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Not to be Missed Entity: Dieulafoy's Lesion!

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Authors' contributions

This work was carried out in collaboration between both authors. Both authors read and approved the final manuscript.

Case Study

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ABSTRACT

Clinically missed Dieulafoy's lesion is a significant cause of gastro-intestinal bleeding with a poor prognosis. We hereby compare an autopsy case of a similar nature; thereby highlighting the importance of autopsy. 69 years old male living in a retirement home was being managed for anemia. He was admitted with a preliminary diagnosis of septicemia with deteriorating renal function. He was found dead and referred for autopsy to our institute. At autopsy, 3 small defects at the gastroesophageal junction measuring about 5-7 mm were seen, which on microscopy revealed a relatively large artery at the submocosal level with rupture. There was about 2 liters of blood in the stomach. This was a Dieulafoy's lesion, the cause of sudden massive gastrointestinal bleeding and subsequent shock. This signifies that autopsy still holds its value in this modern era of diagnostics.

Keywords: Dieulafoy's lesion; gastro-intestinal bleeding; NSAIDs; autopsy.

1. INTRODUCTION

In a recent publication, Christoffersen RK et al. [1] reported a case of sudden and unexpected death in a previously healthy male due to Dieulafoy's lesion (DL) of the esophagus. They concluded that DL is rarely seen in the autopsy room and emphasized the importance of autopsy. We would like to add one more case, wherein an autopsy in the investigation of sudden death revealed massive internal hemorrhage as a consequence of a

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ruptured DL. In our case too, bleeding from a DL proved to be fatal and highlighted the valuable contribution of autopsy in retrospective diagnosis creating awareness about this missed lesion.

DL is a well known form for arteriovenous malformation. It is in the differential diagnosis list of unsuspected gastro-intestinal bleeding, and is not easily recognized [2]. This may be due to lack of awareness, inaccessible site or size [3], and an overlapping clinical presentation with nonsteroidal anti-inflammatory drugs (NSAIDs) intake, peptic ulcer or alcohol abuse [4].

2. CLINICAL SCENARIO

69 years old male with previous two episodes of apoplexy and lumbar L4/L5 disc prolaps was known to have single right-sided kidney. He was living in a retirement home and was being treated for chronic anemia of normocytic normochromic type. He was admitted in the Department of Medicine with a confused state of mind and a preliminary diagnosis of septicemia. He had a high C-reactive protein and peripheral blood smear examination revealed neutrophilic leucocytosis. He was initially treated with Ciprofloxacin and later with Tazocin, as Gram positive bacteria were cultured in his blood with an unknown focus. He was regularly on antiplatelet medicines (Aspirin, Dipyridamole), antihypertensive drugs (Amlodipine, Losartan) and hypolipidemic drug (Simvastatin). He used to consume about 1-2 alcoholic drinks daily. He had deteriorating renal function with rising creatinine and a low Glomerular Filtration Rate. He had no abdominal complaints and was hemodynamically stable. He was found dead in his bed. He was referred for autopsy to our institute in order to determine the exact cause of death.

At autopsy, about 2 liters of fresh blood were found in the stomach and 3 small defects at the gastroesophageal junction measuring about 5-7 mm were noted. On microscopy, one of the sections revealed a large ruptured artery (size 2.15 mm) at the submucosal level with a thrombus-like material (Fig. 1). DL was the cause of sudden massive bleeding leading to hypovolemic shock and death. Other autopsy findings included atherosclerosis of the coronary arteries and the aorta, lung congestion, liver steatosis, and a single right-sided kidney with nephrosclerosis.

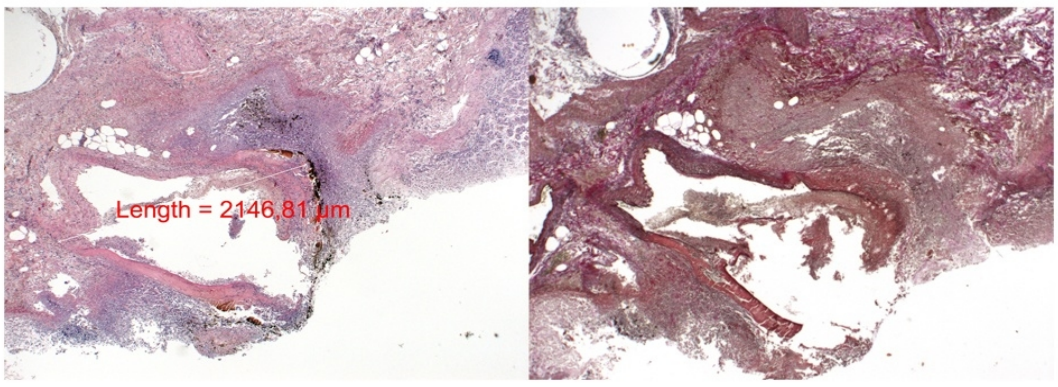

Fig. 1. Ruptured dieulafoy's lesion x 2 haematoxylin & eosin stain and weigert- Van gieson stain

3. DISCUSSION

A DL was diagnosed as the underlying cause of death. This was in accordance with the case report by Christoffersen RK et al. [1] and using established pathological diagnostic criteria such as the presence of a relatively large artery at the level of muscularis mucosae or higher accompanied by a vein of similar caliber with a significant risk of ulceration and bleeding [5]. In the literature, this is also described as a histologically normal vessel that has an abnormally large diameter (1-3mm) and runs a tortuous course within the submucosa protruding through a small mucosal defect varying from 2-5 mm, with a fibrinoid necrotic base [3]. Importantly, there is an absence of inflammation at the edge of the mucosal defect in contrast to that present in peptic ulcer disease [6].

DL can be clinically diagnosed by endoscopy and by using Dy's criteria, angiography or red cell scanning [3] with a clinical presentation of massive hemorrhage which is often recurrent. But in our case there were neither episodes of bleeding, nor abdominal pain, which was similar to the clinical presentation in the publication [1].

The pathogenesis in DL may be related to peptic ulcer disease, alcohol abuse and/or drugs such as NSAIDs, warfarin or aspirin [2]. Despite a lack of direct evidence that the presentation of this lesion is caused by mucosal erosions due to use of aspirin, coumadin, or NSAIDs, the use of these medications has been reported in more than 50% of patients. Peptic ulcer was not seen along with DL in our case. The rupture of DL was visualized histologically in contrast to the publication [1]. Comorbidities—particularly cardiovascular disease, hypertension, chronic renal failure, diabetes, and excessive use of alcohol—have been described in almost 90% of the patients [6]. More than one of these factors were present in our patient's clinical history and this lesion could also be one of the contributing causes for chronic anemia in our patient. This was in contradiction to the case report, which involved a healthy male with unremarkable medical history [1].

It is aptly quoted in the publication that, awareness of this condition is a key to accurate diagnosis [1]. But it can be easily overlooked at endoscopy [4]. Before the advent of endoscopy, the diagnosis was basically made during autopsies or surgeries [7]. It is underdiagnosed rather than being a rare disease [2]. In our patient, there was no clinical suspicion of such condition.

It is therefore clinically important to report this second case of missed DL, as "a potentially life threatening entity". We would also conclude that these autopsy findings are valuable contribution to the retrospective diagnosis and autopsy still holds its value in this modern era of diagnostics.

CONSENT

Autopsy consent was given by the dead person's sister. The manuscript is anonymous.

ETHICAL APPROVAL

Not applicable.

COMPETING INTERESTS

Authors have declared that no competing interests exist.

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