



Case report

A giant gastrointestinal stromal tumor revealed by a life-threatening hematemesis: A case report of a rare entity and a challenging management

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ABSTRACT

Introduction: Gastrointestinal stromal tumors (GIST) are the most common mesenchymal malignancies in the digestive tract. Despite the stomach being their site of predilection, only a few giant GISTs have been discovered in hypovolemic shock and require urgent surgery.

Case presentation: A 62-year-old patient was admitted for hematemesis. Initial examination revealed a compensated hemodynamic shock with a mass in the left hemi abdomen of 20 cm without signs of portal hypertension or peritonitis. After resuscitation, an abdominal CT scan was performed, showing an exophytic mass of the gastric antrum without local or distant lymph node involvement. Later, the patient went into hemodynamic shock, requiring vasoactive drugs. An urgent midline laparotomy was performed, revealing a 20-cm gastric GIST in the anterior wall of the antrum. Wedge resection was performed. The anatomopathological report confirmed the diagnosis of gastric GIST with a moderate risk of recurrence. The patient received adjuvant therapy for 3 years. No recurrence was detected.

Clinical discussion: Bleeding gastric GISTs revealed by a hypovolemic shock is a rare yet critical scenario. Considering GISTs as an etiology of digestive bleeding in life-threatening cases requires a high index of clinical suspicion. Prompt intervention is vital to control hemostasis and ensure patient stability.

Conclusion: Open surgery is mandatory for large bleeding GISTs. The surgical approach must be tailored to the tumor's specific location. Diligent execution of the surgical procedure is vital to prevent tumor rupture. Despite their size, these tumors have a favorable prognosis, enhanced by adjuvant therapy for moderate to high recurrence risk.

1. Introduction

GISTs are common mesenchymal tumors of the digestive tract, with an annual incidence of 10–15 cases per 1,000,000 patients, a rate that has exhibited a rising trend in the last few years [1]. While these neoplasms can manifest at various locations within the digestive system, the stomach emerges as the predominant site for their detection [2].

While GISTs can present with a vast myriad of unspecific symptoms, patients with gastric locations have the lowest rate of admission for acute symptoms and emergency surgical resection [3]. To date, there is limited literature on gastric GISTs presenting life-threatening gastrointestinal bleeding.

We present an exceptional case of gastric GIST discovered following a severe episode of gastrointestinal bleeding requiring emergency surgery. Our aim is to underscore the intricacies associated with the surgical management of sizable gastric GISTs and to explore the specific technical challenges linked to tumor locations. Through this paper, we aspire to contribute valuable insights that may aid in refining the approach to similar clinical scenarios. This case report has been reported in line with the SCARE Criteria [4].

2. Case report

A 62-year-old patient, without current medical diseases or past

Abbreviations: GISTs, gastrointestinal stromal tumors.

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abdominal surgeries, was admitted to our emergency department for recurrent episodes of hematemesis in the last 24 h.

For the last 10 years, the patient has complained of permanent headaches, dizziness, and exhaustion upon performing daily tasks, but she never consulted a physician.

On examination, initially, the patient was lucid and pale, with a blood pressure of 130/60 mmHg, a heart rate of 110 BPM, and a peripheral pulse of good volume with warm and pink extremities. The respiratory assessment was within normal limits. Abdominal examination revealed a firm, painless mass in the left hemi-abdomen, measuring 20 × 6 cm, without local inflammatory signs. No signs of peritonitis or portal hypertension were recorded. The rectal examination did not reveal any masses or hematochezia.

Urgent blood typing, cross-matching, and complete blood counts with iron studies showed normocytic anemia (7 g/dL), confirming the diagnosis of acute anemia and active bleeding. White blood cells and platelet counts were normal. No renal function impairment or electrolyte disorders were found.

Emergency resuscitation was performed, establishing two large-bore intravenous lines, and administering crystalloid IV fluids associated with a continuous IV infusion of proton pump inhibitors. Additionally, a transfusion comprising two units of packed red blood cells and one unit of fresh frozen plasma was conducted. To further assess the situation, an upper gastrointestinal endoscopy was scheduled within the next 6 h. Since the patient exhibited no signs of acute distress, we decided to perform an emergency CT scan for a more comprehensive assessment of the mass developed in the left hemi abdomen. The urgent abdominal scan revealed a large, exophytic mass of the gastric antrum, measuring 18 × 14 × 16 cm, hypodense with discernable areas of necrosis. No local or distant lymph node involvement was observed (Fig. 1).

Two hours into resuscitation, the patient became confused with a

Glasgow coma scale of 9/15, tachycardia of 130 BPM, and a drop in blood pressure of 90/50 mmHg resistant to fluid resuscitation. Our immediate course of action was to stabilize the patient using vasoactive drugs and perform an urgent laparotomy to control hemostasis. The patient underwent an emergency midline laparotomy, revealing a 20-cm exophytic mass in the anterior gastric wall of the antrum. No suspicious lymph node invasion or visible distant metastasis were observed (Fig. 2). We conducted an en-bloc wedge resection of the exophytic mass without effraction, using a surgical stapler. Reinforcement of the resection site was performed via resorbable stitching. In the post-operative course, the patient was admitted to the ICU and discharged 5 days later with an uneventful post-operative course.

The anatomopathological report confirmed the diagnosis of gastric GIST measuring 20 cm with a moderate risk of recurrence according to the Miettinen classification and a mitotic count of 2/50 high-power fields. Immunohistochemistry showed CD 117 and Dog 1 diffuse positivity (Fig. 3).

After a multidisciplinary hearing, the patient underwent adjuvant therapy based on a tyrosine kinase inhibitor for 3 years. No signs of local recurrence or distant metastasis were recorded during follow-up.

3. Discussion

GISTs stand out as the most prevalent form of soft tissue sarcoma, capable of arising anywhere in the digestive tract [5]. With an average size of 7 cm, GISTs measuring more than 10 cm are considered giant tumors [6]. The clinical picture of gastric GISTs is diverse and unspecific, mostly depending on tumor's location and size upon presentation [7]. While this tumor can manifest through various symptoms, gastrointestinal bleeding, in the form of melena or hematemesis, is the most common feature, accounting for 30 to 40 % of all the reported symptoms

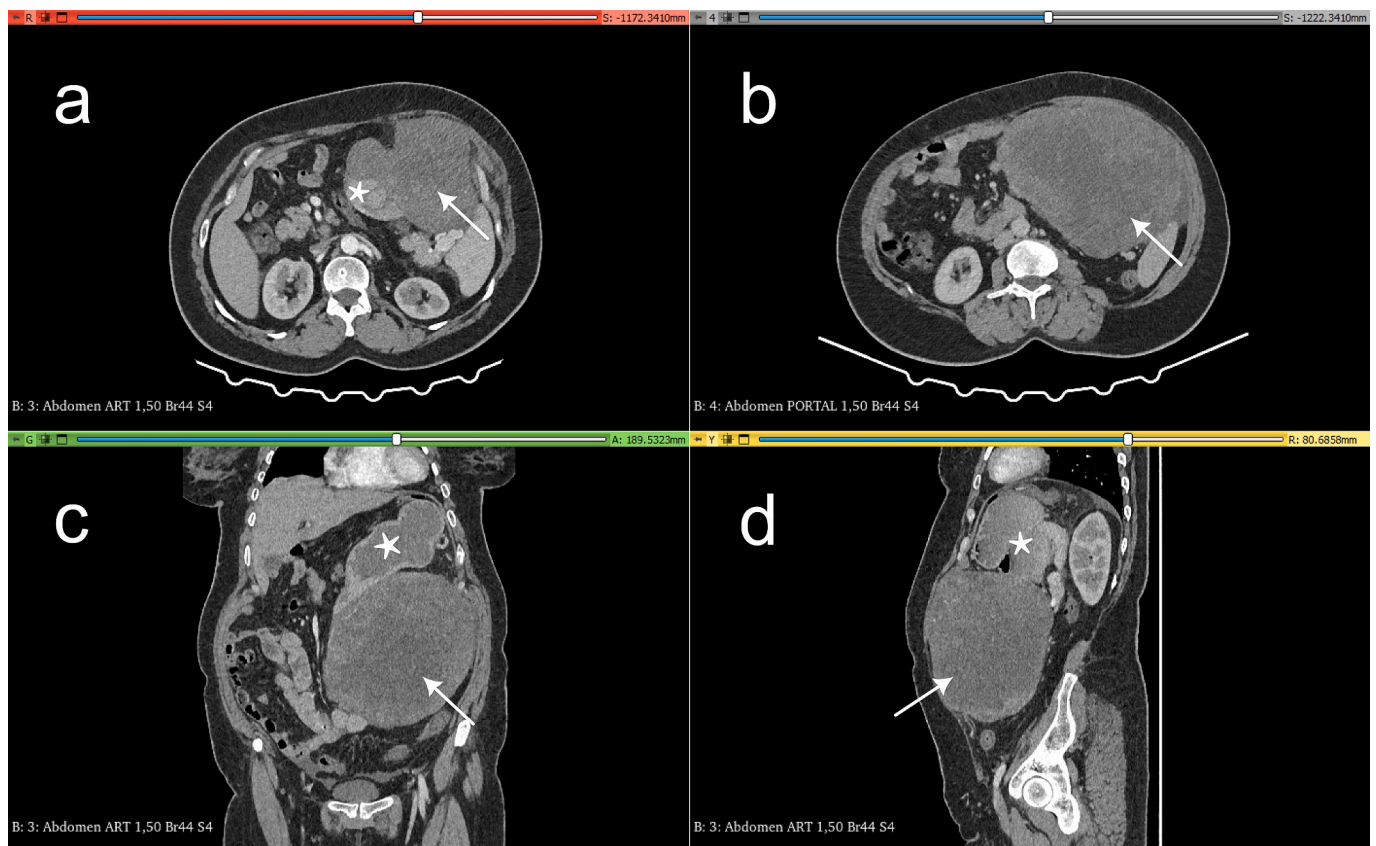


Fig. 1. Preoperative CT scan. (a) Axial view arterial phase, (b) axial view portal phase, (c) coronal view arterial phase, and (d) sagittal view arterial phase showing a sizable tumor (White arrows) attached to the stomach (White stars), with heterogeneous density. No active bleeding was detected on this CT.

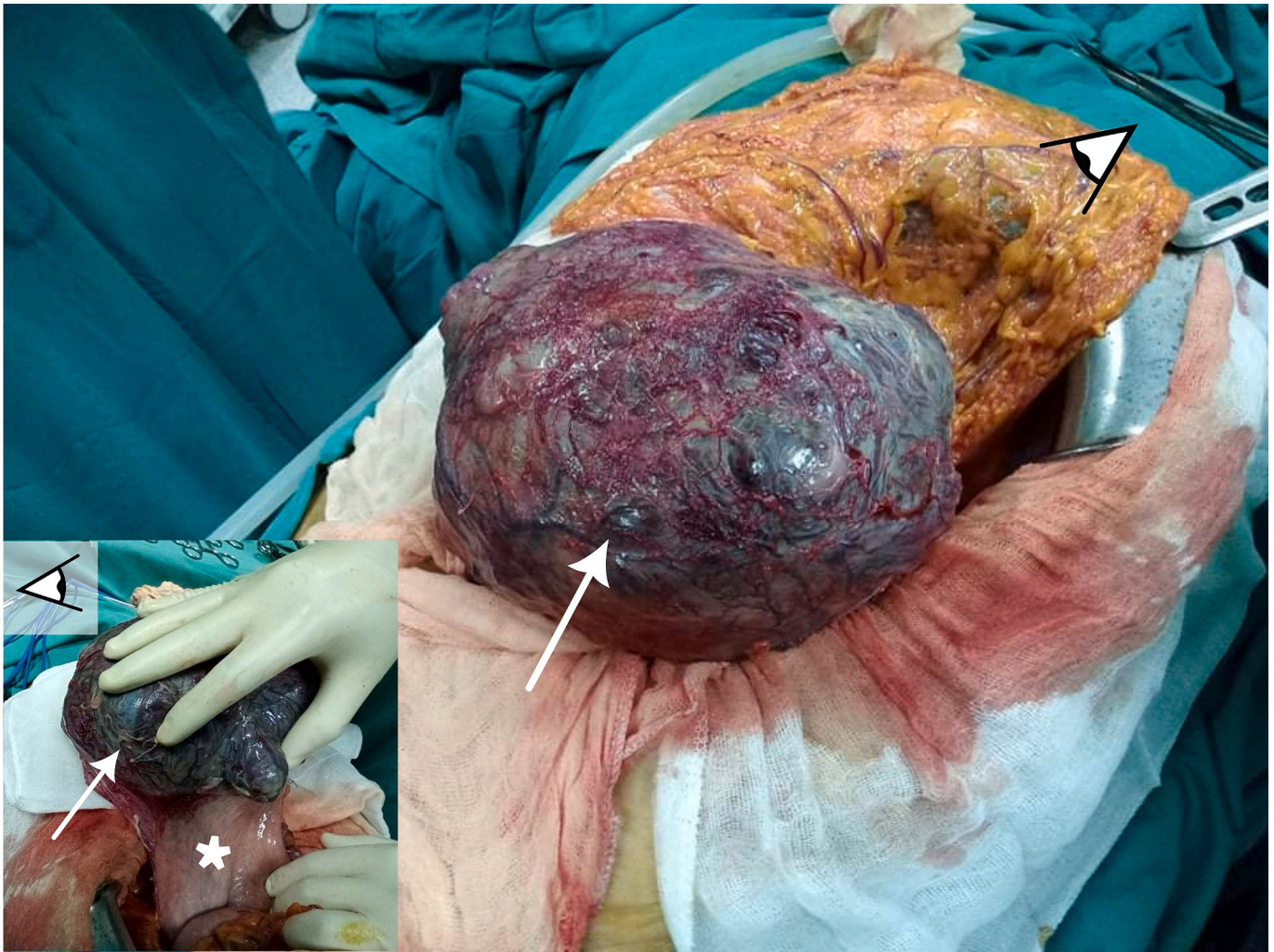


Fig. 2. Intraoperative findings. A huge tumor (White arrows) with rich vascularity was found. The tumor was attached to the antrum of the stomach (White star).

[8].

Although patients with gastric GISTs are mostly referred for gastrointestinal bleeding, instances of life-threatening acute hemorrhage requiring emergency surgical management are rare, with the literature mainly confined to isolated case reports [8]. The genesis of intraluminal bleeding in gastric GISTs results from a complex interplay of factors. Tumor-related factors include prevalent vascular characteristics and relative frailty of gastric GISTs, particularly those exceeding 6 cm in size [9,10]. Moreover, mucosal and submucosal damage due to tumor expansion, invasion of nutritional arteries, and cancer necrosis collectively contribute to vascular rupture and bleeding. Additional factors related to the tumor location involve the synergistic effect of acidic gastric juices and gastric peristalsis [9,10]. For cases involving acute gastrointestinal bleeding in gastric GISTs, upper gastrointestinal endoscopy is recommended within the first 12 h of admissions. This procedure serves to precisely locate the lesion, confirm its tumoral nature, provide the potential for endoscopic management, and assess post-endoscopic recurrence [11]. While an abdominal CT scan may not be the most suitable investigation for acute bleeding, it can unveil indicative signs in favor of gastric GISTs, such as well-defined soft tissue masses with areas of heterogeneous density attenuation from hemorrhage [12]. The gold standard of management of gastric GISTs is based on complete surgical resection with negative margins [5].

Unlike gastric adenocarcinomas, the surgical strategy for gastric GISTs differs, as it does not require lymph node dredging or extensive resection margins [13]. Laparoscopic management of gastric GISTs is an

option recognized for its efficiency, offering a better surgical outcome, shorter postoperative stay, and comparable oncological results to laparotomy [13]. While elective laparoscopic surgery for gastric GISTs has demonstrated favorable results, the evidence supporting its application for acute bleeding is more anecdotal than proven [14]. To date, there is no published meta-analysis assessing the feasibility and efficiency of laparoscopic treatment for acute bleeding in gastric GISTs. Existing literature reviews are largely limited to case reports [14–16]. Management of large gastric GISTs mandates diligent tissue handling and often involves substantial laparotomy incisions for specimen extractions [17]. When feasible, wedge resections represent the most suitable surgical intervention for bleeding gastric GISTs. This approach ensures efficient oncological treatment of the tumor, and its technical simplicity mitigates significant risk factors associated with post-operative mortality, including operative blood loss and prolonged ICU length of stay [18]. Wedge resection is a viable option for the removal of large tumors in the greater curvature and anterior gastric wall. However, technical challenges arise when dealing with tumors located in the lesser curvature, the posterior gastric wall, and the subcardial region [19]. Performing wedge resection for large tumors located in the posterior gastric wall and lesser curvature may result in a significant reduction of the gastric lumen, leading to gastric hourglass configuration [20]. Furthermore, conventional wedge resection for large tumors in the subcardial and fundic regions can lead to post-operative stenosis and functional complications [19]. Large gastric GISTs, due to their anatomical proximity, may exhibit distinct patterns of invasion into adjacent organs: tumors of

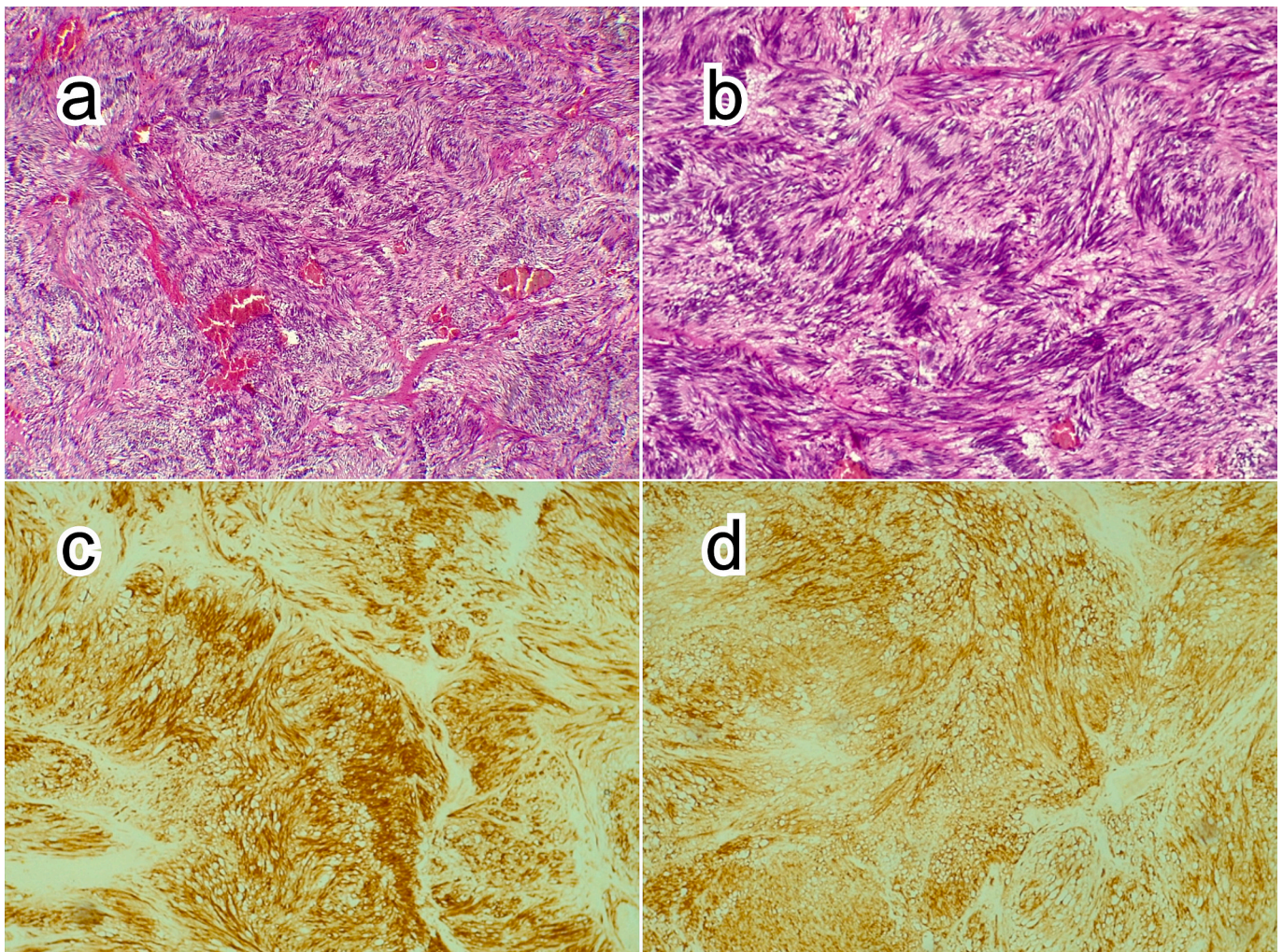


Fig. 3. Histopathology findings. (a) Proliferation in swirling bundles of spindle-shaped tumor cells (HE $\times 40$). (b) Proliferation in swirling bundles of spindle-shaped tumor cells (HE $\times 100$). (c) Positivity of tumor cells with the anti-CD117 antibody ($\times 100$). (d) Positivity of tumor cells with the anti-DOG1 antibody ($\times 100$).

the posterior wall may extend towards the pancreas [17]. Tumors of the lesser curvature may invade neighboring structures like the liver, gallbladder, and small intestine [21]. Additionally, tumors in the subcardial and fundic regions have the potential to invade the spleen [19]. In cases of locally advanced GISTs, multi-visceral resection is required to avoid effraction [22]. In these cases, neoadjuvant therapy must be considered. This approach enhances resectability and improves overall survival, potentially minimizing the need for complex surgeries [22]. In our presented case, midline laparotomy emerged as the only viable option for several reasons. Initially, the patient experienced hypovolemic shock during resuscitation, requiring effective hemostasis control. Additionally, the tumor's substantial size warranted an extensive laparotomy to retrieve the specimen and mitigate the risk of effraction leading to intraperitoneal metastasis. In the context of gastric GISTs, tumors exceeding 10 cm are deemed to carry a moderate to high risk of recurrence, contingent upon their mitotic index [2]. Although surgical management is considered as the primary curative measure for gastric GISTs, the incorporation of adjuvant therapy, based on tyrosine kinase inhibitors, is crucial in cases of moderate and high-risk recurrence as it improves relapse-free survival [2]. Our case, however, presents an intriguing deviation from recent literature reviews that commonly link large-size gastric GISTs with a high mitotic index [2]. Notably, our anatomopathological examinations revealed a massive tumor, yet a surprisingly low mitotic index. Given the tumor's dimensions, exceeding 10 cm, it falls into the moderate risk of recurrence category, warranting

a prescribed adjuvant treatment for 3 years.

4. Conclusion

For large bleeding gastric GISTs, open surgery becomes imperative. The approach to managing these tumors is contingent upon the technical intricacies dictated by the tumor's specific location within the stomach. The surgical procedure requires meticulous execution to prevent inadvertent tumor rupture. Despite their substantial size, these tumors exhibit a favorable prognosis, particularly when complemented by adjuvant therapy, which proves beneficial for patients at moderate to high risk of recurrence.

CRediT authorship contribution statement

Anis Hasnaoui: Conceptualization, Writing-Reviewing and Editing. Racem Trigui: writing-Original draft preparation. Mohamed Ben Hassine: Data curation. Salma Kacem: Writing-Reviewing. Houda Bellamine: Data curation. All authors read and approved the final manuscript.

Declaration of competing interest

The authors declare that they have no competing interests.

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Consent of publication

A written consent was obtained from the patient to publish this case report.

Registration for research study

N/A.

Ethical approval

Ethical approval was deemed unnecessary by our institutional ethical committee, as the paper is reporting a single case that emerged during normal practice.

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Guarantor

Dr. Hasnaoui Anis.
Dr. Trigui Racem.

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