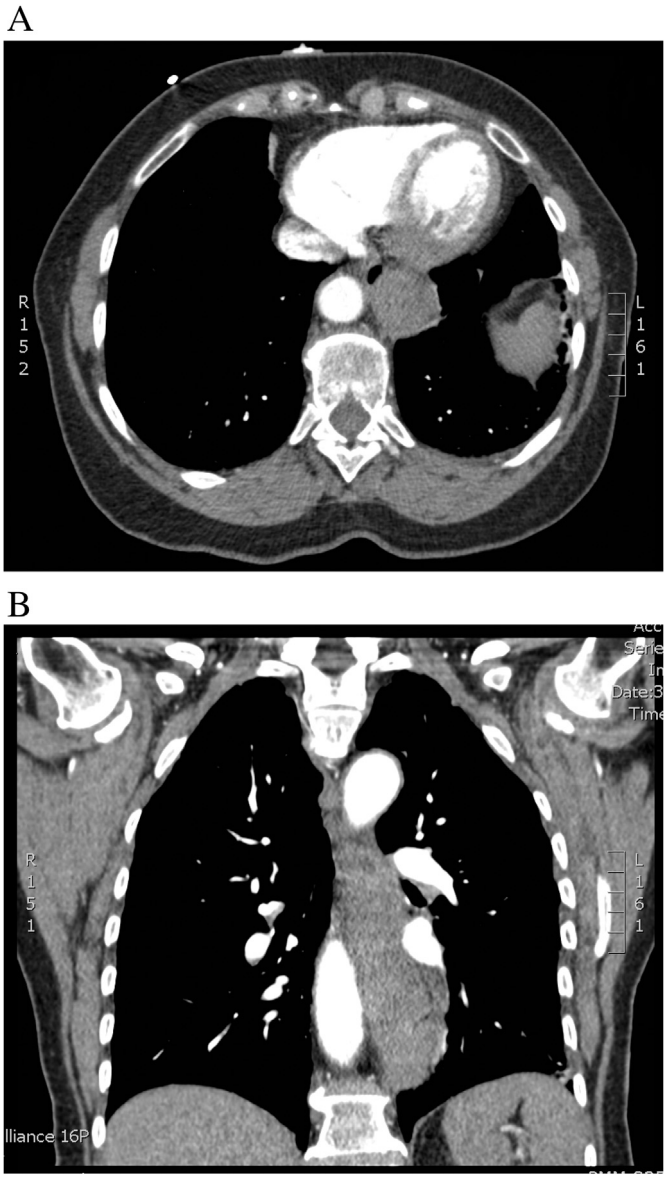

Fig. 1. A and B: CT pulmonary angiogram shows dilated, fluid-filled oesophagus.

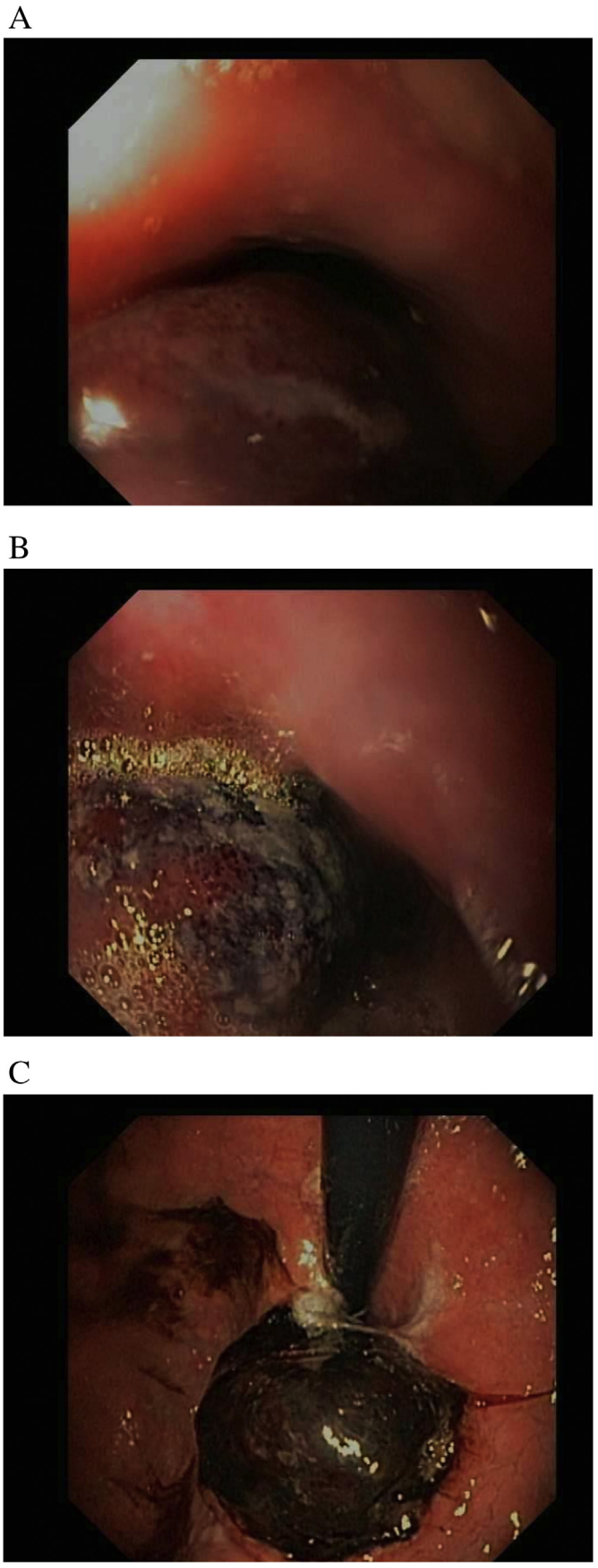

Fig. 2. A & B: Initial OGD on presentation. There is a large, contained submucosal haematoma extending from middle to lower third of the oesophagus. C. On retroflexion, there was a stable, non-bleeding ulcer with an overlying clot in the cardia of the stomach, just below the GOJ.

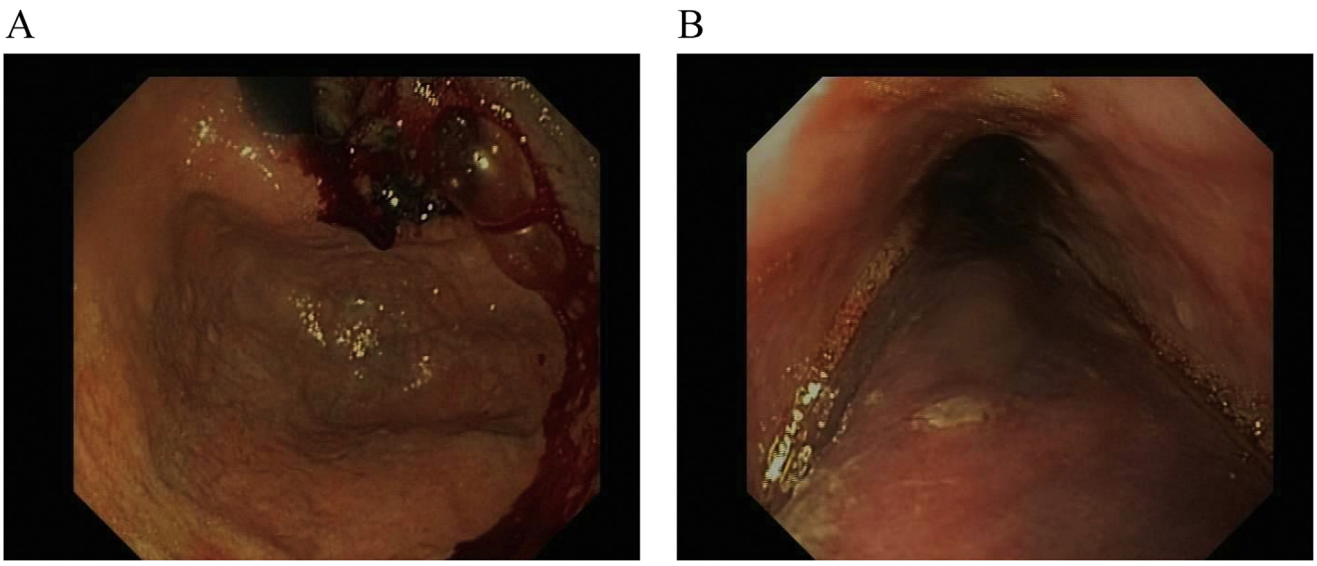

Fig. 3. A: A healing ulcer after 5 days of PPI therapy. B: The initial submucosal haematoma has since organised, and mucosal sloughing and ulceration is evident.

pulmonary angiogram was performed to exclude a pulmonary embolus based on a positive D-Dimer test, which was negative for an embolus but revealed a dilated, fluid filled oesophagus (38 mm in diameter) with a small amount of air in the lumen. There was no lymphadenopathy in the thorax (Fig. 1A & B).

An OGD revealed a large submucosal haematoma extending from just above the gastro-oesophageal junction (GOJ) to 20 cm proximally, involving at least 50% of the oesophageal lumen situated anteriorly and filling most of the lumen (Fig. 2A & B). Just below GOJ on retroflexion a large clot of the cardia of the stomach was seen (Fig. 2C). There was a stable clot overlying the ulcer which was not disturbed. The rest of the stomach and examined duodenum was normal. The patient was commenced on a proton-pump inhibitor (PPI) infusion and admitted to the intensive care unit (ICU) for monitoring.

Another OGD was performed five days later, showed one oozing and cratered gastric ulcer with adherent clot was found in the cardia (Fig. 3A). This lesion was 10 mm in largest dimension. A large oesophageal ulcer with sloughing of the mucosa was found 34–40 cm from the incisors (Fig. 3B). A haematoma with mucosal ulceration was found in the lower third of the oesophagus was found, and this area was slightly stenosed but the scope was able to be passed without difficulty.

The patient remained haemodynamically stable during her stay, and her pain had settled with PPI therapy and cessation of bisphosphonate. She was discharged on oral PPI. A repeat OGD four weeks later showed a completely regenerated area of mucosa with only vague outlines of the previous ulcer still present. There was no stenosis or other complications. The scope passed freely through this area. The previously observed ulcer had now healed with mild scarring noted. The healed ulcer and stomach lining was biopsied on this occasion and histology confirmed mild chronic superficial gastritis. There were no Helicobacter-like organisms present. No evidence of dysplasia or malignancy. Her symptoms during admissions improved significantly, and she will continue her PPI indefinitely. She underwent further manometric testing which revealed a non-specific motor abnormality of the oesophagus. Her lower oesophageal sphincter pressure was normal (12 mmHg) and relaxed with swallow. The study concluded the abnormalities were likely due to age-related oesophageal dysmotility.

3. Discussion

IOD is rare, and various aetiologies have been described. Mechanical trauma, including the ingestion of foreign bodies and pills, and also association with Mallory–Weiss tear has been suggested [6–8]. Iatrogenic causes such as nasogastric tube insertion, OGD, trans-oesophageal echocardiography have all been documented [8–10]. This pathology has known to occur in the presence of coagulopathic disorders or patients who are anticoagulated [11–13]. In a few reported cases IOD can occur spontaneously, and no known cause can be found [4,8,14].

Drug-induced oesophageal injuries have been widely reported, from medications including tetracyclines, non-steroidal anti-inflammatory drugs, potassium chloride, quinidine, and bisphosphonates – particularly alendronate [1,6]. Injuries described in a 2003 review of adverse drug reactions related to oesophageal injuries include dysphagia, oesophageal ulceration, strictures, oesophagitis, haematemesis, and perforation [6]. Bisphosphonate induced injuries of the oesophagus are generally more severe than those caused by other medications [1]. However, IOD resulting from medication use still remains an extremely rare entity [7].

Anticoagulation has been implicated as a primary causative factor for spontaneous oesophageal haematoma. Medications such as warfarin, clopidogrel, and systemic thrombolysis for treatment of acute coronary syndrome have been reported to cause oesophageal haematomas, in some cases in the absence of vomiting, cough, or oesophageal instrumentation. There have been no reported cases of this pathology occurring with the use of fish oil alone [11–13].

Adverse upper gastrointestinal effects are common with bisphosphonate use [1,6,15–17]. Reported adverse events include oesophageal ulceration, oesophagitis, oesophageal stricture, haematemesis, dysphagia, dyspepsia, indigestion, and epigastric or chest pain. Several of these reports were confirmed by endoscopic studies revealing severe and extensive oesophageal ulcerations [18–20]. In clinical studies 1.5% of patients taking alendronate were quoted to have developed oesophageal ulcerations [18].

Alendronate is a nitrogen-containing bisphosphonate (amino-bisphosphonate) that works by preventing osteoclast mediated bone resorption. Sodium alendronate (Fosamax) was approved for

use and marketed in 1996, and within three years had received 331 reports of adverse reactions, more than half related to gastrointestinal effects. 87% of these were female [18]. In the literature, alendronate is currently the most commonly reported cause of medication induced oesophagitis [1]. Alendronate has been associated with development of gastric erosions and ulcerations in as little as the first few weeks from initiation of therapy [10], to over a year after commencement [8]. This medication is contraindicated in patients who cannot remain upright for at least 30 min, have oesophageal dysmotility and delayed emptying, and hypocalcaemia.

This report presents a case of an acute bleeding of a gastric ulcer close to the gastro-oesophageal junction, which led to haematoma formation, subsequently extending into a dissection of the submucosa of the oesophagus. Several factors could have contributed to the development of IOD in this patient. The most likely is that the combination of fish oil and age-related oesophageal dysmotility had significant contribution to the development of this patient's presentation. In addition, the concurrent use of alendronate, and given the well recognized adverse side effect profile, could raise the suspicion that this medication could also have contributed. However, there is no substantiating evidence to this or reports in the literature to date.

4. Conclusion

Intramural oesophageal dissection is a rare disorder of the GI tract. Several causative factors have been investigated. IOD often occurs in elderly patients, predominantly female, with significant comorbidities. Although difficult to identify the true cause of IOD in this case, anticoagulation, oesophageal dysmotility, and potential mucosal injury precipitated by bisphosphonate use could be considered. There is little information regarding the diagnosis, management and follow-up of this condition, and although the intention of this article is not to promulgate a standard therapy, it is hoped that this could provide a further insight to the natural history and progression of IOD, as well as raising in the literature a potentially unrecognised adverse effect of bisphosphonate use. This will likely need further investigation. Care should be taken nevertheless in prescribing bisphosphonates in elderly patients who may not be able to fully adhere to instructions for use, or those with severe GI reflux, oesophageal motility disorders, strictures or achalasia.

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Ryo Mizumoto – first author.
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Conflicts of interest

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