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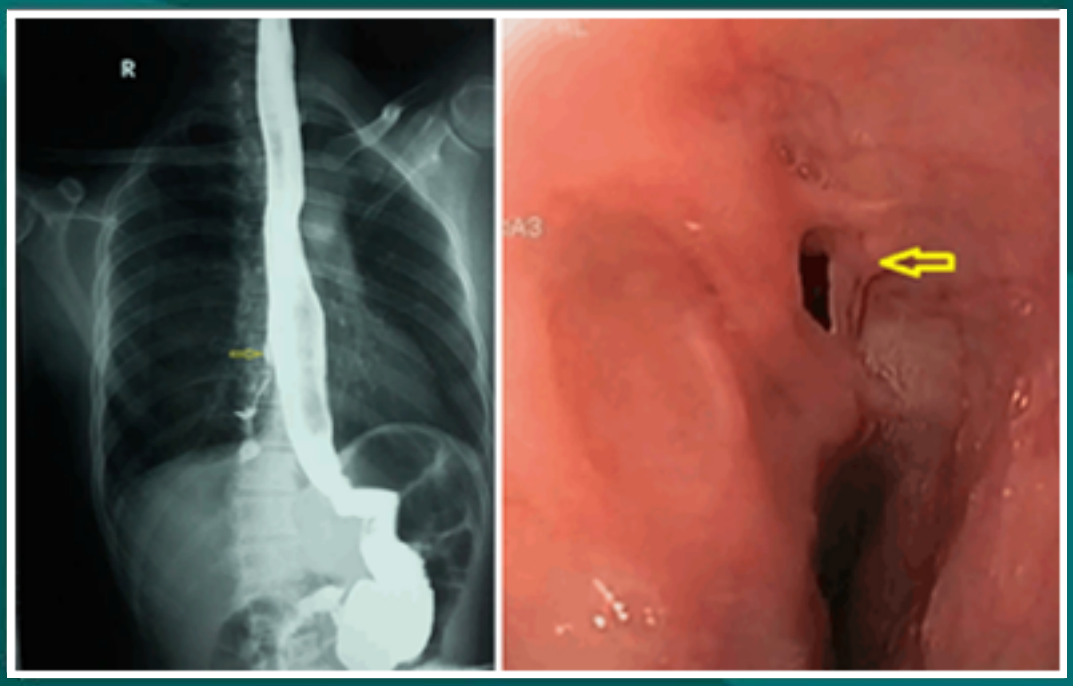

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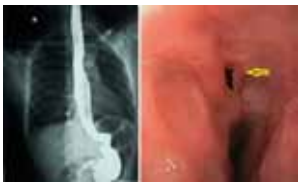

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CASE REPORT OPEN ACCESS | PEER REVIEWED

Dramatic response to HAART in HIV-induced hemophagocytic lymphohistiocytosis

Karan K. Topiwala, Ellen F. Eaton, Ricardo A. Franco

ABSTRACT

Introduction: Hemophagocytic lymphohistiocytosis (HLH) is a difficult diagnosis to make and carries with it a high mortality rate, yet it is treatable. Case Report: We present a 48-year-old African-American male, recently diagnosed with AIDS who had not yet initiated HAART (highly active antiretroviral therapy). He presented with high fever and altered mental status with progressive liver failure and worsening cytopenias. He showed no response to broad spectrum anti-microbial coverage and no infectious etiology could be established. There was no evidence of an opportunist infection or hemophagocytosis (HPC) on a subsequent bone marrow biopsy. In view of his rapid clinical deterioration with multi-organ failure, in the absence of an infectious cause and a stable CD4 cell count, he was diagnosed with HIV induced HLH using the modified-2009 criteria. He was started on HAART and showed a dramatic response with clinical improvement over the next three days, without the need to resort to immunosuppressive agents. The patient was discharged two weeks later, and has remained free of problems over the last eight months. Conclusion: This case report summarizes the importance of an early diagnosis of HIV induced HLH and the importance of early HAART initiation.

Keywords: Hemophagocytic lymphohistiocytosis (HLH), Hemophagocytosis, Highly active antiretroviral therapy (HAART) Hepatitis B, Herpes simplex virus (HSV), T lymphocytes

How to cite this article

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INTRODUCTION

Hemophagocytic lymphohistiocytosis (HLH) is a life-threatening syndrome characterized by an unregulated activation of antigen presenting cells, T lymphocytes, and macrophages, without their appropriate down-regulation by natural killer and cytotoxic cells. Although the frequency is unknown, HIV alone or in combination with infection or malignancy has been recognized as a cause of HLH since 1992 [1]. HLH in context of HIV can occur with acute or chronic HIV or even after initiation of HAART [2]. A dramatic resolution of HLH after the initiation of highly active antiretroviral therapy (HAART), as in our case report, is only infrequently reported. We thus hope to underscore the importance of rapid diagnosis and early initiation of HAART to treat the immune activation of HLH and the underlying viral process.

CASE REPORT

A 48-year-old African-American male presented with altered mental status and one week of fever, confusion and visual-tactile hallucinations. He reported malaise and subjective fever since one month. He had lost 40 pounds over the last two years which prompted a diagnosis of HIV/AIDS, made two months prior to his recent presentation. At that time his viral load was 630,000 copies/mL and CD4 count was 24/mm^3. He had not yet initiated antiretroviral therapy and was on bactrim prophylaxis for *Pneumocystis pneumonia*. His only other medication was hydrochlorothiazide for hypertension, which was well controlled. He reported being sexually active with men. Physical examination was significant for fever of 103°F and a regular tachycardia. He was alert to person, place and location but deferred other questions to his mother due to confusion. There were no focal neurological findings. His initial laboratory workup showed pancytopenia (WBC 2.5x10^3/µL, HCT 21, platelets 145x10^3/µL) with markedly elevated serum ferritin (51,376 ng/mL, normal: 23–336 ng/mL) levels along with raised serum transaminases (ALT-450 IU/L, normal:15–58 IU/L; AST-790 IU/L, normal:14–40 IU/L) and normal alkaline phosphatase-109 IU/L (normal: 39–117). Routine cerebrospinal fluid (CSF) analysis, neuroimaging and chest X-ray were within normal limits.

An initial differential diagnosis included, disseminated histoplasmosis, hemophagocytic lymphohistiocytosis (HLH) due to HIV, leptospirosis, fulminant viral hepatitis from herpes simplex virus (HSV) or hepatitis B and disseminated Mycobacteriosis. Given the acuity of his presentation he was given an initial empirical coverage with vancomycin, piperacillin and tazobactam, acyclovir and amphotericin B for suspected sepsis and hepatitis. However, there was little improvement in his clinical status the following day with worsening pancytopenia (WBC, HCT and platelets are 1.9 x10^3/µL, 25 and 90x10^3/µL respectively) and liver function (ALT and AST rose to 1500 and 2000 IU/L, respectively with an INR at 2.2 (normal: 1). Additionally, he developed renal failure (serum creatinine 1.7 mg/dL, normal: 0.5–1.2 mg/dL). A comprehensive infectious workup including a viral hepatitis panel and others (CSF culture and cytology, CSF-AFB, HSV PCR and IgM, CMV antigen, HHV-6 DNA, adenovirus DNA, parvovirus PCR, VDRL, leptospirosis antibody, Bartonella IgG and IgM, AFB blood isolators, cryptococcal antigen, urine histoplasma antigen, blood and urine cultures, stool giardia and cryptosporidium tests) were checked and found to be negative. A bone marrow biopsy was subsequently performed to look for any evidence of an opportunistic infection or hemophagocytosis. The results were indicative of HIV/AIDS related changes including plasmacytosis, megaloblastic change, megakaryocyte hyperplasia, reticulin fibrosis, stromal damage, histiocyte hyperplasia, and hypercellularity (Figure 1).

In the absence of an infectious etiology, a diagnosis of HLH was made using the 2009 criteria (Table 1) and the patient was started on HAART (raltegravir, emtricitabine and tenofovir). He improved dramatically within three days of initiating HAART. His agitation resolved along with improvement in his fever and liver function tests. He was discharged two weeks after starting HAART. He is being followed in the clinic and is doing well, eight months later. Below is a figure demonstrating his laboratory trends after HAART initiation (Figure 2).

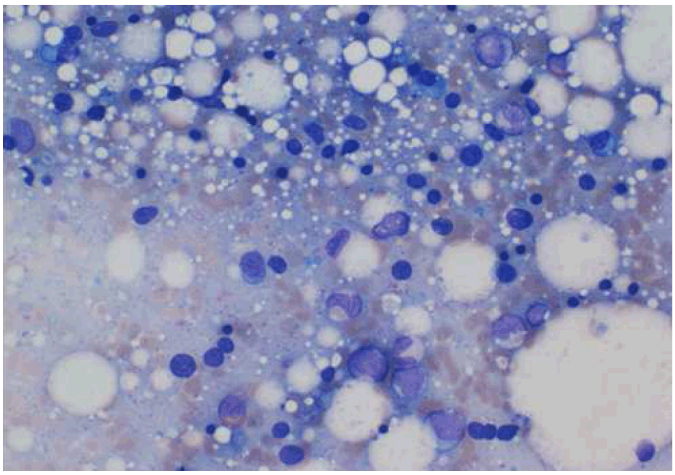

Figure 1: Slightly hypercellular marrow (50–60%) with trilineage hematopoiesis. Stromal changes consistent with history of HIV/AIDS effect on marrow: plasmacytosis, megaloblastic change, megakaryocyte hyperplasia, reticulin fibrosis, stromal damage, histiocyte hyperplasia, and hypercellularity. No morphologic evidence of malignancy. No evidence of opportunistic infection (special stains for fungus, cytomegalovirus, herpes simplex virus (HSV), mycobacteria, spirochetes and *Bartonella* were negative.

Table 1: 2009 HLH diagnostic criteria [9]

Identification of a HLH-associated gene mutation: (PRF1, UNC13D, STX11, STXBP2, Rab27A, SH2D1A, or BIRC4)
or
Three of the following four clinical criteria:
(i) Fever ≥38.5°C (ii) Splenomegaly (iii) Peripheral blood cytopenia: at least two cell lines (iv) Hepatitis
and
one of the following four laboratory criteria:
(i) Hemophagocytosis in bone marrow, spleen, lymph node, or liver (ii) Ferritin >500 ng/mL (iii) Elevated soluble CD25 (iv) Low or absent NK cell activity
Supportive criteria:
(i) Hypertriglyceridemia (ii) Hypofibrinogenemia (iii)Hyponatremia

Abbreviations: [HLH Hemophagocytic lymphohistiocytosis].

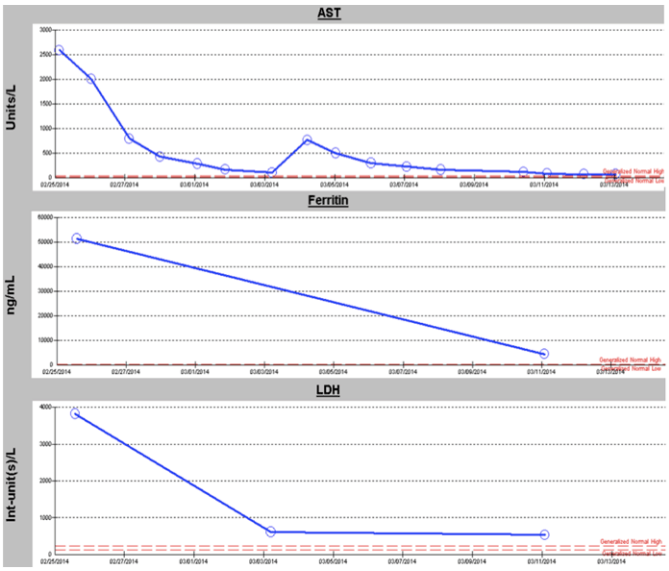

Figure 2: Laboratory trends after highly active antiretroviral therapy (HAART) initiation. Note the extremely elevated levels of serum ferritin and its rapid decline along with aspartate aminotransferase and lactate dehydrogenase levels, after the initiation of HAART.

DISCUSSION

HLH is a life-threatening syndrome characterized by an unregulated activation of antigen presenting cells, T lymphocytes, and macrophages, without their appropriate down-regulation by natural killer and cytotoxic cells. The major manifestations include sepsis, fever, cytopenias, splenomegaly and hepatitis. The activated macrophages in HLH may also phagocytize host cells producing hemophagocytosis (HPC). HLH can be primary due to genetic predisposition or secondary to an infectious or rheumatologic processes. When HLH occurs in the setting of rheumatologic disorders such as lupus or rheumatoid arthritis, it is termed Macrophage Activation Syndrome (MAS). EBV was the most common infectious trigger in one study of 250 suspected HLH-cases (28% of the cases), followed by histoplasmosis (19%), cytomegalovirus (14%), bacteria (4%), mycobacteria (4%), blastomycosis (5%) [3]. Although the frequency is unknown HIV alone or in combination with an infection or malignancy has been recognized as a cause of HLH since 1992 [1]. Neidt et al. published autopsy reports of 56 AIDS patients in 1985, 20% of whom had HLH [4]. In 1997, Grateau et al. found a 0.6% incidence of HLH among HIV infected persons in one clinic, but this was likely an under-representation as it relied on bone marrow aspiration [5]. Of HIV infected patients with HLH, 56/58 had an additional hematologic or infectious etiology in one review: tuberculosis, cytomegalovirus and Hodgkin's disease were the most common associated conditions [1]. Several case reports, including ours, report HIV infection alone as the trigger for HLH, but the incidence is unknown [6]. HLH in context of HIV can occur with acute or chronic HIV or after initiation of HAART [2]. In advanced AIDS, HLH is usually secondary to infection or malignancy [1, 5]. Diagnosis of HLH in HIV infected patients is also limited by poor sensitivity of bone narrow. Bone marrow biopsy is thought to be 60% sensitive [7] in all populations, and 63% sensitive in HIV infected patients [1]. Other reports of severe HIV-related HLH have failed to demonstrate biopsy-proven HPC [8]. As in our case, it is common for patients to present with only three or four of the eight diagnostic criteria, as initially laid down in 2004 [9]. Thus, experts proposed a modification (Table 1) necessitating only three of four physical findings and one of four laboratory derangements [10]. Lastly, treatment of HLH in the HIV infected patient should focus on treating the underlying cause. The HAART potentially plays an important role in reverting clinic-pathological changes of severe HLH as exemplified in our case report. The importance of HAART is also suggested, by the dramatically reduced mortality of HLH in the post-HAART era (31%) compared to the pre-HAART era (50–100%) [1]. Severe HLH usually requires conventional immunomodulatory agents such as corticosteroids, cyclosporin, intravenous immunoglobulin (IVIG), anti-thymocyte globulin and TNF antagonists. Refractory cases respond well to Alemtuzumab, where it is used as a bridge to allogenic stem cell transplantation [11]. Two case reports of HIV related HLH with significant splenic involvement have also benefitted from splenectomy [8]. In our case, HAART alone could provide complete recovery of multi-organ failure without the use of further immunosuppressive therapies.

CONCLUSION

Our case highlights chronic HIV alone as a trigger for hemophagocytic lymphohistiocytosis (HLH). It stresses the use of more liberal diagnostic criteria as proposed in 2009. It further underscores the importance of an early treatment initiation with highly active antiretroviral therapy (HAART) in the HIV associated HLH cases, as it may potentially prevent the use of immuno-modulatory agents, in this immune-deficient population and provide complete recovery even in severe cases.

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Author Contributions

Karan K. Topiwala – Acquisition of data, Analysis and interpretation of data, Drafting the article, Final approval of the version to be published
Ellen F. Eaton – Conception and design, Acquisition of data, Analysis and interpretation of data, Critical revision of the article, Final approval of the version to be published
Ricardo A. Franco – Conception and design, Analysis and interpretation of data, Critical revision of the article, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest
Authors declare no conflict of interest.

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SUGGESTED READING

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ABOUT THE AUTHORS

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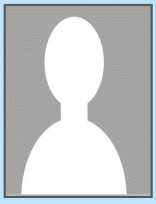

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CASE REPORT OPEN ACCESS | PEER REVIEWED

A rare case of congenital esophagobronchial fistula in an adult

V. YaminiChitra, K. N. Paramesh, Alamelu Haran, Nitin D. Tengli

ABSTRACT

Introduction: Congenital esophagobronchial fistulas in adults are extremely rare, acquired fistulas being more common. The aim of this paper was to present a rare case of congenital esophagobronchial fistula in a 37-year-old male of type II Braimbridge's classification and to emphasize on the diagnostic modality of choice and the appropriate mode of treatment. Case Report: A 37-year-old male presented with chronic cough with ingestion of food, especially liquids of 13 years duration and recent onset hemoptysis. He was evaluated with upper gastrointestinal endoscopy, bronchoscopy, computed tomography scan of chest and the definitive test was barium swallow which confirmed it. He underwent transthoracic excision of the fistula with repair of both esophageal and bronchial ends. A peroperative endoscopy helped localization of the tract. Postoperative outcome was excellent with no leak and patient is totally asymptomatic after 12 weeks of surgery. Conclusion: Congenital esophagobronchial fistulas in adults, due to insidious nature need high index of suspicion as early diagnosis by barium swallow and surgical treatment gives excellent results. Peroperative endoscopy is mandatory to localize the tracts, helps do an intraoperative leak test and avoid esophageal stenosis during repair.

Keywords: Barium swallow, Congenital esophagobronchial fistula, Peroperative endoscopy, Transthoracic excision

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INTRODUCTION

Fistula between esophagus and bronchus may be congenital or acquired. Acquired fistulas can be inflammatory, traumatic or neoplastic. Congenital esophago bronchial fistula (EBF) with atresia present immediately in infancy, are sudden and are diagnosed early and treated [1]. Congenital EBF without atresia are insidious, can present in adulthood also or missed often especially if they communicate with a lobar bronchus [1].

To differentiate between congenital and acquired EBF in an adult is usually difficult [2]. Adult EBF may present acutely with sudden respiratory distress, chronically with repeated respiratory infections or can be totally asymptomatic. Less than 200 cases of adult EBF are reported so far in literature [2].

Herein, we report a rare case of 37-year-old male with congenital EBF without atresia (type 2 according to Braimbridge's classification) [1] which was diagnosed by barium esophagogram. Patient underwent transthoracic fistulectomy with the repair of both esophageal and bronchial ends. Postoperative outcome was successful with complete resolution of symptoms and closure of fistula.

CASE REPORT

A 37-year-old male presented to our department with history of cough with expectoration immediately after taking food, especially liquids for last 13 years. He had two bouts of hemoptysis in the last 15 days which had made him seek medical attention. Clinically, chest on auscultation had crackles in right infra-scapular region. Chest X-ray revealed patchy opacities in the right lower zone paracardiac region. Computed tomography (CT) scan of thorax revealed consolidation in posterior segment of the right lower lobe with no evidence of lung sequestration or cyst. Upper gastrointestinal endoscopy showed a fistulous opening in the mid esophagus 32 cm from the incisor teeth. It did not show any evidence of malignancy, granulomatous disease or any other acquired basis for the fistula. Simultaneously, methylene blue was injected into the fistulous tract and bronchoscopy was done which was normal. Barium swallow showed fistulous communication between mid esophagus and right lower lobe bronchus at lower border of T7 with barium passing downward into the right lung (Figure 1).

Preoperative evaluation done included pulmonary function tests, echocardiogram and electrocardiogram. Incentive spirometry was started for better postoperative outcome. Preoperatively, 1 fr size guide wire was introduced endoscopically into the fistulous tract to aid identification of fistula. Patient underwent right posterolateral thoracotomy. There were minimal adhesions around the fistulous site which was identified about 5 cm below the level of entry of azygos vein into superior vena cava. Azygos vein was isolated, ligated and cut to aid esophagus to be encircled, to localize the fistulous tract. As the guide wire was not palpable through the tract, intraoperative endoscopy was done and fistula tract location was confirmed. The fistulous tract was dissected, it was 10 mm long. The tract was excised, both the esophageal and bronchial ends were healthy and were closed with 4-0 Vicryl. An intraoperative leak test was done using endoscopy which confirmed the integrity of the repair and an intercostal drain placed (Figure 2–4).

Postoperative period was uneventful. Intercostal drain was removed on postoperative day-4 after a barium swallow to confirm that there was no leak and patient was discharged on postoperative day-6, after starting oral soft diet (Figure 5).

Histopathology showed the mucosa to be lined by stratified squamous epithelium. There was no evidence of inflammation, granuloma or carcinoma confirming the congenital nature of the fistula. Patient is totally asymptomatic after 12 weeks of surgery.

DISCUSSION

The majority of EBF in adults are acquired. The usual causes are inflammatory like tuberculosis, trauma and neoplasms. The EBF was first described as early as in 1916 by Heiderich [3]. Congenital EBF are rare, three times more common on the right than the left [4].

According to Braimbridge's classification, type I is a fistula associated with a wide-necked congenital diverticulum of the esophagus with inflammation at the tip. Type II, which is the simplest and most common, consists of a short tract running directly from the

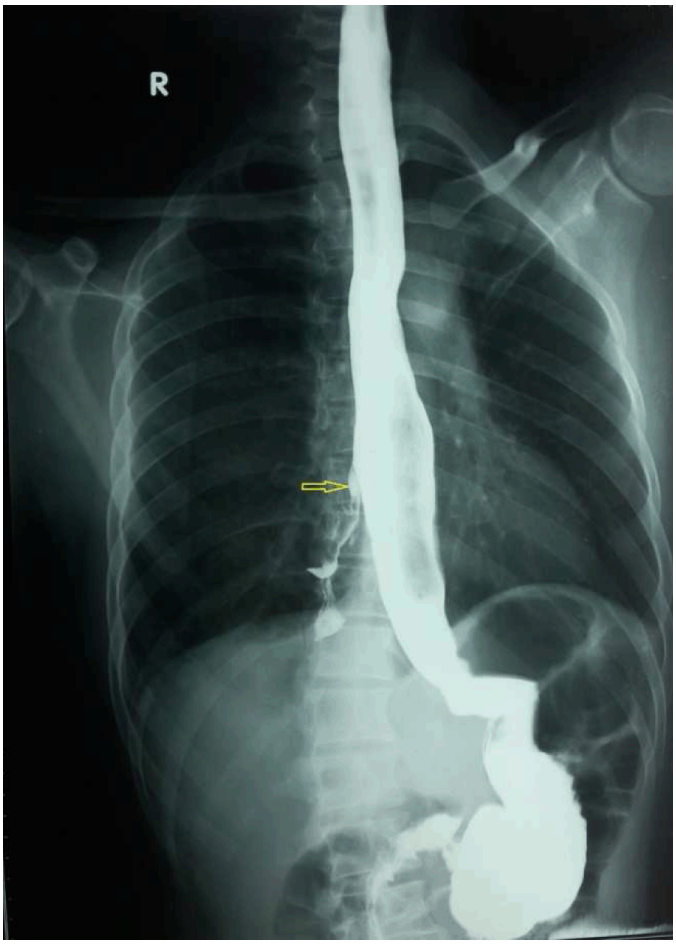

Figure 1: Barium esophagogram showing esophagobronchial fistula between the middle third of the esophagus and the right lower bronchus with downward passage of barium (shown with arrow).

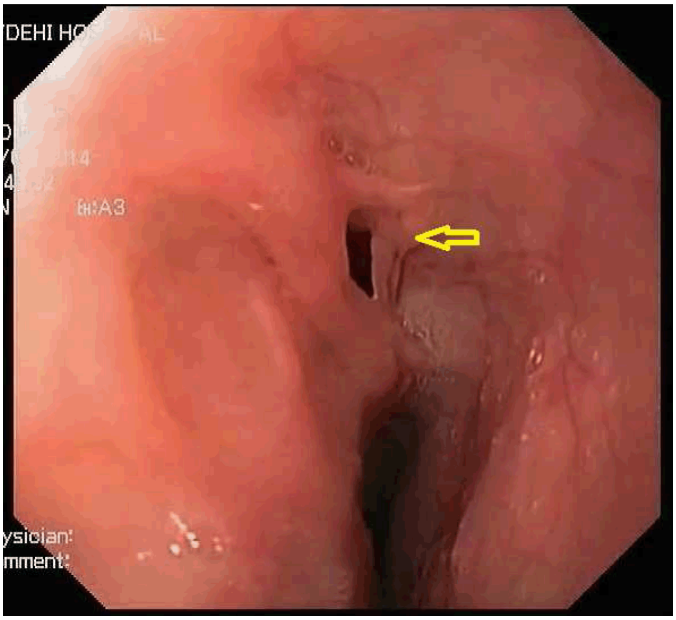

Figure 2: Endoscopic findings: The fistula opening in the middle esophagus (arrow).

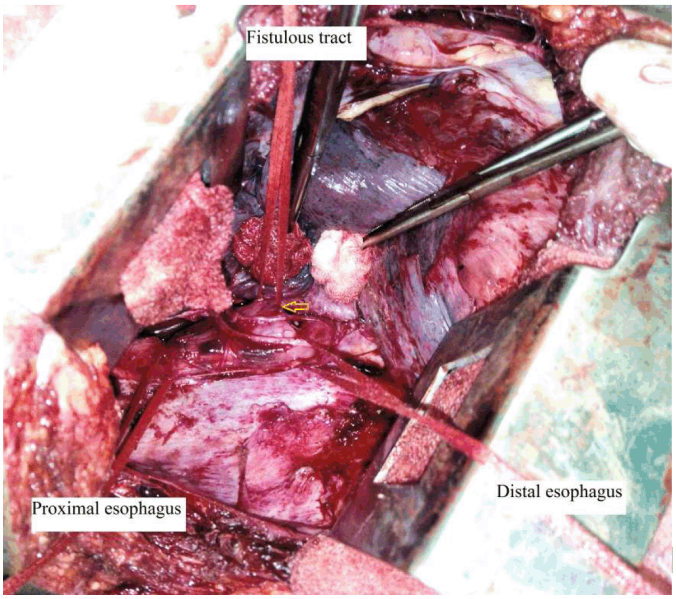

Figure 3: Intraoperative photograph showing bronchoesophageal fistula. The fistula opening in middle esophagus (shown with arrow), proximal, distal esophagus and fistulous tract is also looped.

esophagus to the lobar or segmental bronchus. In type III, the fistulous tract connects the esophagus to a cystic pulmonary change, and in type IV a fistula runs into a sequestered pulmonary segment. The patient described here is of type 2 according to this classification [1].

Criteria for congenital EBF are suggested pathologically by the absence of surrounding inflammation and adherent lymph nodes along with the presence of a mucosa and a definitive muscularis mucosa within the fistulous tract. Surgically, uncomplicated and easy dissection of the fistula and absence of inflammation suggests a congenital fistula [5].

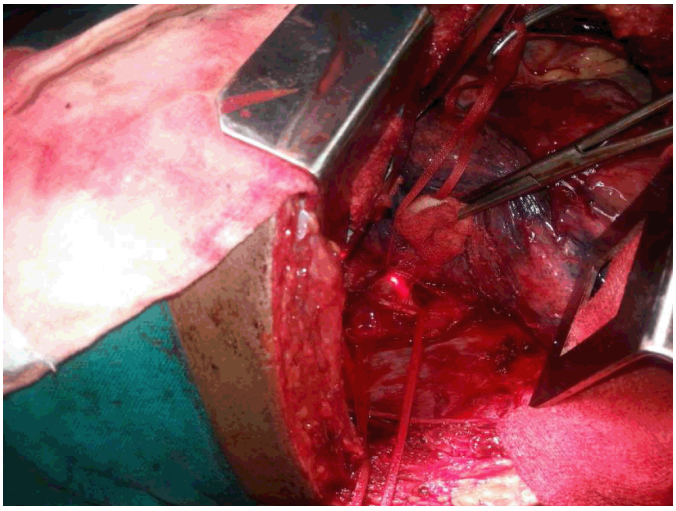

Figure 4: Intraoperative photograph with intraoperative upper gastrointestinal endoscopy (light of endoscopy is visible through the tract).

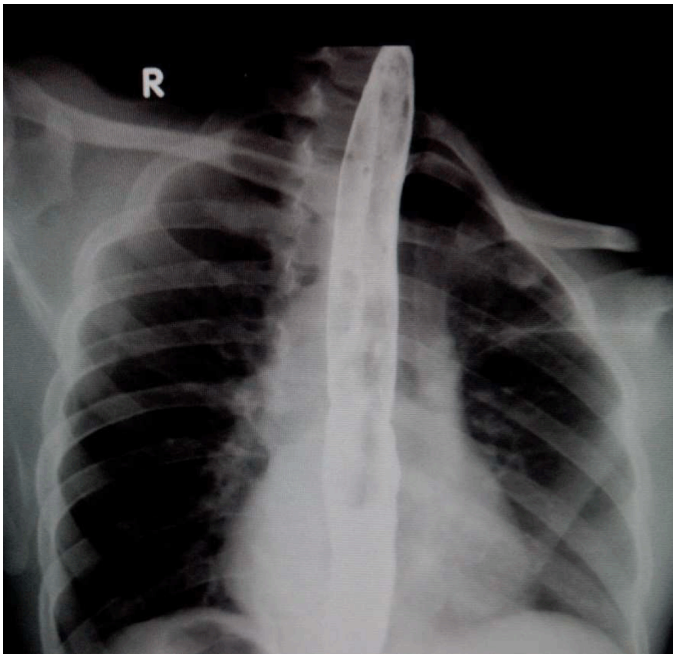

Figure 5: Postoperative barium esophagogram showing normal esophagus and no evidence of fistula.

Reasons for delay in the onset of symptoms and presentation in the adult may be due to

(i) A fistula tract which runs upwards and may close during swallowing. But in our patient barium was passing downward during the swallow.

(ii) A membrane which later ruptures.

(iii) A fold of esophageal mucosa which overlaps the orifice but subsequently becomes less mobile [1].

Both sexes are affected almost equally (male 53% and females 47%) [6].

Symptoms are often intermittent, due to chronic bronchopulmonary infection. They are chronic cough (96%), pneumonia (56%), hemoptysis (17%). Ohno's sign characterized by symptoms of strangulation and

paroxysmal coughing with swallowing liquids occurs in the presence of a very large communication [7].

The diagnosis is usually made by barium esophagogram [1]. A thin barium is given, in a position where patient gets most of the symptoms. Endoscopy and bronchoscopy should be done but will not always demonstrate the opening in esophagus and bronchus [4]. Ramo et al. have shown that bronchoscopy is negative in 67% of cases and esophagoscopy in 40% [4]. Bronchoscopy can be normal, if the communication is in the distal segmental portion of the tract as in our case [7]. Dynamic bronchoscopy with pediatric bronchoscope may help [7]. Right bronchus is most commonly affected [7]. Computed tomography scan may help to rule out type 4 fistulas and aortography can document the sequestrated lung [8] and aid in pulmonary resection if needed. Peroperative endoscopy helps accurately in identifying the tracts [2], to confirm patency of the esophagus if the repair is very proximal or on the esophageal wall itself, as in wide short tracts and also aids in doing an intraoperative leak test as done in our case.

Thoracotomy and resection of the fistulous tract with primary repair of both the bronchial and esophageal defects with 4-0 Vicryl the ideal treatment of choice. Interposition of pleural/diaphragmatic flap helps reduce the recurrence. In our case, it was not done as the tract was long and the bronchial and esophageal tissue post repair were healthy. The diseased lung tissue and sequestrated lobes if any should be resected at the same time.

Postoperative barium swallow helps document the healed defect in the esophagus.

Endoscopic management by submucosal dissection and isolation of the fistulous tract with clipping has been tried in tracheoesophageal fistula but has been unsuccessful [9]. Other endoscopic techniques like histoacryl glue injection have been tried in recurrent congenital trachea esophageal fistulas, or when patient has refused surgery with varied results.

Thoracoscopic repair assisted by peroperative endoscopy and stapling of the fistulous tract by surgeons experienced in minimally invasive surgery can reduce the morbidity associated with open thoracotomy [10].

Either thoracoscopic or open thoracotomy repair with excision of the fistulous tract and good repair of the esophageal and bronchial defects with tissue interposition when needed, gives permanent cure and relief of symptoms in the patients with EBF.

CONCLUSION

Congenital esophagobronchial fistulas in adults, due to rare occurrence and insidious nature need high index of suspicion as early diagnosis by barium swallow and surgical treatment by open or minimally invasive technique gives excellent results.

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Author Contributions

V. YaminiChitra – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

K. N. Paramesh – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Final approval of the version to be published

Alamelu Haran – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Final approval of the version to be published

Nitin Tengli – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Final approval of the version to be published

Guarantor

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Conflict of Interest

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CASE REPORT OPEN ACCESS | PEER REVIEWED

Bilateral retrobulbar neuritis following cessation of ethambutol

Vivekanand Undrakonda, Yashodhara B. M., Sarita Gonsalves, Shashikiran U., Smita Kapoor

ABSTRACT

Introduction: Ethambutol related ocular toxicity, is a well-documented fact. In spite of this a regular visual acuity examination is missed during treatment and follow-ups. Case Report: A 35-year-old male diagnosed with tuberculosis of spine, who was on antitubercular drugs for more than one year presented with complains of acute diminution of vision in both eyes two months following cessation of ethambutol. Visual electrode potential (VEP) showed prolongation of latency of P100 bilaterally based on which a diagnosis of ethambutol induced bilateral retrobulbar neuritis was made. Patient was started on intravenous methylprednisolone 1 g/day for three days which was followed by 11 days of oral steroids (1 mg/kg) which was tapered off over next 15 days. On follow-up patient showed signs of improvement in visual acuity and visual fields over the next six months. Conclusion: Visual symptoms may revert if prompt action is taken that include discontinuation of ethambutol and supplementation of pyridoxine along with steroids like in our case. Pharmacovigilance on patients receiving antitubercular drugs for exact dosage, drug combinations and duration is necessary to avoid untoward complications.

Keywords: Antitubercular drugs, Ethambutol toxicity, Retrobulbar neuritis, Scotoma, Visual acuity

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INTRODUCTION

Acute onset of painless loss of vision in both eyes in a patient who is on antitubercular drugs pointing towards ethambutol toxicity, which is a well-documented fact. Ethambutol can affect the peripheral optic nerve fibers, causing defects in the peripheral visual field. Although optic neuritis is the most common side effect, retrobulbar neuritis is also a well-documented fact with involvement of either axial fibres or periaxial fibres [1, 2].

Dose related ethambutol related retrobulbar neuritis in patients who received it for more than two months was reported as 18% in patients receiving >35 mg/kg/day, 5–6% with 25 mg/kg/day and <1% with 15 mg/kg/day [3].

Rarely, it can cause visible retinal manifestations, including hyperemia and swelling of the optic disc, flame-shaped hemorrhages on the optic disc and in the retina, and macular edema. Other rarer side effects of ethambutol

include peripheral neuropathy, cutaneous reactions (rash, pruritus, urticaria, etc.), thrombocytopenia and hepatitis [4].

CASE REPORT

A 35-year-old male presented to our ophthalmology outpatient department with chief complaints of marked painless diminution of vision (left eye > right eye) over a period of three days. His best corrected visual acuity recorded was 6/36 in right eye and 1/60 in left eye (Snellen chart). Gross observations of the anterior segments of both eyes were normal except for ill-sustained pupillary response to light in both eyes. Dilated fundoscopy was unremarkable.

One year back, he was diagnosed with tuberculosis of spine (thoracolumbar vertebra) based on a history of chronic cough, weight loss, backache, loss of appetite and results of diagnostic tests such as hemogram showed markedly elevated erythrocyte sedimentation rate (ESR) of 99 mm/hr, hematocrit of 36.2% with a bone scan with Tc-99m MDP which revealed multiple osteoblastic changes over T7 to T12 with multiple bilateral rib involvement. Following which a magnetic resonance imaging scan (plain and contrast) of lumbosacral spine showed features suggestive of diffusely infiltrating disease, with a possibility of multifocal tuberculosis in same region (Figure 1). Other parameters such as blood glucose, chest X-ray (Figure 2), liver function tests and renal function test were normal. Sputum test results were negative for acid-fast bacilli. The patient was started on antitubercular (ATT) drugs-isoniazid 300 mg/day, rifampicin 600 mg/day, pyrazinamide 1500 mg/day and ethambutol 1200 mg/day with vitamin B6 20 mg/day. Two months after initiation of ATT patient underwent a detailed ophthalmic examination which included visual acuity examination, macular function tests, and visual fields, all of which were unremarkable. After three months of initiation of ATT with four drugs, patient was advised to stop pyrazinamide and ethambutol and continue on two drugs isoniazid and rifampicin (which comes as combination tablet) as part of continuous phase treatment. The patient was lost to follow-up for next nine months. On repeat, consultation after one year patient was found to be still on isoniazid, rifampicin (combination tablet) and ethambutol, although he was advised to stop ethambutol three months after initiation of ATT. A repeat MRI scan of spine was performed after one year after initiation of ATT showed signs of improvement following which ethambutol was stopped and patient was advised to continue on isoniazid 300 mg/day, rifampicin 600 mg/day, and vitamin B6 20 mg/day for 6 more months. Four months following stopping of ethambutol patient presented with gross diminution of vision in both eyes.

Investigations

Visual field assessment using Goldman perimeter showed presence of gross central visual field (CVF) defects with Red-green dyschromatopsia. Visual electrode potential (VEP) showed prolongation of latency of P100 bilaterally. Computed tomography scan of brain and MRI scan of optic nerve both with and without contrast were normal (Figure 3). The cANCA was negative. A cerebrospinal fluid examination could not be performed.

Treatment

Based on the findings of VEP patient was started on intravenous methylprednisolone 1 g/day for three days which was followed by 11 days of oral steroids (1 mg/kg) which was tapered off over next 15 days.

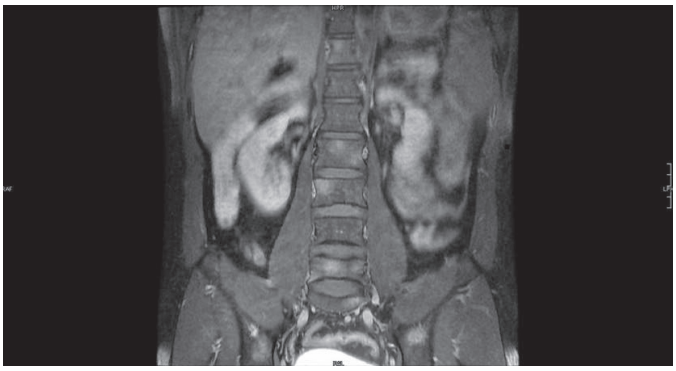

Figure 1: Magnetic resonance imaging scan of spine showing diffuse multifocal tuberculosis of T7 to T12.

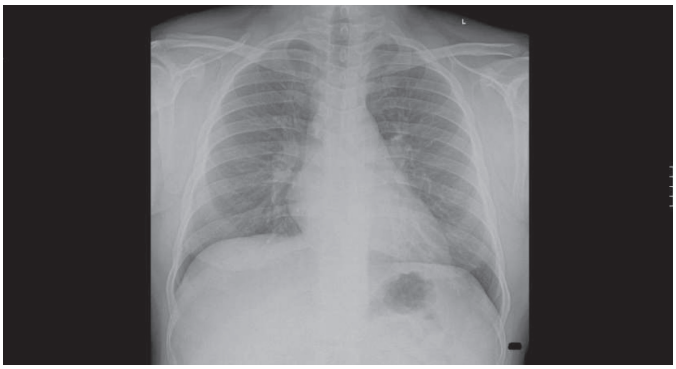

Figure 2: Normal chest X-ray.

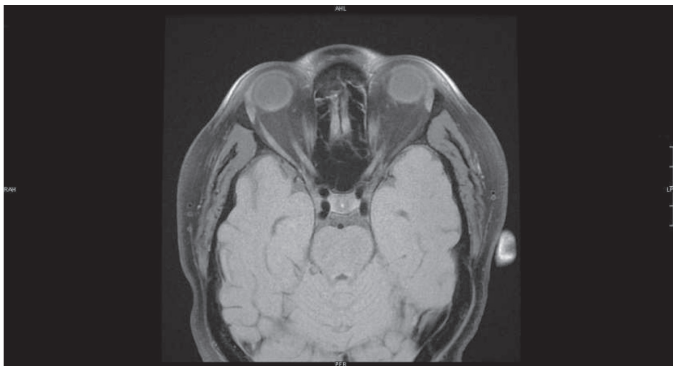

Figure 3: Normal magnetic resonance imaging scan of optic nerve.

Outcome and follow up

After one month, during review patient showed signs of improvement in visual acuity and visual fields. His visual acuity improved to 6/24 in right eye and 3/60 in left eye. Visual field assessment showed constriction in central visual field defects in the subsequent follow-ups.

DISCUSSION

Antitubercular drugs produce unwanted side effects, especially when used at high dosages and usually for periods of more than two months [5, 6]. Retrobulbar neuritis, optic neuropathy and chiasmopathy due to ethambutol is a known neurotoxic complication. The onset of visual symptoms can occur within a period of 10 days to 90 days after initiation of therapy [7–9]. Detailed ophthalmoscopic examination reveals a bilateral and often an unequal decrease in visual acuity, loss of color vision, bitemporal hemianopsia or centrocecal scotoma on perimetry. Fundoscopy generally shows bilateral disc hyperemia with blurred borders. Rarely, fundoscopy may be normal like in our case report. Methylprednisolone along with cessation of the drug is considered useful, as was seen by the visual improvement in our patient. However, the recovery is often incomplete.

Visual acuity, contrast sensitivity, and multifocal ERG are sensitive tests to detect ethambutol toxicity in subclinical stages and hence very useful tools for monitoring patients under ethambutol therapy for ocular toxicity [10]. MRI scans of the optic nerves and chiasm, with normal findings in toxic and/or nutritional optic neuropathy, could be useful to differentiate between bilateral centrocecal scotomas and compressive or infiltrative lesions of the optic chiasm.

After three months of initiation of ATT with four drugs, our patient was instructed to stop pyrazinamide and ethambutol and continue on two drugs isoniazid and rifampicin which comes as combination tablet as part of continuous phase treatment for one year. But unfortunately, patient misunderstood the instructions as two tablets continued isoniazid, rifampicin which comes as a combination tablet with another tablet ethambutol for next nine months and was never on follow-up during this period.

After one year when our patient came for follow-up, he was instructed to stop ethambutol. Four months following discontinuation of ethambutol, which remains the mainstay of treating ethambutol-associated ocular toxicity, our patient developed bilateral retrobulbar neuritis.

Isoniazid associated combination formulations are easier to administer and also may reduce medication errors. These formulations are means of minimizing inadvertent monotherapy. It is quite common for patients with tuberculosis to be taking a variety of other medications, hence combination therapy is preferred to monotherapy to improve compliance in patients with comorbid conditions. The combination of rifampicin, isoniazid, ethambutol, and pyrazinamide for two months followed by combination of rifampicin and isoniazid for a total period of 6, 9, 12 or 18 months is the most frequent protocol used for treatment of spinal tuberculosis [11].

Early detection of adverse effects of drugs, failure of treatment and emergence of drug resistance due to non-compliance could be overcome by establishing a human bond between the patient and the provider through DOTs. In the developing world, evidence from uncontrolled studies shows that the introduction of DOTS has increased completion of therapy and cure rates from 25–50% (with unsupervised treatment) to 80–90%, with relapse rates of less than 5% [12, 13]. Despite all the advantages of DOTS regimen, many orthopedic surgeons continue to give daily regimens. This is basically due to the fact that the efficacy of short-course intermittent therapy like DOTS regimen is not scientifically proven [14].

How could such complications be prevented: The first step is to identify patients in whom ethambutol is relatively contraindicated. These include patients who are unlikely to notice or describe visual symptoms, such as patients with dementia, mental retardation and children. Others include patients with pre-existing ophthalmological diseases with poor baseline vision. These patients should not be treated with ethambutol. The second step is to educate all patients treated with ethambutol on its side effects. Third step: Patients taking ethambutol should be instructed to discontinue the drug immediately at the onset of any visual symptoms and seek medical consult. Fourth step: When patients are prescribed combination therapy they should be closely monitored for compliance as well as side effects. The duration therapy for each drug should be defined and monitored. Physicians prescribing the drug should be aware of this and the drug should be used with proper patient education and ophthalmological monitoring especially when used in combination [15–19].

An immunological reason for bilateral retrobulbar neuritis was suspected and cANCA was performed to rule out other collagen vascular disorders and granulomatous conditions. Although few positive c-ANCA test results have been reported in patients with tuberculosis, Hodgkin's lymphoma, human immunodeficiency virus infection, nasal septal perforation, monoclonal gammopathies, and drug-induced Wegener-like disease. In this case, cANCA was negative [20–21]. Although the MRI scan showed no involvement of optic nerve, in view of positive VEP findings patient was started on methylprednisolone and oral steroids [22].

CONCLUSION

It is necessary that practitioners watch patients receiving antitubercular carefully for exact dosage, drug combinations and duration in order to avoid untoward complications. Visual symptoms may revert if prompt

action is taken that include discontinuation of ethambutol and supplementation of pyridoxine along with steroids like in our case. It is also vital that such cases are reported through active pharmacovigilance programs, so that we can prevent future errors.

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Author Contributions

Vivekanand Undrakonda – Substantial contributions to conception and design, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published
Yashodhara B. M. – Substantial contributions to conception and design, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published
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Smita Kapoor – Substantial contributions to conception and design, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

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CASE REPORT OPEN ACCESS | PEER REVIEWED

Atypical Takotsubo cardiomyopathy after xylocaine injection during dental procedure

Matthieu Marchetti, Benjamin Monteil

ABSTRACT

Introduction: Takotsubo cardiomyopathy (TTC) is an entity firstly described in 1991, characterized by acute but reversible left ventricular systolic dysfunction in the absence of atherosclerotic coronary artery disease. Acute physical or psychological stresses seem to be the most common causes of takotsubo. We reported a case of atypical TTC, during a dental procedure, including 2% xylocaine injection into gum. Case Report: A 51-year-old Caucasian female with a past medical history of depression underwent dental procedure. The dentist injected a solution of 2% xylocaine into the gum and three minutes following the procedure, the patient complained for chest pain, during one hour. Electrocardiogram revealed T waves inversions into inferolateral leads. Troponin level performed at admission was 23 ng/mL (N ≤ 0,05). Clinical examination, echocardiography and coronary angiography were normal. Cardiac magnetic resonance imaging (MRI) found a global left ventricular hypokinesis, with ejection fraction of 40%, without late Gadolinium enhancement. Conclusion: This case reports an atypical presentation of TTC, induced by xylocaine injection, during the dental procedure. This is an unexpected cardiac side effect of the xylocaine drug mechanism. This case has an interest for the specialties of emergency medicine, cardiology and for dentists usual practice.

Keywords: Takotsubo, Cardiomyopathy, Local anesthesia, Xylocaine

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INTRODUCTION

Takotsubo cardiomyopathy (TTC) is an entity which mimics acute coronary syndrome and characterized by reversible left ventricular systolic dysfunction [1]. Acute physical or psychological stresses are frequently found, but not systematic [2]. Diagnosis is based on the criteria of the Mayo clinic [3]. Postmenopausal women are the most affected population. Several additional tests may be performed to confirm the diagnosis are electrocardiogram, cardiac biomarkers, echocardiography, coronary angiography and ventriculography and magnetic resonance imaging (MRI) scan [4]. Left ventricular dysfunction is pathognomonic, with a circular akinesis or dyskinesis, affecting different medium and apical portions of the left ventricle in its typical form [5]. Here, we report a case of atypical TTC in a 51-year-old female who underwent a dental procedure including 2% xylocaine injection into gum. Xylocaine is used for local anesthesia in many specialties. This is a rare side effect, with atypical presentation of the disease,

due to a global dysfunction of the left ventricle rather than segmental.

CASE REPORT

A 51-year-old Caucasian female with a past medical history of depression underwent dental procedure. She had no allergy, no history of smoking or alcohol consumption, and no family history of heart disease. The dentist injected a solution of 2% xylocaine into gum. Three minutes following the procedure, the patient complained for chest pain, during one hour. She was admitted to a cardiac intensive care unit. Her blood pressure was 125/65 mmHg, her pulse rate was 70 beats per minute (regular), and her spontaneous arterial oxygen saturation was 97%. Her physical examination was normal. Her electrocardiogram (ECG) revealed T waves inversions in AVF, DII, DIII, V4, V5 and V6 (Figure 1). Troponin level performed at admission was 23 ng/mL (N≤0,05). C-reactive protein (CRP) concentration was normal. Chest X-ray was normal. Cardiac catheterization and ventriculography were normal. An early trans-thoracic echocardiogram and cardiac MRI found a global left ventricular hypokinesis with depressed ejection fraction of 40%, without late gadolinium enhancement (Figures 2–4). Chest pain stopped the first day and the patient was discharged from hospital six days later, with beta blocker treatment (bisoprolol 5 mg/day). Left ventricular ejection fraction has been checked normal (65%) after five weeks of treatment. We concluded to atypical TTC induced by injection of xylocaine.

DISCUSSION

Takotsubo, also known as “left apical ballooning”, is defined by acute but reversible left ventricular systolic dysfunction in patients for whom atherosclerotic coronary artery disease was excluded [6]. About 1–2% of all acute myocardial infarction are TTC [7]. Patients are most likely

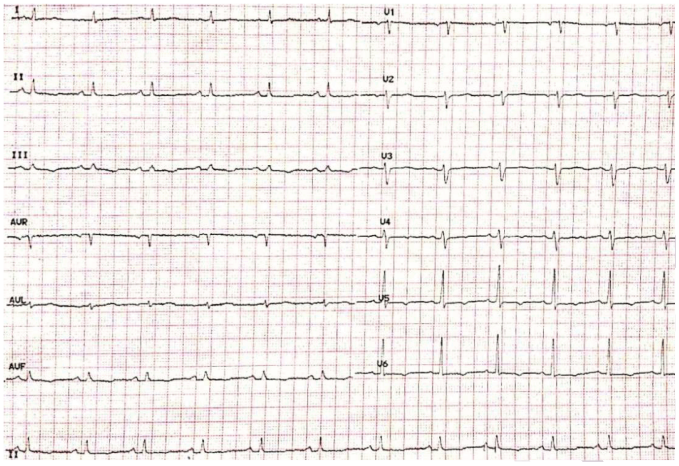

Figure 1: ECG: T waves inversions in AVF, DII, DIII, V4, V5 and V6.

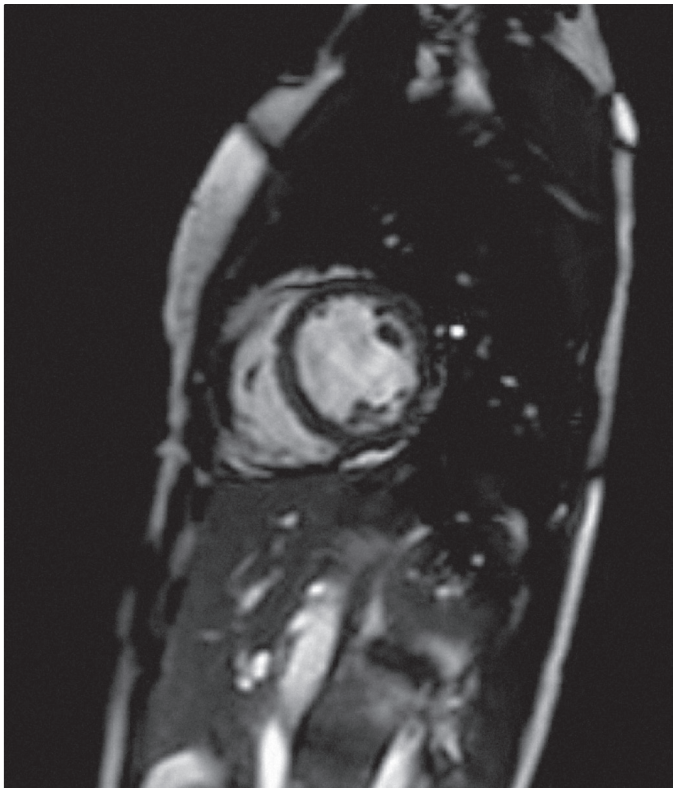

Figure 2: MRI: global left ventricular hypokinesis with depressed ejection fraction of 40 %, without late gadolinium enhancement.

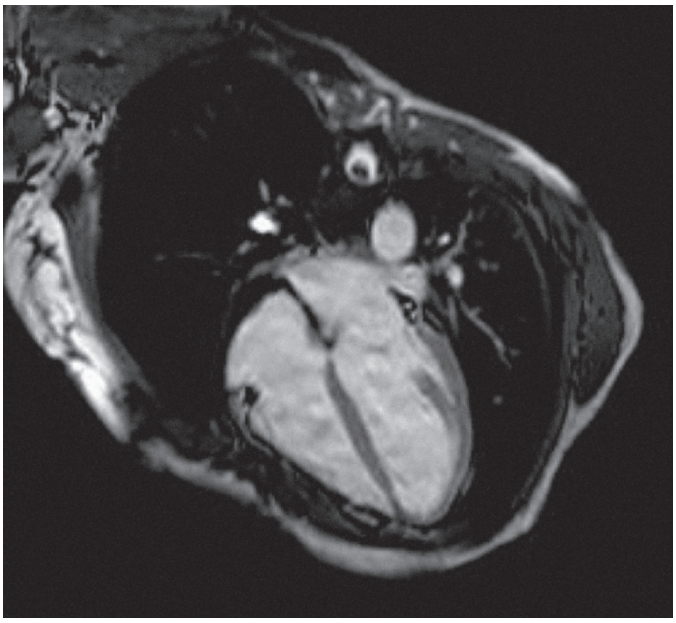

Figure 3: MRI: global left ventricular hypokinesis with depressed ejection fraction of 40 %, without late gadolinium enhancement.

postmenopausal women who experienced acute physical or emotional stress. Ventriculography typically reveals left ventricular apical akinesis or hypokinesis and basal hyper-contractility [8]. The diagnosis of takotsubo was defined through clinical consensus based on the following

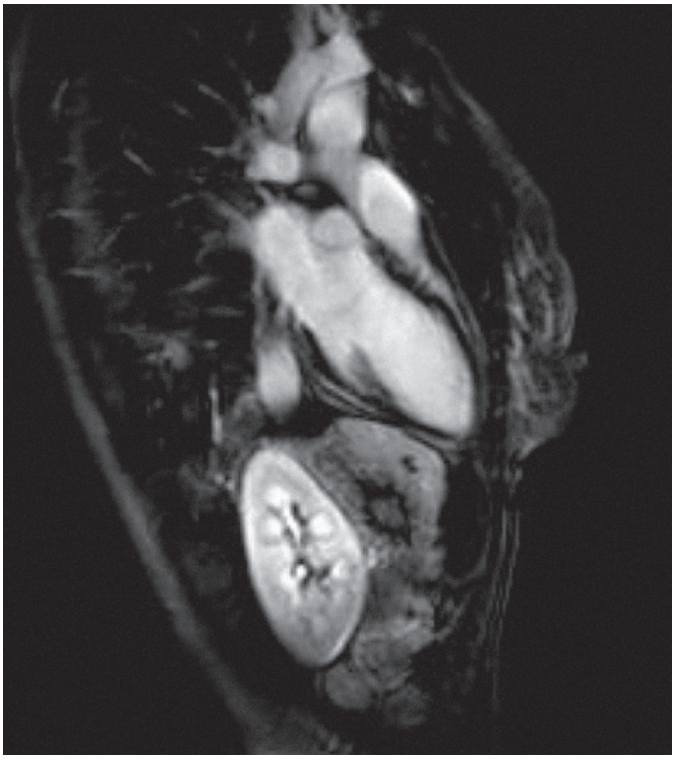

Figure 4: MRI: global left ventricular hypokinesis with depressed ejection fraction of 40 %, without late gadolinium enhancement.

criteria proposed by the Mayo clinic: acute cardiac event typically presenting with chest pain or dyspnea; dyskinesis of the left ventricular segments (hypokinesis or akinesis); wall motion abnormalities extending beyond an epicardial territory distribution; a stressful trigger is often, but not always present; absence of significant obstructive coronary artery disease or angiographic evidence of acute plaque rupture; ECG abnormalities (ST or T-wave modifications) or troponin elevation; absence of pheochromocytoma and absence of myocarditis or typical ischemic transmural late gadolinium enhancement on MRI [9]. Takotsubo syndrome is probably related in part to catecholamine overload [10]. Intravenous administration of catecholamines and other beta-receptor agonists have been associated with takotsubo syndrome occurrence. In a study, nine cases of stress cardiomyopathy occurred immediately after intravenous administration of epinephrine (six cases) or dobutamine (three cases) [11]. As in our case, a study report two cases of TTC occurring in teenage girls undergoing cosmetic rhinoplasty [12]. In these cases, injection of xylocaine with epinephrine was realized into subcutaneous planes. A systematic review of the literature revealed only one case in a dental journal after local anesthesia [13]. Recently, a Takotsubo syndrome occurring after consumption of an energy drink, containing sympathomimetic substances, specifically caffeine and 1,3-dimethylamylamine, had been reported [14]. Twenty different drugs were recognized as possible drug-induced Takotsubo. In the majority of cases, iatrogenic takotsubo are associated with sympathomimetics drugs [15]. To our knowledge, few cases have been reported of patients with atypical presentation of TTC. In some cases they had global left ventricular dysfunction, instead of wall motion abnormalities related to epicardial territory distribution [16, 17]. This rare presentation could be explained by a global microcirculatory impairment effect during the acute phase of TTC [18]. Little data exists on this atypical presentation of the disease. Our case demonstrates that TTC physiopathology is still little-understood.

CONCLUSION

Takotsubo was first described in 1991. In most cases, patients with takotsubo cardiomyopathy (TTC) are postmenopausal women who present chest pain following an acute stress. Since this is a new clinical entity, knowledge are likely to evolve in the coming years. In particular, its physiology must be better defined. Our case shows that some drugs, like local anesthetics, may be responsible of takotsubo with atypical presentation.

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Author Contributions

Matthieu Marchetti – Substantial contributions to conception and design, Acquisition of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published
Benjamin Monteil – Substantial contributions to conception and design, Acquisition of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

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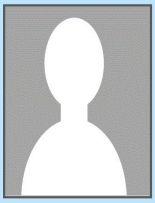

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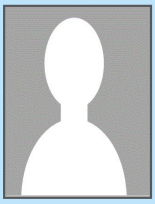

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CASE REPORT OPEN ACCESS | PEER REVIEWED

Successful tenecteplase use in sudden shock from CT-proven massive pulmonary embolism

Jason P. Stopyra, William P. Bozeman

ABSTRACT

Introduction: Emergency, inpatient, and critical care physicians frequently evaluate for and treat patients with deep venous thrombosis (DVT) and pulmonary embolism. While most patients with diagnosed pulmonary embolism remain stable and do not require aggressive therapy, hemodynamic compromise due to pulmonary embolism can occur suddenly and without warning. Case Report: In the community emergency department fibrinolysis is the only available lifesaving option for massive pulmonary embolism. In this report, we describe a patient who suddenly became profoundly unstable moments after establishing the diagnosis by computed tomography scan. Systemic fibrinolysis was administered using the single bolus agent tenecteplase; the patient stabilized and recovered without bleeding complications, neurologic or cardiovascular deficits. Conclusion: All physicians, especially those in a community setting who treat patients with venous thromboembolic disease should be familiar with and prepared to give these agents.

Keywords: CT-proven, Fibrinolysis, Pulmonary embolism, Thrombolysis

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INTRODUCTION

Venous thromboembolic disease includes deep venous thrombosis (DVT) and pulmonary embolism (PE), and can present initially with non-specific signs and symptoms. Many patients have DVT/PE in their working differential diagnosis and clinicians in many settings must maintain a high level of suspicion to diagnose this life-threatening condition. After diagnosis, stable patients with PE are typically treated with parenteral anticoagulation and simple monitoring during conversion to oral anticoagulation therapy [1].

In the setting of hemodynamic instability, due to large PE an immediate reduction in clot burden is needed. In most settings systemic fibrinolysis is the most readily available of the several treatment options and should be undertaken emergently [1, 2]. Fibrinolytic agents are commonly and successfully used in patients with suspected PE who initially present with hemodynamic compromise, but these are most often treated empirically based on clinical suspicion since confirmatory testing is precluded by clinical instability. In some cases, however, an initially stable patient with suspected or confirmed PE may suddenly become unstable and require fibrinolysis. Emergency, critical care, and inpatient physicians in particular must be prepared for this situation and comfortable with the use of systemic fibrinolysis during this pivotal time. Though academic and international studies have been published on this topic, the use of fibrinolysis is even more important because it is the clinician's only option to prevent mortality [3–5].

CASE REPORT

A 33-year-old female presented via ambulance to small community hospital emergency department complaining of feeling weak and dizzy. Two hours prior to calling EMS she began to feel "a funny fluttering in chest" associated with weakness, sweating and near syncope upon standing. She recalled two similar, but less severe episodes over the past week. On questioning, she also reported several episodes of left sided chest pain and dyspnea on exertion over the previous week. Two weeks prior, she had an open reduction and internal fixation of a fractured left metatarsal, with cast immobilization since that time. Other medical history included hypertension, gastroesophageal reflux, and hypercholesterolemia. Medications included lovastatin, lisinopril, esomeprazole, and oxycodone/acetaminophen. She was a non-smoker and family history was unremarkable.

Initial examination showed a conscious, alert female in no distress. Her vital signs on arrival included a heart rate of 145 bpm, respiratory rate of 20 bpm, blood pressure 106/60 mmHg, and oxygen saturation of 97% on oxygen via nasal cannula. Oxygen saturations on room air were not recorded prior to EMS providers initiating oxygen therapy. The physical examination revealed a pleasant young woman with mild tachypnea, tachycardia, normal heart and lung examinations, and her left leg in a cast. An electrocardiogram (EKG) showed sinus tachycardia with right axis deviation, but no dysrhythmias or acute ischemic changes. A chest X-ray, complete blood count, electrolytes, and cardiac enzymes (troponin I and creatine phosphokinase) were all within normal limits.

The risk of PE was considered to be high so a subcutaneous dose of enoxaparin (1 mg/kg) was administered empirically. She was given a fluid bolus and morphine for chest discomfort. She was transported to the radiology area of the emergency department and spiral computed tomography (CT) angiography was performed of her pulmonary vessels. After completion of the scan, just as the patient was moved back to her stretcher, she lost consciousness and displayed a brief episode of generalized tonic-clonic motor activity. Carotid pulses were not palpable during the episode indicating either cardiac arrest or profound hypotension. During the following 30–60 seconds she was rapidly moved to the emergency department treatment area for resuscitation. However, within those seconds her pulses became palpable, and her mental status and blood pressure rapidly improved and returned to baseline over the next three to five minutes.

Review of the CT images obtained moments before showed a large central pulmonary embolism. (Figure 1). A heparin drip was initiated and consent was obtained for systemic fibrinolysis. Tenecteplase 45mg (0.5 mg/kg) was administered intravenously.

Approximately fifteen minutes later, the patient had another sudden episode of increased dyspnea, anxiety and chest pressure, followed by a 45-second loss of consciousness with myoclonus and cyanosis of the face and neck. Blood pressure could not be measured but central pulses remained palpable. The cardiac monitor showed sinus tachycardia throughout the episode. Ventilations were assisted with bag valve mask (BVM). The patient again regained consciousness and subsequently stabilized. She was promptly transferred to the intensive care unit of a nearby tertiary care facility by helicopter.

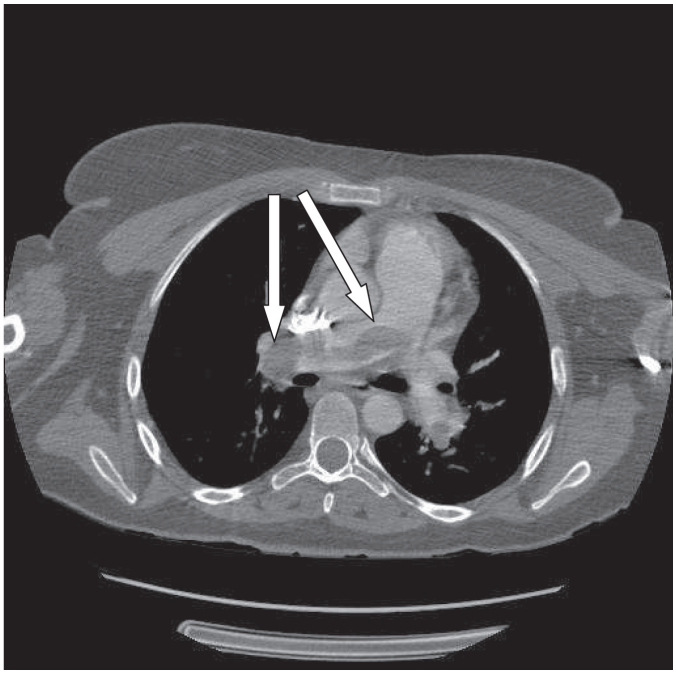

Figure 1: Computed tomography scan image demonstrating bilateral large central pulmonary embolism (arrows).

Upon arrival at the tertiary facility's ICU the patient displayed normal vital signs and had no further episodes of syncope or hypotension. A transthoracic ultrasound was performed on the next day, showing a dilated right ventricle with paradoxical septal motion. Left ventricular size and function were normal. Lower extremity Doppler studies revealed widespread deep venous thrombosis of the distal left leg and an inferior vena caval filter was placed. Serial cardiac enzymes showed a peak troponin I of 0.39 ng/mL on the second hospital day. Warfarin therapy was initiated and she was discharged home after nine days without neurologic deficits or bleeding complications. On follow-up one year later she continued to do well.

DISCUSSION

This case demonstrates an initially stable patient who presented with findings worrisome for pulmonary embolism (PE) that rapidly became unstable. Traditional risk stratification indicators of massive pulmonary embolism that may benefit from fibrinolysis, including hypotension, hypoxia, and elevated cardiac enzymes,

were absent prior to her sudden clinical decompensation. After prompt treatment with systemic fibrinolysis in the emergency department, she stabilized and was later discharged without neurologic deficit or other adverse outcome. Without aggressive treatment the patient may well have had additional hemodynamic decompensation and a poor or fatal outcome.

Despite the patient's non-specific initial chief complaint of feeling weak and dizzy, careful questioning and physical examination made the physician strongly suspect pulmonary embolism. Her rapidly evolving process gave no warning of its change from stable to unstable with the need for aggressive measures. It was fortunate and unusual that the first episode of hemodynamic instability occurred just after the CT scan was completed, allowing confirmation of the suspected diagnosis.

Fibrinolytic agents are widely accepted as first line therapy in the setting of hemodynamic compromise due to massive PE. There is ongoing discussion whether this therapy should be expanded to other high risk patients without hemodynamic compromise, such as those with elevated cardiac biomarker or right ventricular dysfunction on EKG, echocardiogram, or CT angiography [1, 2, 6, 7]. Other treatments such as catheter-guided thrombectomy or surgical embolectomy, if available emergently, should also be considered to treat massive PE [1]. In this case, tenecteplase (TNKase®) was used for systemic fibrinolysis. This agent is administered as a single weight-based bolus injection; it is immediately available in our community emergency department for ST elevation myocardial infarction (STEMI) and has also been used for PE [4]. Other commonly available fibrinolytic agents include reteplase (Reteplase®), which is administered in two 10 mg doses 30 minutes apart, and alteplase (recombinant tissue plasminogen activator, r-tPA or Activase®) which is typically given as a 100 mg infusion over 2 hours. All three agents are approved by the United States Food and Drug Administration for STEMI. Alteplase and the older agents streptokinase and urokinase are formally approved for PE [2, 7].

The choice of fibrinolytic agent for PE remains one of physician judgment, preference, and availability. The tenecteplase used in this case has several potential advantages over other agents. These include single bolus dosing, which makes it attractive for resuscitation situations and may alleviate confusion about infusion protocols and co-administration of heparin, and may result in more rapid plasmin formation and clinical effects [7]. In addition, the possibility of bleeding complications is always of concern when considering systemic fibrinolysis. In previous STEMI trials, tenecteplase has had fewer non-cerebral bleeding complications than alteplase [8].

Most emergency physicians and cardiologists practicing in community hospitals are comfortable giving fibrinolytic agents to STEMI patients, in part because EKG criteria clearly show confirmatory evidence to indicate their use. The diagnosis of massive PE is frequently less clear, since unstable patients needing the therapy are frequently too unstable to undergo CT scanning. Other imaging modalities such as transesophageal ultrasound and formal angiography which can confirm the diagnosis may not be available in a timely manner. Consequently, in the setting of massive PE, systemic fibrinolytics frequently must be administered based on clinical suspicion alone.

As demonstrated in this case, even an initially stable patient can decompensate suddenly and without warning. Therefore, all physicians who may treat patients with known or suspected DVT or PE should be familiar with fibrinolytic therapy and prepared to administer these agents rapidly when indicated. A fibrinolytic eligibility checklist to exclude contraindications should be completed in some form even in stable patients at the time the diagnosis is entertained since the necessary information may be unavailable or too time-consuming to collect if a patient becomes unstable and requires rapid treatment.

CONCLUSION

All emergency and inpatient physicians, especially those in a community setting should maintain a high level of suspicion for pulmonary embolism and should be prepared to administer systemic fibrinolytic agents when needed in the setting of massive pulmonary embolism, even if patients are initially stable upon presentation.

KEY POINTS

- Patients with deep venous thrombosis and pulmonary embolism can become unstable rapidly and without warning.
- Immediate clot reduction can be lifesaving in this situation; systemic fibrinolytic agents are commonly available and frequently used.
- All medical providers who treat patients with venous thromboembolic disease should be familiar with this treatment modality.

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Author Contributions

Jason P. Stopyra – Substantial contributions to conception and design, Drafting of article, Revising it critically for important intellectual content, Final approval of the version to be published

William P. Bozeman – Substantial contributions to conception and design, Drafting of article, Revising it critically for important intellectual content, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

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CASE REPORT OPEN ACCESS | PEER REVIEWED

Chronic cutaneous lesions in immunocompromised patients

Wei-Chieh Lee, Wei-Sin Li, Chih-Hsiung Lee

ABSTRACT

Introduction: Chronic cutaneous lesions are common in immunocompromised patients and are attributable to a wide range of potential microbial pathogens. Common infections may have a variety of unusual manifestations, and unusual pathogens can also play an important role in these infections. Therefore, "how to approach" these lesions becomes a difficult problem. Case Report: A 42-year-old male had medical history of immune thrombocytopenic purpura (ITP), chronic hepatitis C with liver cirrhosis, Child's-Pugh-Turcotte score A, and end stage renal disease requiring maintenance hemodialysis. The patient also suffered from multiple ecchymoses and tender plaques on all four limbs for one month. A pathology of skin biopsy showed lobular panniculitis and a strong positive finding of acid-fast bacilli (AFB). A bone marrow biopsy also showed a strong positive finding of AFB but no granulomatous inflammation. The patient was treated as disseminated tuberculosis infection and experienced anti-tuberculosis (TB) drug-related adverse effects. Finally, non-tuberculous mycobacteria (NTM) was diagnosed. There are difficulties regarding how to survey and treat immunocompromised patients due to multiple side effects and comorbidities. In recent years, NTM has been characterized as an emerging pathogen. Conclusion: Cutaneous lesions in immunocompromised patients are complex because of a wide range of potential microbial pathogens. Common infections may have unusual manifestations in immunocompromised patients. The NTM is an emerging opportunistic pathogen in severely immunocompromised patients with acquired immunodeficiency syndrome or a transplantation in recent years. The incidence of pulmonary or extrapulmonary NTM infection has increased dramatically in recent years. We should consider NTM infection if a clinical condition does not improve after several months of anti-TB therapy.

Keywords: Chronic Cutaneous Lesion, End stage renal disease, Immune thrombocytopenic purpura, Nontuberculous mycobacteria

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INTRODUCTION

Chronic cutaneous lesions are common in immunocompromised patients and are attributable to a wide range of potential microbial pathogens. Common infections may have a variety of unusual manifestations,

and unusual pathogens can also play an important role in these infections. Therefore, "how to approach" these lesions becomes a difficult problem.

CASE REPORT

A 42-year-old male had medical history of immune thrombocytopenic purpura (ITP), chronic hepatitis C with liver cirrhosis, Child's-Pugh-Turcotte score A, and end stage renal disease requiring maintenance hemodialysis. The patient also suffered from multiple ecchymoses and tender plaques on all four limbs for one month. The lesions secreted bloody discharge (Figure 1A–D). Cellulitis was initially treated at our emergency department. The dermatologist suspected erythema induratum and suggested a skin biopsy. However, the procedure was postponed due to severe thrombocytopenia. Fever, chills, recent trauma, insect bites, pet contact, and travel history were all ruled-out.

A high fever with chills started three days after treatment for cellulitis with empirical antibiotics (intravenous oxacillin: one gram every four hours). Ulcerative plaques with bloody discharge on all four limbs became gradually more severe. New ecchymotic nodules with pus occurred. The patient's vital signs were: blood pressure 166/108 mmHg, heart rate 106 beats per minute, respiratory rate 18 breaths per minute, temperature 39.3°C.

The patient's blood workup revealed hemoglobin concentration of 7.7 g/dL, a leukocyte count of $13.3x10^3$ cells/mm^3 with an absolute neutrophil count of 87%, and a platelet count of $20x10^3$ cells/mm^3. C-reactive protein (CRP) was 69 mg/L. Chest radiography showed multiple nodules over the right upper lung and left upper lung, compared to previous radiographs (Figure 2). The patient did not have respiratory symptoms but was isolated because pulmonary tuberculosis could not be excluded. High resolution computed tomography scan showed a diffuse ground-glass picture over the central and upper lung, two cavitary lesions in the right middle lung (1.6 cm) and right lower lung (1.6 cm), and minimal pleural effusion (Figure 3A–D).

The high fever and chills persisted after antibiotics were modified several times. A skin biopsy was performed, and pathology showed lobular panniculitis and a strong positive finding of acid-fast bacilli (Figure 4). A bone marrow biopsy also showed a strong positive finding of acid-fast bacilli but no granulomatous inflammation. Both biopsies had negative gram stains and periodic acid-schiff stain. Disseminated tuberculosis infection involving skin and bone marrow was suspected. Anti-tuberculosis (TB) medication, including rifampin 360 mg, isoniazid 240 mg, and pyrazinamide 150 mg per day, was started. The high spiking fever persisted and new plaques with discharge continued to occur.

After a four-day course of anti-TB medication, hyperbilirubinemia (total bilirubin: 2.5 mg/dL and direct bilirubin: 1.64 mg/dL) and hyperuricemia occurred. The anti-TB medication protocol was modified as follows: rifampin 600 mg, INH 300 mg and EMB 800 mg per day. Anti-TB treatment was stopped due to progressive jaundice. After 8 days of observation, hepatic function improved, and rifampin 450 mg, INH 200 mg per day and EMB 800 mg per two days was started. It was frustrating that the intermittent fever persisted and the skin lesions healed poorly after anti-TB treatment continued for two months. Unfortunately, the dose could not be increased due to hepatic function.

A culture of bone marrow and sputum showed the presence of *Mycobacterium haemophilum* approximately 82 and 98 days later. Therefore, the final diagnosis was disseminated *Mycobacterium haemophilum*, which

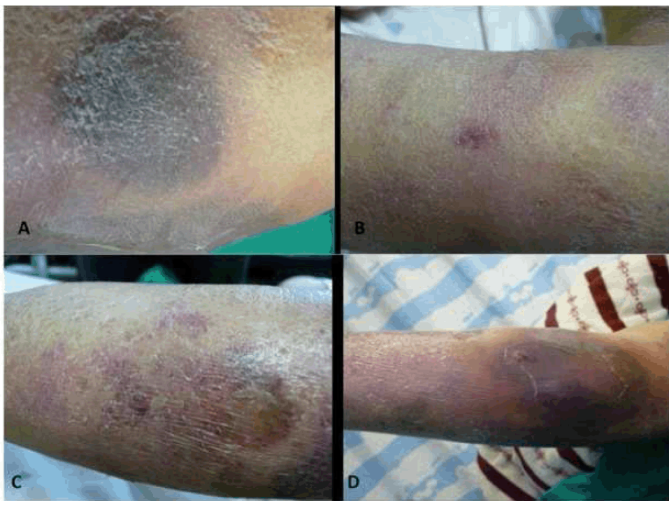

Figure 1: (A, B, C, D) Tender ecchymoses and purpuric plaques over the lower limbs and, elbows with bloody discharge or pus.

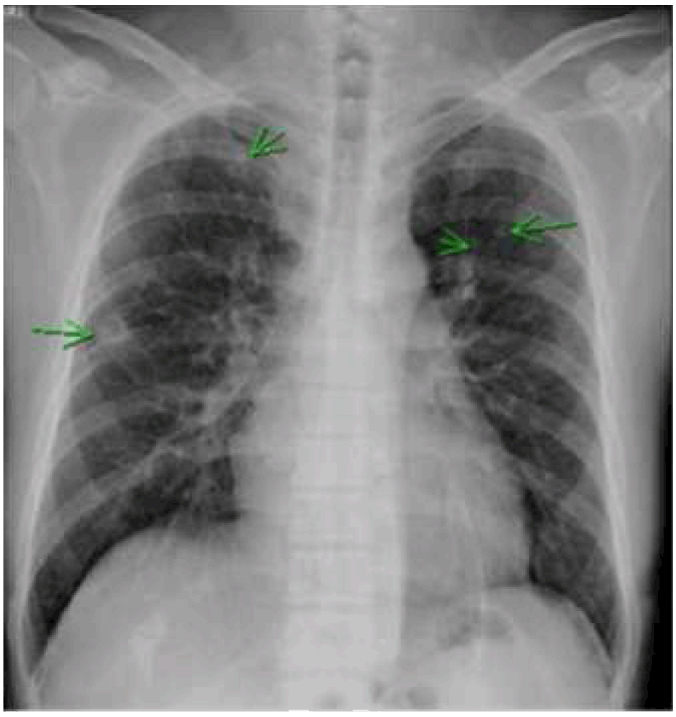

Figure 2: Multiple nodules over the lung (green arrows).

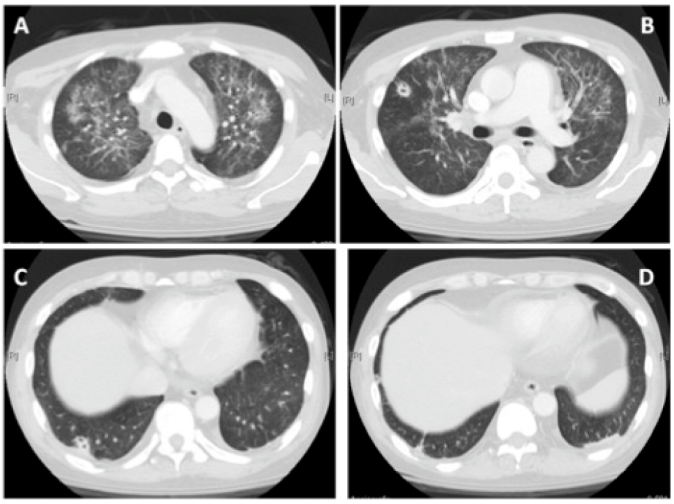

Figure 3: (A, B, C, D) Diffuse ground-glass picture over of the central and upper lung, two cavitary lesions in the right middle lung (1.6 cm) and right lower lung (1.6 cm) and minimal pleural effusion.

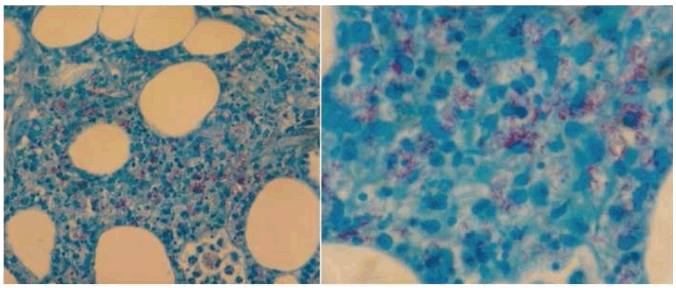

Figure 4: Lobular panniculitis and strong positive finding of acid-fast bacilli.

involved the skin, bone marrow, joints and lung. The antibiotics were changed to rifabutin 300 mg per day and clarithromycin 500 mg every twelve hours. After that, the fever subsided and treatment was continued for one year. The patient received regular outpatient department follow-up every three months and no pus or ulceration was noted.

DISCUSSION

Cutaneous lesions in immunocompromised patients are complex because of a wide range of potential microbial pathogens. Common infections may have unusual manifestations in immunocompromised patients. We could list several differential diagnosis as formulas VINDICATE for chronic cutaneous lesion with ulceration (Table 1) [1]. Figure 5 showed an algorithm to approach chronic cutaneous lesions in immunocompromised patients.

Lobular panniculitis is a group of diseases whose hallmark is inflammation of subcutaneous adipose tissue. Lobular panniculitis can be related to trauma, connective tissue disease (systemic lupus erythematous or scleroderma), lymphoproliferative disease (lymphoma or histiocytosis), pancreatitis or pancreatic cancer, and other causes [2]. Positive acid-fast staining pathogens include all *Mycobacteria* (TB, NTM), Actinomyces (*Nocardia*, *Rhodococcus*, *Gordonia*, Tsukamurella, Dietzia, nuclear inclusion bodies (Lead poisoning, Bismuth poisoning), and cysts of some coccidian parasites (*Cryptosporidium parvum*, *Isospora belli*, and *Cyclospora cayetanensis*).

The skin biopsy showed positive acid-fast staining and favored cutaneous tuberculosis infection causing

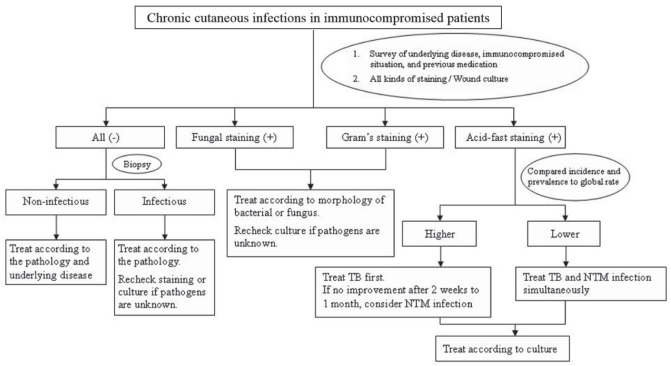

Figure 5: An algorithm to approach chronic cutaneous infections in immunocompromised patients.

Table 1: Differential diagnosis as formulas VINDICATE for chronic cutaneous lesion

V	Vascular	Peripheral arteriosclerosis, venous ulcer, and varicose ulcers
I	Infections	Viruses, Bacteria, Spirochetes, Parasites, Fungus
N	Neoplasm	Sarcomas, mycosis fungoides
D	Degenerative	Peripheral neuropathy, syringomyelia, and peroneal muscular atrophy
I	Intoxication	Ulcer of chronic dermatitis
C	Congenital	Ulcers of sickle cell anemia
A	Autoimmune	Periarteritis nodosa, pyoderma gangrenosum
T	Trauma	Burns and radiation and secondary to unhealed lacerations and decubitus ulcers
E	Endocrine	Diabetic ulcers

ulceration and lobular panniculitis. Anti-TB treatment was started, but the fever and cutaneous lesions persisted even after two months of anti-TB treatment. NTM infection was considered. The cultures of bone marrow and skin showed *Mycobacterium haemophilum*. Disseminated *M. haemophilum* infection, involving the skin, lung and bone marrow, was diagnosed.

The patient often had hematomas on the lower limbs due to ITP. Our hypothesis was that hematomas were a good media for *M. haemophilum* growth. However, no obvious connection between immune thrombocytopenic purpura and *M. haemophilum* has been reported previously. Cell-mediated immunity is a significant factor in disease caused by *M. haemophilum* [3, 4].

M. haemophilum is sensitive to amikacin, clarithromycin, ciprofloxacin, rifampin and rifabutin, but it is usually resistant to other first-line antimycobacterial medications [3, 5]. Currently, according to guidelines from the 2007 American Thoracic Society/Infectious Diseases Society of America, patients with irreversible immunosuppression probably require maintenance therapy for months to years. Duration of therapy should be guided by the patient's underlying condition and clinical response. Severe disease (disseminated disease, osteomyelitis, and pulmonary disease) that responds well to treatment should be treated for a minimum of several months (e.g., 6–12 months) [6, 7].

In our case, we switched medication to rifabutin 300 mg once per day and clarithromycin 500 mg twice per day after confirmation of *M. haemophilum* infection. The fever subsided rapidly and no new nodules were noted on the lower extremities. After one week of therapy, the patient was discharged but continued follow-ups and medical therapy at our outpatient department. Treatment continued for one year. The skin lesions healed and no pus or ulcerations were noted after one year. The patient's hepatic function remained within the normal range [8].

CONCLUSION

A number of learning points are illustrated by this case. Firstly, careful examination of the skin may provide the earliest clues to the accurate diagnosis of systemic infections. Generalized skin lesions are often diagnosed as cellulitis. We cannot forget that cutaneous lesions in immunocompromised patients are complex because of a wide range of potential microbial pathogens. Common infections may have unusual manifestations in immunocompromised patients. Skin biopsy is usually necessary to confirm the diagnosis. Secondly, we also need to realize that nontuberculous mycobacteria (NTM) is an emerging opportunistic pathogen in severely immunocompromised patients with acquired immunodeficiency syndrome or a transplantation in recent years. The incidence of pulmonary or extrapulmonary NTM infection has increased dramatically in recent years. We should consider NTM infection if a clinical condition does not improve after several months of anti-TB therapy.

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Author Contributions

Wei-Chieh Lee – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Wei-Sin Li – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published

Chih-Hsiung Lee – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

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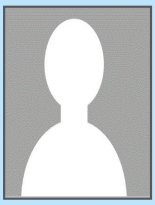

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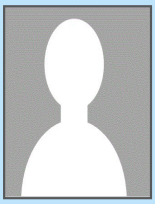

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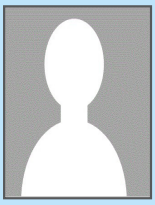

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CASE REPORT OPEN ACCESS | PEER REVIEWED

An unusual case of gastric gangrene, diaphragmatic gangrene and autosplenectomy due to mucormycosis in a diabetic patient

Amrit Manik Nasta, Kushal Bairoliya, Shashi Ranjan

ABSTRACT

Introduction: Stomach has a rich vascular supply and undergoes gangrene in rare circumstances. Some of the known causes include gastric volvulus, diaphragmatic herniation, caustic injury and infectious necrotising gastritis. Mucormycosis, a life-threatening infection caused by fungi of the subphylum Mucoromycotina, order Mucorales, usually affects the para-nasal sinuses, orbit, brain and lung. Case Report: We report an unusual presentation of gastric mucormycosis in a 50-year-old diabetic male, leading to perforative peritonitis with diaphragmatic gangrene and auto-splenectomy. Conclusion: Clinical suspicion of mucormycosis in immunosuppressed to aid diagnosis and adequate treatment in the form of antifungal agents and surgery may help improve outcome.

Keywords: Autosplenectomy, Diaphragmatic gangrene, Gangrene of stomach, Gastric Mucormycosis

How to cite this article

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INTRODUCTION

Mucormycosis of stomach is a rare cause of gangrene and perforation of gastric wall [1]. Fungi of the order Mucorales are causative agents of Mucormycosis, commonly of genus *Rhizopus* and *Mucor*. Risk factors for the development of invasive mucormycosis include diabetes, immunosuppressive states, corticosteroid use, organ or stem cell transplantation, and increased levels of available serum iron [2]. Common sites of infection are the paranasal sinuses, orbit, brain and lung. Gastrointestinal mucormycosis, in the past, was usually seen in premature neonates with widespread dissemination of the disease [3]. We report an unusual presentation of gastric mucormycosis leading to gangrene and perforation, associated with gangrene of diaphragm and autosplenectomy, in a diabetic male.

CASE REPORT

A 50-year-old male, known diabetic, presented to the emergency medical department with complaints of breathlessness and left sided chest pain since one week. Pain was continuous, dull aching and present at rest. He had no history of similar chest complaints in the past. There were no abdominal complaints. There was no significant surgical history or history of any trauma to the chest. On examination, patient was conscious and oriented, afebrile, pulse 100/minute, blood pressure 90/70 mmHg, respiratory rate was 22/minute. Air entry was decreased in left lower zones of the lung with dullness on percussion. Abdominal examination revealed mild tenderness in left hypochondriac region, otherwise unremarkable. Chest X-ray revealed left hydro-pneumothorax, abdominal ultrasound revealed free fluid in left sub-phrenic space. Computed tomography (CT) scan of chest and abdomen revealed perforation of fundus

of stomach with extravasation into left pleural space, rent in left hemi-diaphragm and absent spleen (Figure 1). After initial resuscitation, patient underwent an emergency exploratory laparotomy which showed gangrene of posterior wall of stomach with demarcation, with a 1x1 cm perforation. A left diaphragmatic rent about 6x6 cm with gangrenous edges, absent spleen and moderate perigastric collection were other findings (Figures 2 and 3). Rest of the bowel was normal. Posterior gangrenous segment of stomach was excised and wall was closed using 55 mm linear staplers. Gangrenous diaphragmatic edges were excised, left intercostal drain placed and defect plugged with 15x15 cm composite mesh, as edges could not be approximated. Washes were given and distal feeding jejunostomy was done. Histopathology of resected stomach revealed necrosis with mucormycosis (Figures 4 and 5). Postoperatively, patient complicated with staple line leak and sepsis, and expired on postoperative day-3.

DISCUSSION

The gastrointestinal tract is a rare site of mucormycosis. Gastrointestinal mucormycosis was seen mainly in neonatal age group, often in association with disseminated disease [3, 4]. Other rare cases of gastrointestinal mucormycosis were previously described in association with other immunocompromised states, like HIV, systemic lupus erythematosus, and organ transplantation [5]. Cases of hepatic mucormycosis have also been associated with ingestion of herbal medications [6]. As this infection is acute and rapidly fatal; it is often diagnosed with post-mortem.

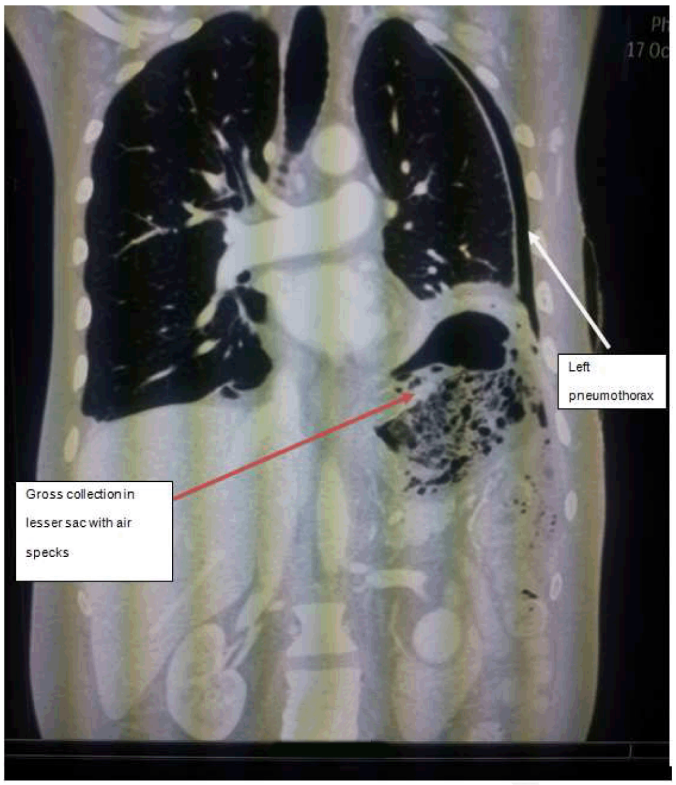

Figure 1: Contrast-enhanced computed tomography scan of chest abdomen and pelvis, coronal view, showing collection in the lesser sac with air specks and absent spleen with a left pneumothorax.

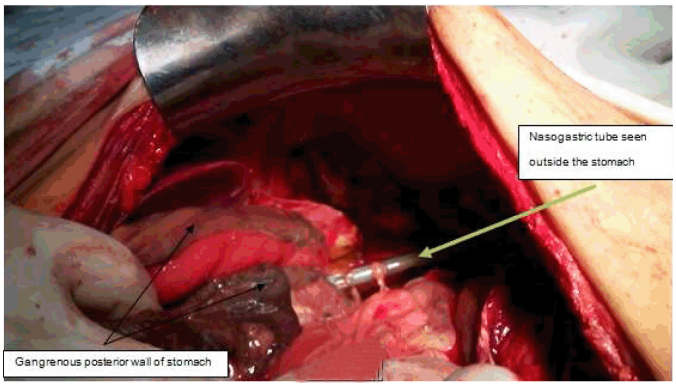

Figure 2: Intraoperative photograph showing the gangrenous posterior wall of the stomach with nasogastric tube seen outside through a perforation.

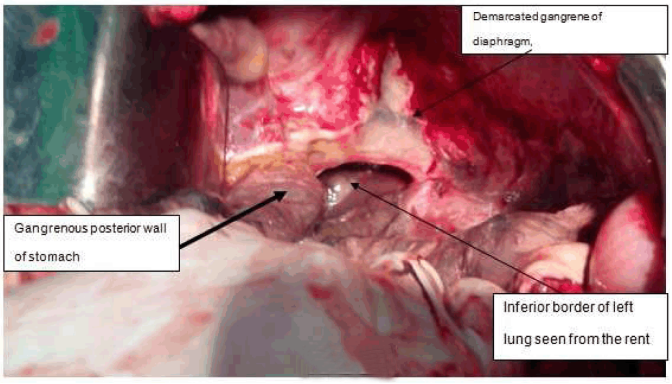

Figure 3: Intraoperative photograph showing the gangrenous posterior wall of the stomach with demarcated gangrene of the diaphragm. The inferior border of left lung is seen through a rent in the diaphragm.

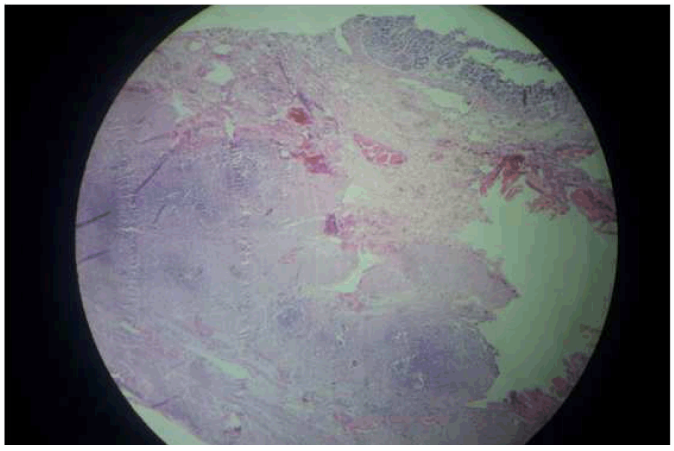

Figure 4: Low power H&E stained section showing gastric mucosa. Underlying muscularis propria shows confluent areas of necrosis with giant cells.

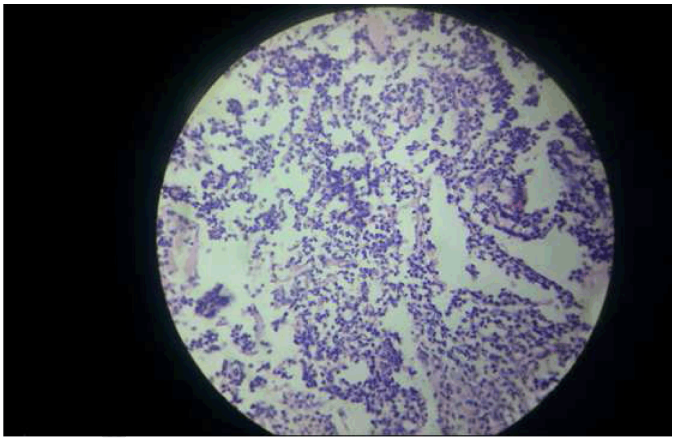

Figure 5: High power H&E stained section showing neutrophilic infiltrate around septate hyphae branching at right angles suggestive of Mucor.

Mucormycosis of gastrointestinal tract, depending on the site of pathology, may present as pain, distention, nausea and vomiting commonly [7]. Fever, hematemesis, melena or hematochezia may also occur. The patient is often misdiagnosed to have an intra-abdominal abscess. The diagnosis may be made by biopsy of the suspected area during surgery or endoscopy [7]. Our patient had an unusual presentation with mainly chest symptoms due to hydropneumothorax.

In a case reported by Morton et al., a patient had Crohn's disease, who presented with perforative peritonitis and there was no specific reason to suspect mucormycosis [8]. Patient was on steroid therapy for Crohn's disease. Only the appearance of the fungi on histopathology caused the diagnosis to be made and appropriate therapy to be initiated. This case highlights the need to maintain a high index of suspicion for invasive fungal infections like mucormycosis, in immunosuppressed patients.

Mucormycosis is difficult to treat with antifungals alone, like Amphotericin B and triazoles like Posaconazole. In addition, surgery is required to remove all infected tissue. Mucormycosis shows angioinvasion and tissue necrosis which results in poor local penetration of anti-fungal agents. With an ongoing epidemic of obesity and diabetes, and the increasing population of patients receiving immunosuppressive therapy for inflammatory diseases, solid organ or stem cell transplantation, it is not surprising that recent studies have reported alarming increases in the incidence of mucormycosis [9]. It is likely that the clinicians will encounter this disease more frequently in the coming years, especially in the nosocomial setting.

CONCLUSION

Invasive mucormycosis is a life-threatening infection, with varied clinical presentations including gastrointestinal involvement. Clinical suspicion in immunosuppressed to aid diagnosis and adequate treatment in the form of antifungal agents and surgery may help to improve outcome.

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Author Contributions
Amrit Manik Nasta – Conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Critical revision of the article, Final approval of the version to be published
Kushal Bairoliya – Conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Critical revision of the article, Final approval of the version to be published
Shashi Ranjan – Acquisition of data, Analysis and interpretation of data, Final approval of the version to be published

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Conflict of Interest
Authors declare no conflict of interest.

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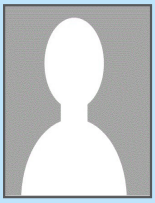

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CASE REPORT OPEN ACCESS | PEER REVIEWED

Spontaneous heterotopic triplet pregnancy: Tubal and intrauterine twin gestation

Ebelechukwu Chris Nnoli, Andrew Lefkovits, Clark Bruce

ABSTRACT

Introduction: Although there have been reports of heterotopic pregnancies in published literature, there has not been a documented report, to the best of our knowledge of a spontaneous conception of intrauterine twins along with a tubal ectopic without risk factors. Case Report: A 29-year-old female with a prior obstetric history of spontaneous intrauterine twins presented with a second spontaneous conception of intrauterine twins along with a tubal ectopic pregnancy. The patient was taken to the operating room after tubal rupture, where the ectopic conceptus was removed laparoscopically without complications. She was discharged the following day to follow-up as an outpatient. At the time of writing, her last prenatal visit at 24 weeks gestation was uncomplicated. Conclusion: Though rare, it is prudent that there be continued awareness of the possibility of a heterotopic pregnancy even in the absence of risk factors especially in a patient with symptoms. When confirmed and the patient is clinically stable, surgical management via laparoscopy is a safe alternative.

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INTRODUCTION

Heterotopic pregnancy is when gestation present in two or more sites of implantation. It is a rare event with an occurrence rate of less than 1 in 30,000 in spontaneous pregnancies. With the introduction of assisted reproductive technology, the occurrence rate is between 1 in 100 and 1 in 500 [1]. After extensive literature search and finding no recorded case, we report a case report of a rare occurrence of a spontaneously conceived twin intrauterine gestation along with a tubal ectopic pregnancy.

CASE REPORT

A 29-year-old female presented to the emergency department at our hospital with complaints of mild abdominal pain associated with nausea. Physical examination findings were normal with stable vital signs and a slightly elevated white blood cell count of $14x10^3$ cells/mm^3. A transvaginal ultrasound scan revealed di-amniotic di-chorionic twin intrauterine pregnancy at 6 weeks and 2 days gestation as well as normal sized ovaries with arterial and venous flow to both ovaries. The patient reported no use of assisted reproductive techniques in conception. Of significance, she also reported spontaneous conception of di-amniotic di-chorionic twins, delivered vaginally at 35 weeks, 3 years

prior to presentation to the emergency department. Her history was only notable for breast augmentation after her twin delivery, multiple urinary tract infections, former 14-pack per year smoker and the use of Depo-Provera for 12 years. After satisfactory workup in the emergency department and with consultation from the obstetrics and gynecology team, the patient was discharged to follow-up as an outpatient for a repeat ultrasound. At eighth week and four days she presented to the office, where a repeat transvaginal ultrasound scan revealed twin gestation with an ectopic pregnancy situated in the right tube, close to the right ovary (Figures 1–3). Given the patient's stable clinical status upon presentation, she was scheduled for laparoscopic surgery the following morning. A few hours upon leaving the office, the patient presented to our emergency department complaining of severe lower abdominal pain. An acute abdomen was diagnosed on physical examination with stable vital signs, white blood cell count of $16x10^3$ cells/mm^3, hemoglobin 11 g/dL, hematocrit 34%. A diagnosis of a ruptured ectopic was made clinically mainly based on the acute abdomen. As a result, the patient was taken for an emergent laparoscopy. Upon entry into the abdomen, the right tubal pregnancy with partial implantation on the broad ligament had ruptured with significant hemoperitoneum. A fimbriectomy was carried out without complications. The patient tolerated the procedure well and was discharged the following day in stable condition.

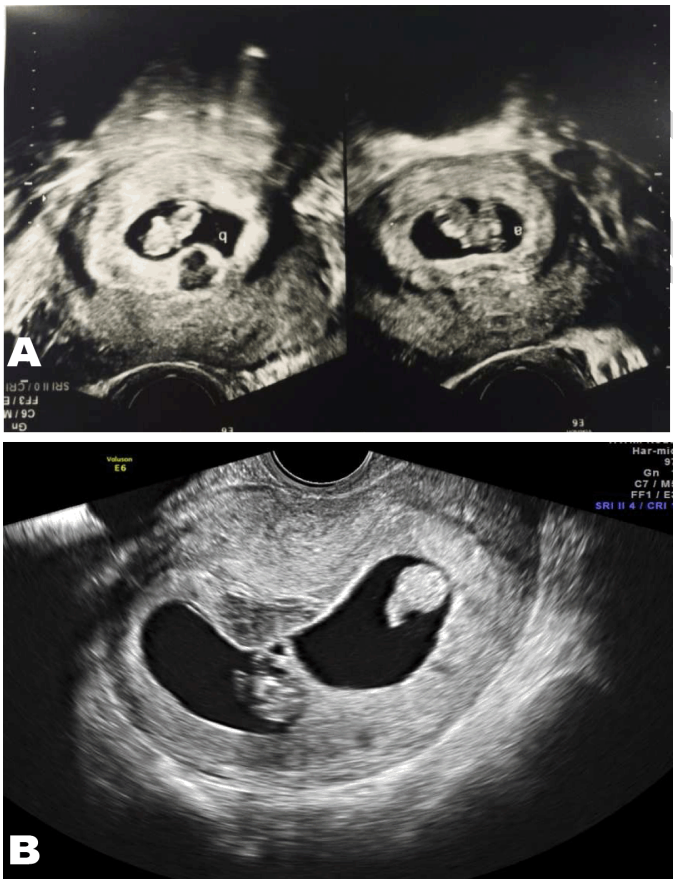

Figure 1 (A, B): Ultrasound images showing both twins. Twin B is close to the ectopic pregnancy.

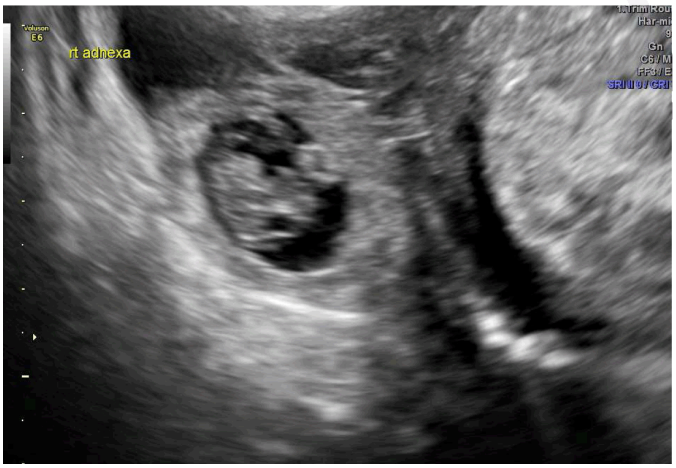

Figure 2: A closer look at the ectopic in the right adnexa.

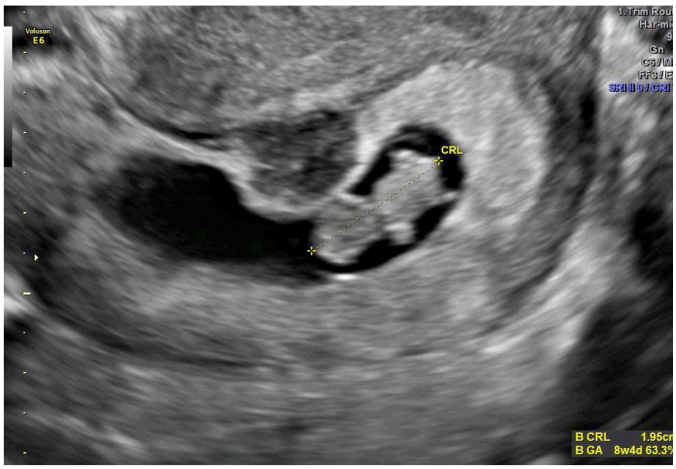

Figure 3: The ectopic close to one of the twins.

DISCUSSION

There are various forms of heterotopic pregnancy including but not limited to twin tubal pregnancy and intrauterine pregnancy, bilateral tubal and intrauterine pregnancy and intrauterine pregnancies associated with cornual, cervical or ovarian pregnancies [1, 2]. A large number of reported cases of twin heterotopic pregnancies are associated with assisted reproductive techniques, with reported rates of 1 in 100–500 [1]. With spontaneous conception, the rate decreases to 1 in 30,000 [1]. Our patient is unique in that this pregnancy is the second spontaneous pregnancy resulting in twin gestation. She had none of the risk factors that would predispose her to having an ectopic pregnancy, namely a history of pelvic inflammatory disease, endometriosis, tubal surgery that may cause damage to the tubes, previous ectopic and assisted reproductive techniques (In vitro fertilization being the most important risk factor) [1, 3].

Transvaginal ultrasounds are vital in diagnosing heterotopic pregnancies as early as five weeks up to 34 weeks with ours diagnosed a little over eight weeks [1, 3, 4]. The clinical presentation of a heterotopic pregnancy

varies widely, with abdominal pain the most commonly reported symptom [3, 4].

When our patient presented at sixth week with complaints of abdominal pains, a transvaginal ultrasound was done which documented the twin gestation though it is not surprising that the ectopic was missed as the sensitivity of ultrasound is low [1].

Diagnosing the ectopic gestation of a heterotopic pregnancy is difficult, resulting in delay of diagnosis which can put the mother at risk of complications, namely tubal rupture, increased risk of blood transfusion and shock. We encountered the same difficulty at initial presentation prompting discharge to follow-up as an outpatient. When she returned to the office, a transvaginal ultrasound was able to detect fetal heart rates in both the ectopic and the twin fetuses, which confirmed the diagnosis. Successful medical management of ectopic pregnancy using methotrexate or KCl has been described in literature though this should not be used in the event of a ruptured ectopic [2]. Surgery is one of the treatments of ectopic pregnancy, which can be done via laparotomy or laparoscopy [1–3]. We proceeded with laparoscopy to help minimize interference with the intrauterine pregnancy but more so because she was hemodynamically stable, as evidenced by her clinical presentation and vital signs. According to published reports, laparoscopic management of heterotopic pregnancy allows for a faster recovery time and minimal requirements for antibiotics and pain medications, which was true in our case [5]. As long as operating time is kept under an hour and a 10–12 mmHg intraperitoneal pressure maintained, there is minimal effect on mother and fetus [5]. Although a study released by Heather et al. suggest that heterotopic pregnancies are more likely to result in spontaneous abortion, at the time of writing this report, the patient continues to do very well with this pregnancy and just recently completed her 24th week prenatal visit without complications [6].

CONCLUSION

This case demonstrates the uniqueness of heterotopic pregnancy without risk factors or assisted reproductive techniques and the use of laparoscopy as a safe surgical alternative for its treatment as long as the patient is hemodynamically stable.

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CASE REPORT OPEN ACCESS | PEER REVIEWED

Septic cavernous sinus thrombosis secondary to a mycotic pseudoaneurysm of a cubital arteriovenous fistula

Ahmed Mohamed Elhassan Elfaki Osman,
Saif Eldin Mohammed Ali Ibrahim

ABSTRACT

Introduction: The cavernous sinuses are part of the dural sinuses. In 1831, Bright described cavernous sinus thrombosis (CST) as a complication of epidural and subdural infections. Cavernous sinus thrombosis usually results as a complication of paranasal sinus infection or infections of the face, in an area called the 'danger triangle', trauma, bacteremia or ear infections. Case Report: A 35-year-old female presented with one month history of high grade fever and progressively increasing periorbital swelling. This presentation was preceded by failure and infection of her arteriovenous fistula (AVF) three weeks beforehand, which was resistant to medical therapy. On examination, she was febrile and had periorbital swelling with bilateral closure of the eyes. The left jugular vein was distended. At the site of the AVF, there was a pulsatile swelling which was also discharging pus. Following aneurysmectomy and AVF ligation, an angiogram of the head and neck showed a long segment occlusion on the left internal jugular vein extending to the left brachiocephalic vein plus a cavernous sinus thrombus. A diagnosis of a septic CST was made and management was conservative. We report this case because, to the best of our knowledge, no literature was found describing a mycotic AVF complicated by a jugular and brachiocephalic vein thrombosis causing a septic CST. Conclusion: In patients presenting with a mycotic AVF, the risk of developing a septic CST should be kept in mind. The goal of intervention should be to control the source of infection and prevent complications.

Keywords: Aneurysm, Cavernous sinus thrombosis (CST), Mycotic arteriovenous fistula, Septic cavernous sinus thrombosis

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INTRODUCTION

The cavernous sinuses are part of a group of sinuses which, together, form the dural sinuses (composed of sagittal, lateral and cavernous sinuses). In 1831, Bright described cavernous sinus thrombosis as a complication of epidural and subdural infections. Cavernous sinus thrombosis (CST) is the most important thrombosis compared to other intracranial thrombosis, because of its complex relationship, both neurovascular and anatomic [1]. It usually results as a complication of paranasal sinus infection [1] or infections of the face, in an area called the "danger triangle" which lies between the corners of the

mouth to the bridge of the nose including the nose itself and the maxilla) [2]. Other causes may include trauma, bacteremia or ear infections [1].

CASE REPORT

A 35-year-old female, who is on regular hemodialysis (HD) via an arm arteriovenous fistula (AVF), originally presented to the medical department complaining of high grade fever and failure of her AVF associated with a swelling and pus discharge. A diagnosis of a mycotic AVF was made and she received intravenous (IV) antibiotics for three weeks with no improvement. During that time, the patient complained of a progressively increasing periorbital swelling which became associated with complete ptosis of the right eye and partially on the left eye. The swelling was also accompanied by skin discoloration and pus discharge (Figure 1). She was referred to us for further management of her condition.

On general examination, the patient looked ill and pallor was noted on the palms. Both the radial and ulnar pulses were intact bilaterally with a pulse rate of 112 beats per minute. Her blood pressure was 105/60 mmHg and her temperature was 39.1°C (102.38°F). There was a generalized swelling on the face, more prominent periorbitally, with complete ptosis of the right eye and an incomplete ptosis of the left eye (Figure 1). The periorbital swelling was firm, ulcerated and discharging pus bilaterally. No neurological deficit was noted. The left jugular vein was prominent and distended along its course (Figure 2). Upper limbs were asymmetrical (Figure 3), the left upper limb was swollen. On the left cubital fossa, there was a pulsatile, firm swelling which was discharging pus. Blood samples and a swab from the AVF were taken for laboratory investigations which revealed signs of infection.

With the source of infection being the mycotic AVF pseudoaneurysm, the initial step of management included aneurysmectomy and AVF ligation to control the systemic infection. Postoperatively intravenous antibiotics and analgesia were administered. The fever subsequently subsided. Next, computed tomography angiogram of the head and neck showed a long segment occlusion on the left internal jugular vein extending to left brachiocephalic and subclavian veins (Figure 4) and evidence of a cavernous sinus thrombus (Figure 5); a diagnosis of a septic cavernous sinus thrombosis was made. The treatment included anticoagulant therapy, IV 3rd generation cephalosporin and periorbital eye dressing with honey and water. After the periorbital swelling and local infection subsided, she was discharged on oral anticoagulant therapy and daily eye dressing. She was later booked for debridement of a necrotic skin infection just below the right eye (Figure 6). After complete resolution of her condition, she now presents with ectropion of the right lower eye lid.

Figure 1: Bilateral periorbital swelling with skin changes.

Figure 2: Prominent left jugular vein.

Figure 3: Left arm edema.

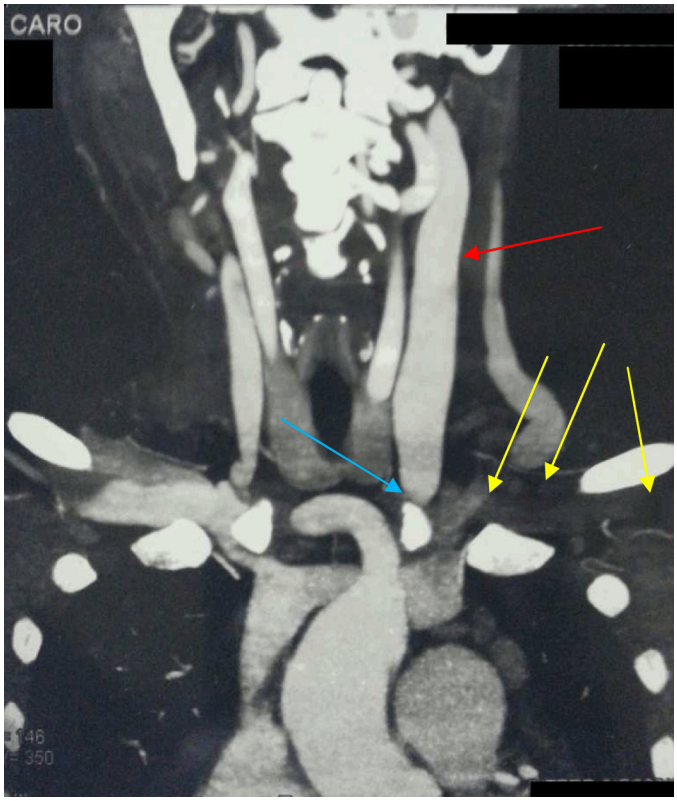


Figure 4: Subclavian and brachiocephalic vein thrombosis (yellow arrows), left internal jugular vein distension (red arrow), thrombosis (blue arrow).

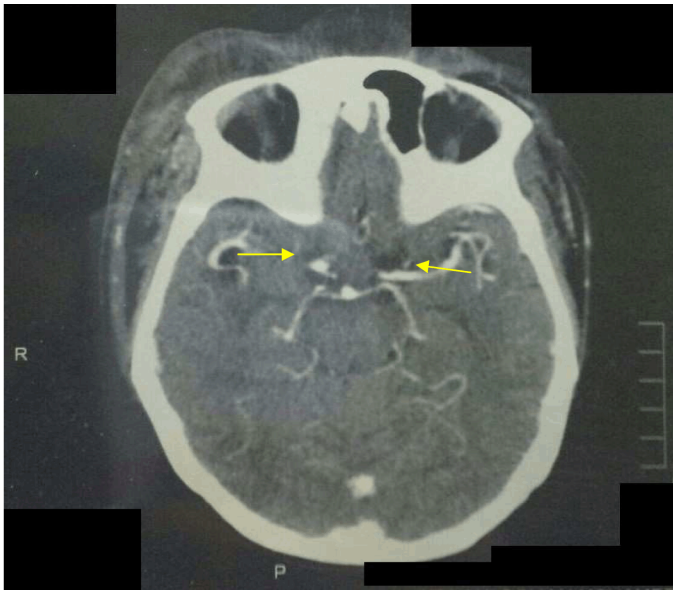
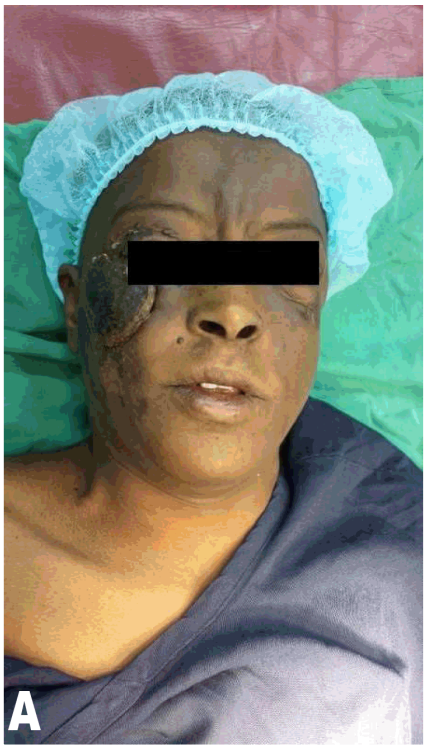


Figure 5: Bilateral cavernous sinus lesions shown on computed tomography scan of brain.



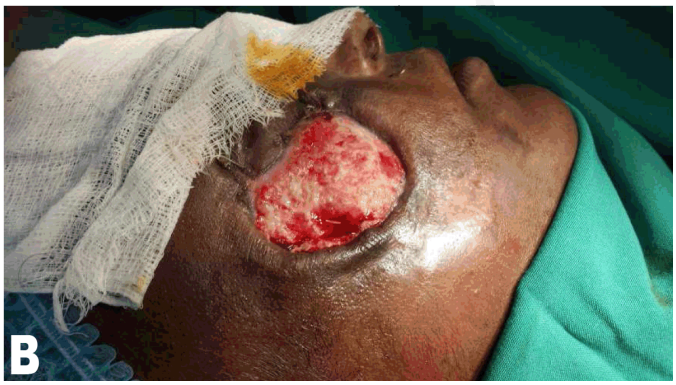


Figure 6: (A) Resolution of cavernous sinus thrombosis, and (B) Wound debridement under local anesthesia.

DISCUSSION

With regards to the anatomy, the cavernous sinuses are irregularly shaped and trabeculated cavities which, together with other sinuses, are collectively known as the dural sinuses. The cavernous sinuses lie at the base of the skull on either side of the sella turcica and of all the dural sinuses, they are the most centrally located in the skull. Relations to the cavernous sinuses include the sphenoid sinus, which lies inferomedially from the cavernous sinus and the optic chiasm, located anterior to the cavernous sinuses. Structures that pass through the cavernous sinuses include the internal carotid artery with its surrounding sympathetic plexuses. Attached to the lateral wall of the sinus are the oculomotor, trochlear and abducens nerves (cranial nerves III, IV and VI, respectively) and embedded in the wall are the ophthalmic and maxillary divisions of the trigeminal nerve. This complex and intimate relationship of vital structures within and around the cavernous sinuses accounts for the characteristic presentation of CST.

Venous blood drains from the superior and inferior ophthalmic veins to the facial veins which drain into the cavernous sinuses. The cavernous sinuses also receive venous blood from the sphenoid and middle cerebral veins. Both cavernous sinuses then drain into the inferior

petrosal sinuses which drain, via the superior petrosal sinuses, into the inferior jugular veins and sigmoid sinuses. Depending on the dominant pressure gradients, blood can flow in either direction due to the fact that this complex structure of cerebral veins contains no valves and infections usually extend from one cavernous sinus to the other because multiple connections exist between them [1].

In a study conducted by Stolic R regarding complications of AV fistulas, it was found that infections were very rare and respond well to antibiotic treatment lasting 4–6 weeks and that AVF ligature is only indicated if it is a source of recurrent septic pulmonary emboli [3]. In this case, a source of jugular vein occlusion and septic CST. Lucian et al., undertook a study in which they concluded that complications of autogenous arteriovenous fistulas are divided into two categories—acute and chronic. The acute complications included thrombosis, bleeding and hematoma formation, while the chronic complications were thrombosis, anastomotic pseudoaneurysms, venous aneurysm, venous pseudoaneurysm, skin necrosis, hand ischemia, hyperdynamic syndrome, hand edema, lymphorrhea and infection. Regarding anastomotic pseudoaneurysms and infections as a chronic complication, their rate was 0.8% and 0.1%, respectively. The formation of the pseudoaneurysm is almost always preceded by a septic process, either intraoperatively or due to an infected hemodialysis catheter or wrongful AVF cannulation techniques [4].

A case reported by Watsona and Russoa describes arterialized flow within a cavernous sinus due to an upper extremity arteriovenous dialysis fistula. The arterialized flow was due to occlusion of the left brachiocephalic vein resulting retrograde flow of blood via the left internal jugular vein. Despite it being a rare presentation, common causes for left brachiocephalic vein occlusion include venous catheter placement and malignancies [5]. In a retrospective study of 100,942 patients in a three-year period, conducted by Oymak et al., the prevalence of brachiocephalic veins and the superior vena cava (SVC) occlusion was rare. Only 33 patients (0.03%) were diagnosed with brachiocephalic or SVC thrombosis. They evaluated the causes and found that malignancies, chronic diseases, central venous lines, peripheral venous lines and thrombophilia came in at 42%, 39%, 27% and 38%, respectively. The manifestations included arm, head and neck swellings in 97% of the patients, which coincide with the findings in the patient being reported, and pulmonary embolism in 36% of the patients. No accounts of cavernous sinus thrombosis were observed [6]. Otten et al. performed a study of nine patients with brachiocephalic vein occlusion and found that three of these patients had AV fistulas as their means of dialysis with no history of central lines. They concluded that a sufficient number of patients develop brachiocephalic vein occlusion to consider a diagnosis, especially in patients with malignancies or central venous lines [7]. The list of complications provided in an article by Mueller regarding internal jugular vein thrombosis included pulmonary embolism, subclavian vein and superior sagittal sinus thrombosis, superior vena cava syndrome, pseudotumor cerebri, laryngeal and lower airway edema and infected thrombophlebitis, but again, no mention was given regarding CST as a complication [8]. Boedeker et al. described, in general, the complications of internal jugular vein thrombosis, which included pulmonary embolism, sepsis with septic emboli to different organs and tissues and thrombus propagation intracranially [9].

CONCLUSION

Cavernous sinus thrombosis (CST) can result as a complication of infections within the danger triangle of the face or paranasal sinuses, trauma, bacteremia or ear infections. Brachiocephalic and jugular vein thrombosis propagation is another important cause. Most importantly, in patients presenting with a mycotic arteriovenous fistula (AVF), the risk of developing a septic CST as a complication should be kept in mind and intervention, either conservative or surgical, should be based upon the risks versus benefits of either form of management and prevention of further complications.

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Ahmed Mohamed Elhassan Elfaki Osman – Acquisition of data, Drafting the article, Final approval of the version to be published

Saif-Eldin Mohammed Ali Ibrahim – Substantial contributions to conception and design, Revising it critically for important intellectual content, Final approval of the version to be published

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CASE REPORT OPEN ACCESS | PEER REVIEWED

Vulvar myiasis: A rare case report

Savita Singhal, Parul Bhugra

ABSTRACT

Introduction: Myiasis is a parasitic infestation of tissues of living vertebrae by the larvae of dipterous flies. The condition usually occurs on exposed part of the body. The location of myiasis is very rarely seen on the vulvar area. Case Report: We present a case of vulvar myiasis affecting a 72-year-old female. The patient was investigated for any predisposing factor. The larvae were removed after application of turpentine oil. Conclusion: Genital myiasis is very uncommon. Awareness of the disease and careful examination of the lesion is important for making correct diagnosis.

Keywords: Infestation, Larvae, Myiasis, Vulvar

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INTRODUCTION

Human myiasis is caused by fly larvae capable of penetrating body orifices as well as healthy or necrotic tissues. Myiasis is more common in rural areas. In the urban areas, the disease is usually found among people with poor hygienic habits and low education level. Although human myiasis occurs rarely, it probably occurs at a higher rate than is currently thought. A careful examination is needed to identify vulvar myiasis as it can be confused with many common diseases such as insect bite, cellulitis, etc.

CASE REPORT

A 72-year-old female presented in the gynecology outpatient department of our institute with the chief complaint of pain over the genital area for five days. The pain was severe, intermittent, throbbing in nature, associated with a crawling sensation and discharge from the genital area. The discharge was blood stained and foul-smelling. General physical and systemic examination revealed no abnormalities. On local examination, there was ulcer of size approximately 5x3 cm, irregular in shape, involving right labia majora, labia minora and vagina (Figure 1). Floor of the ulcer was covered with foul smelling slough with numerous maggots. Maggots were also filling the vagina. On palpation, the surrounding area was tender, edge and base of the ulcer was indurated. Inguinal lymph nodes were enlarged and showed the signs of acute lymphadenitis. The patient was admitted for investigations and treatment. The patient was found to be HIV and VDRL non-reactive. Her blood sugar level was within normal limits. The culture from the swab taken from the vulva and vagina showed mixed flora and no sexually transmitted organisms were identified. Histopathological examination of the biopsy specimen from the ulcer edge showed no evidence of malignancy. The treatment was started with antibiotics and analgesics. The larvae were removed manually with the help of forceps after application of turpentine oil and the lesion was cleaned thoroughly. Daily antiseptic

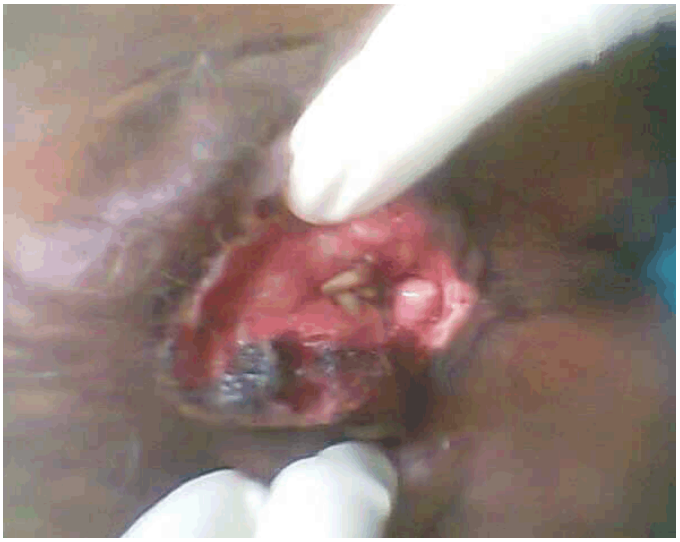

Figure 1: Vulvar myiasis.

dressing and turpentine oil application continued for the presence of any deeply burrowed larvae. The patient was discharged after four days of hospital stay on antibiotics and analgesics.

DISCUSSION

Myiasis is a zoonosis resulting from an infestation by the larvae of flying dipterous insects. There are two forms of myiasis: obligate, in which the larvae feed themselves on living tissues and facultative myiasis, where the larvae opportunistically take advantages of wounds or degenerative necrotic conditions [1]. Depending on the body parts involved, myiasis can be classified as mucocutaneous, ophthalmic, nasal, nasopharyngeal, intestinal or urinary tract myiasis.

The incidence of myiasis is probably underestimated as it occurs principally in rural areas where cases are usually not registered. Greenberg identified, four risk factors for infestation by larvae (i) helpless or debilitated person (ii) bleeding or odors of decomposition present (iii) summer season and (iv) neglect in nursing care [2]. Acquired immunodeficiency and the habit of some populations of sitting on the ground, for example, during certain religious rites are other predisposing factors [3]. The larvae can enter through intact skin or a wound. Myiasis may also be caused by the deposition of the ova on the wounds, clothes or by the ingestion of eggs of obligatory myiasis producing flies [4].

The disease occurs mostly on uncovered parts of the body. Infestation is very rarely seen in the vulvar area. The possible modes of vulvar infestation has been suggested to be through (i) fomites, especially with the washed undergarments being dried in the open sunlight on which flies may lay their eggs (ii) due to deposition of the ova on the genital region because flies were attracted by the odor caused by a lack of proper hygiene or by the foul smell caused by multiple infections and the patient did not wear undergarment while sleeping, after sexual intercourse or because the use of undergarment is uncomfortable because of already present genital infection [5, 6].

Mucocutaneous myiasis is of three types, furuncular myiasis, traumatic myiasis and creeping myiasis. In the furunculoid form, over a period of several days a painful furuncle develops in which the larvae is present. The lesion has a central punctum, which discharges serosanguineous fluid. The lesions may be accompanied by lymphangitis and regional lymphadenopathy. Traumatic or wound myiasis may be seen in neglected ulcers or wounds. The larvae (maggots) can be seen, often in large numbers in the suppurating tissues. In creeping myiasis, an itching pink papule develops, followed by a tortuous line that extends by 1 to 30 cm a day [4, 7].

Mucocutaneous myiasis can be misdiagnosed as adenopathy, cellulitis, skin abscess, insect bite and subcutaneous cysts [8].

The aim of the treatment is to remove the larvae and treat any secondary bacterial infection. In wound myiasis, the larvae can be removed mechanically with forceps and preferably, destroyed to prevent them from pupating. When larvae are burrowed onto deeper tissues, they can be made to come out by application of gauze soaked in turpentine oil. Furuncular myiasis may be treated by occlusion of the opening of the furuncle with animal fat, paraffin, mineral oil or petrolatum. After this maggots can be removed with the aid of surgical tweezers and the nodule may be gently expressed. Sometimes, surgical incision may be required to extract the larvae. If a surgical approach is used, the incision should avoid lacerating the larvae, as the retained larval parts may precipitates a foreign body reaction. In a creeping eruption, the larvae can be extracted with a needle [6, 9].

CONCLUSION

The location of myiasis at genital region is a very rare occurrence. Knowledge of the characteristic clinical findings and the close inspection of lesions are key to diagnosing myiasis. Pain, swelling and redness can easily be mistaken for sexually transmitted infections. With the correct diagnosis, unnecessary antibiotics and surgical procedures can be avoided.

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Author Contributions

Savita Singhal – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Parul Bhugra – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published

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The corresponding author is the guarantor of submission.

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CASE REPORT OPEN ACCESS | PEER REVIEWED

Internal hernias: Emergency department radiological dilemma

Cigdem Ozpolat, Halil Ibrahim Atalay, Sefer Ozkaya, Musa Adanc, Arzu Denizbasi, Ozge Onur, Serkan Emre Eroglu, Haldun Akoglu

ABSTRACT

Introduction: Internal abdominal hernias present a non-specific clinical presentation. Clinical diagnosis is often difficult so imaging studies. It plays an important role in the early diagnosis. Its diagnosis remains difficult even after the computed tomography (CT) scans disseminated accessibility and use. We present a case that was confirmed CT scan and treated with subsequent surgery. Case Report: A 37-year-old female presented to the emergency department with diffuse abdominal pain, developing suddenly an hour ago. In her physical examination, there was diffuse tenderness, and rebound and guarding in right upper and lower quadrants. A CT scan of the abdomen and pelvis with intravenous contrast demonstrated dilatation and left migration of the loops of jejenum, migration of inferior mesenteric vein to the left, and free fluid in abdomen. In the operation, it was seen that ileum was herniated around cecum (paracecal hernia). Conclusion: Internal abdominal herniations are rare conditions. Clinical presentation may be non-specific, and diagnosis is difficult if it is not thought. But delayed diagnosis results in increased mortality and non-viable intestinal tissue. So emergency physicians should aware of this condition and radiological images for early surgery consultation.

Keywords: Internal herniation, Intestinal obstruction, Paracecal hernia, Radiological dilemma

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INTRODUCTION

An internal hernia is a rare condition defined as the protrusion of abdominal viscera into one of the fossae, foramina, recesses, or congenital defects within the abdominal and pelvic cavity [1]. The sex ratio exposed a male prevalence of 3:2 [2]. The aperture can be normal, encased with a sac or either abnormal, not possessing a sac. Congenital anomalies due to improper intestinal rotation, previous trauma, vascular or inflammatory diseases, or postsurgical iatrogenic are predisposed factors to internal herniation. Internal hernias are generally classified into six types: paraduodenal, pericecal, foramen of Winslow, transmesenteric, pelvic and supravesical, and intersigmoid [2]. Internal hernias may present as intestinal obstruction and account for 0.5–4.1% of all cases [3]. Congenital and acquired defects in the mesentery of the cecum or appendix, may lead to development of a pericecal hernia. Anatomically, there are four types of peritoneal recesses of various sizes and depths identified in the pericecal region, including the superior ileocecal recess, inferior ileocecal recess, retrocecal recess and paracolic sulci [4, 5]. Internal abdominal hernias present a non-specific and intermittent clinical presentation.

Therefore, clinical diagnosis of internal hernias is often difficult and thus imaging studies play an important role in the early diagnosis. Its diagnosis remains difficult even after the computed tomography (CT) scans disseminated accessibility and use. We present a case of atypical presentation of acute abdomen and was diagnosed as internal herniation with CT scan, thereafter confirmed with subsequent emergency surgery.

CASE REPORT

A 37-year-old female presented to the emergency department with diffuse abdominal pain, developing suddenly an hour ago. The pain was severe, not colicky and radiated through to her back. Sitting upright and leaning forward relieved the pain. She was also complaining from nausea and vomiting. She was passing flatus. Before, she had not experienced any similar pain. Her vital signs were normal. In her physical examination, there was diffuse tenderness, and rebound, guarding in right upper and lower quadrants. In blood gas analysis, her venous blood pH: 7.19, HCO_3: 16 mmol/L, CO_2: 47 mmHg, lactate 4.2 mmol/L, base excess: 10.6 mmol/L. In her total blood count, white blood cell was 13400/L. Her liver function tests were minimally elevated. As her pain was persistent and unexplained. A computed tomography scan of the abdomen and pelvis with intravenous contrast demonstrated dilatation and left migration of the loops of jejenum, migration of inferior mesenteric vein to the left, and free fluid in abdomen (Figures 1, 2). With the prediagnosis of internal herniation, fluid resuscitation and supportive care were initiated for preparation for surgery. In the first, operation laparotomy with mid line incision made, 500 cc fluid was aspirated, it was seen that 50 cm segment of the terminal ileum was herniated medial to cecum (paracecal hernia), ileum was extracted from the defect. It was seen that loops of jejenum and terminal ileum up to 50 cm proximal to ileocecal valve were dilated and viable. Hot compresses were situated. Resection was not applied due to revascularization of ischemic loops was seen after 10 minutes. The defect was repaired and operation was terminated. After three days, patient deteriorated; in the exploration ischemia was detected beginning from 10 cm proximal of terminal ileum up to the 200 cm ileum. Ischemic loops were dissected, serosal tearing in ileum and sigmoid colon repaired. Efferent and afferent tips together subtracted from right upper quadrant. After four days, probably from micro perforations and ventilator associated pneumonia, in blood culture first methicillin resistant *Staphylococcus auerus then Pseudomonas aeruginosa* proliferated, despite of full septic shock treatment (fluid therapy; vancomycin, clindamycin, gentamycin antibiotic therapy; vasopressors with noradrenalin and dopamine), the patient died in intensive care unit due to sepsis and multi-organ failure.

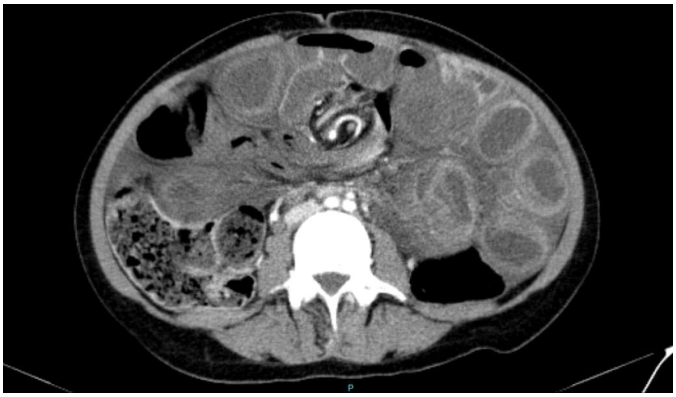

Figure 1: Axial section demonstrating 'Whirlpool sign'.

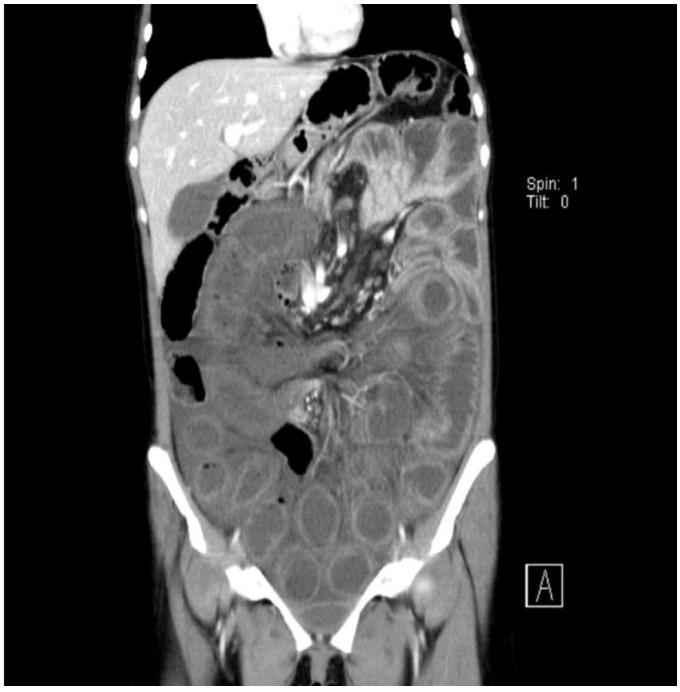

Figure 2: Coronal slice showing dilated and left migration of the loops of jejenum.

DISCUSSION

In autopsies, internal hernia has been reported to range from 0.2–0.9% [2]. In the reality, we know that 5.8% cases of small bowel obstruction are due to internal hernia [4]. Internal herniation should be considered as a differential diagnosis in patients presenting with symptoms of small bowel obstruction without a history of prior abdominal surgery. Normally, standard hernias results from defects in the retaining walls of the abdomen. But internal hernias are due to the organ protrusion through an opening or pouch of the peritoneum. Patient may be asymptomatic or may have ileus symptoms and findings like constant epigastric pain or intermittent colicky periumbilical pain. There may be nausea or vomiting. The severity of the pain is related to the presence of ischemia or necrosis. As no specific symptoms are associated with the condition, it is rarely diagnosed preoperatively. Clinical examination is non-specific and laboratory findings are rarely helpful.

Although the occurrence is rare, delayed diagnosis and treatment are associated with a high mortality rate. Abdominal CT scan is important for intestinal obstruction [6]. Computed tomography scan gives information about location of obstruction, the possible underlying causes, such as malignancy, stenosis within the bowel wall, and other intraluminal problems such as intussusception, feces or or Bezoar like substances [7]. Computed tomography scan has become the first-line imaging technique in patients with suspected acute abdomen. Etiologies of acute abdomen like internal hernias are often difficult to identify with physical examination [8]. Computed tomography scan showed classic signs of internal herniation as 'Whirlpool sign', crowding of bowel loops in the upper compartment and the absence of cecum in the right iliac fossa, replacement of abdominal organs and vasculature. If strangulation of the intestine is suspected including other causes also, an internal hernia should be considered as in our case. Delay in diagnosis and treatment is often observed in internal hernia cases and results high mortality rate of up to 49% [9]. In the operation, often intestinal obstruction associated with non-viable bowel detected. So the treatment invariably requires urgent surgery. Usually, open surgery is performed. Only a few cases of laparoscopic hernia management have been reported [10]. If there is intestinal necrosis, an adequate resection is mandatory. Nevertheless, there is no clear and established consensus on surgical management when the herniated contents are grossly viable.

CONCLUSION

In the symptoms of intestinal obstruction, a high index of suspicion for internal hernia is necessary to prevent diagnostic delay and mortality in emergency departments.

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Author Contributions

Cigdem Ozpolat – Substantial contributions to conception and design, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published
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Haldun Akoglu – Acquisition of data, Revising it critically for important intellectual content, Final approval of the version to be published

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The corresponding author is the guarantor of submission.

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CLINICAL IMAGES OPEN ACCESS | PEER REVIEWED

Renal carcinoma complicating autosomal dominant polycystic kidney disease

Francisco Rivera, Celia López Redondo

CASE REPORT

On February 2013, a 76-year-old male was sent to our nephrology unit to evaluate renal disease. He was asymptomatic and had been diagnosed with hypertension, overweight, benign prostatic hypertrophy and chronic obstructive pulmonary disease several years ago. His one daughter was also diagnosed with a renal cyst. His serum creatinine was 1.6 mg/dL, eGFR 42 mL/min, albumin/creatinine ratio in spot urine sample 2.8 mg/g and normal urinary sediment. Ultrasonography showed a slight enlargement of both kidneys and the presence of multiple bilateral cysts, predominantly with cortical distribution, classified as Bosniak I. No complex cysts that required monitoring or solid lesions were found (Figure 1). Therefore, he was diagnosed with Autosomal Dominant Polycystic Kidney Disease (ADPKD) and follow-up was drawn. He was successfully treated with losartan 50 mg/day. On July 2014, a new ultrasound control revealed the appearance of an echogenic nodule on the upper pole of the right kidney with vascularization in Doppler mode (Figure 2). The study was completed by computed tomography scan that confirmed the presence of a solid nodule of 23 mm on the right kidney with early contrast enhancement after i.v. iodine contrast administration. These findings strongly suggested the existence of superimposed renal cell carcinoma (RCC) (Figure 3). Considering his age and co-morbidities conservative treatment was planned.

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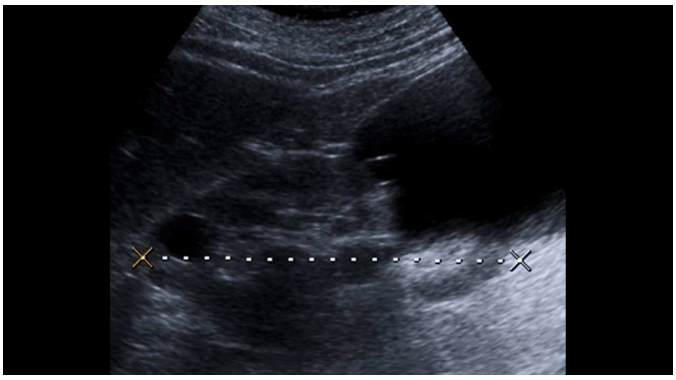

Figure 1: Ultrasound image of right kidney showing multiple cysts with cortical distribution, classified as Bosniak I.

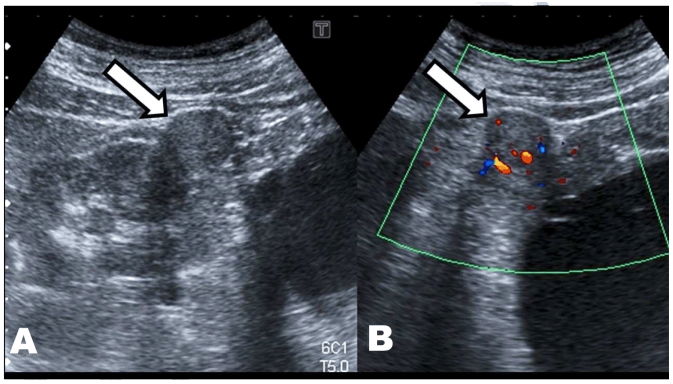

Figure 2: Ultrasound image of the upper pole of right kidney. (A) B-mode image with an echogenic nodule, with rounded morphology (white arrow), (B) Doppler mode reveals the presence of vascularization in this nodule (white arrow).

DISCUSSION

The association between RCC and ADPKD has been described although this association has raised some controversy. While several case reports of RCC complicating ADPKD have been described and it has been found premalignant epithelial cells in the cyst epithelia, epidemiologic and autopsy studies have not shown a significant higher incidence of RCC in ADPKD [1, 2].

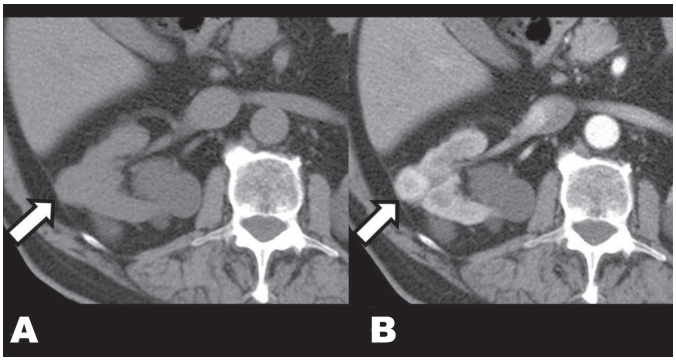

Figure 3: (A) Computed tomography scan showing without intravenous contrast: exophytic isodense nodule compared to the rest of the renal parenchyma (white arrow), (B) Computed tomography scan in arterial phase after the administration of intravenous contrast: early enhancement predominates at the periphery of the nodule (white arrow).

On the other hand, more recent studies have described that kidney-related prevalence of RCC in patients with ADPKD and advanced chronic renal failure treated by hemodialysis or renal transplantation was high [3, 4]. These apparent discrepancies could be explained by the difficulties in the diagnosis of RCC in this setting and emphasizes the importance of close clinical surveillance and the interpretation of several radiological studies such ultrasonography, CT scan and MRI scan [5, 6].

Herein, we report a case of RCC complicating ADPKD who has several characteristics. Firstly, he did have neither hematuria nor symptoms of occult neoplasia. Secondly, renal function was decreased although dialysis was not needed. Finally, the combination of ultrasonography and unenhanced and contrast-enhanced CT scan studies were able to achieve a diagnosis without using RMI, as it has been recently recommended.

CONCLUSION

Renal cell carcinoma (RCC) can appear as a complication of autosomal dominant polycystic kidney disease (ADPKD). The diagnosis of RCC in this setting needs a thoroughly radiological evaluation, which should include ultrasonography and computed tomography scan studies.

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Celia López Redondo – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

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CLINICAL IMAGES OPEN ACCESS | PEER REVIEWED

Progressive hearing loss: A case report on surfer's ear

Paresh Kushta Dessai, Sapna Sada Raut Dessai

CASE REPORT

A 30-year-old male reported with the complaint of progressive bilateral hearing loss over a period of six months. There were no associated symptoms of otalgia, tinnitus, discharge or bleeding from the ears. Patient also could not recall any history of trauma to the face. ENT surgeon, who referred the patient for computed tomography (CT) evaluation of hearing loss, had performed the otoscopy and audiometry examinations. Otoscopic findings had revealed, a narrow external auditory canal, tympanic membrane was barely visualized on both sides and was intact. Bilateral moderate conductive hearing loss was noted. He was then advised high resolution computed tomography (HRCT) scan of the temporal bone.

Following findings were noted on HRCT. Broad-based osseous overgrowth of external auditory canal bilaterally, were noted on axial CT images. (Figure 1). Sagittal CT images showed evidence of dense bony protuberances arising from the tympanic and the petrous bone into the external auditory canal space. This resulted in significant narrowing of the ear canal and thus conductive hearing loss bilaterally (Figure 2). On obtaining a detailed history, it was noted that the patient was a frequent swimmer. Thus, the diagnosis of surfer's ear was made. The patient was referred for surgical management of the exostoses.

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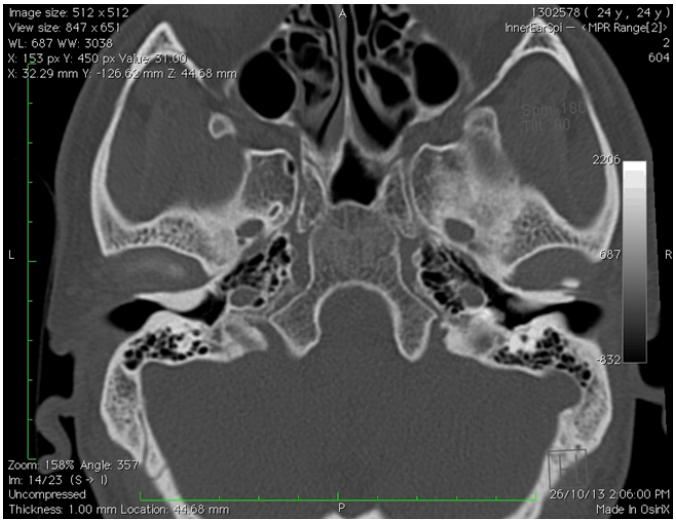

Figure 1: High resolution computed tomography axial image of bilateral temporal region showing bony outgrowth in the external auditory canals.

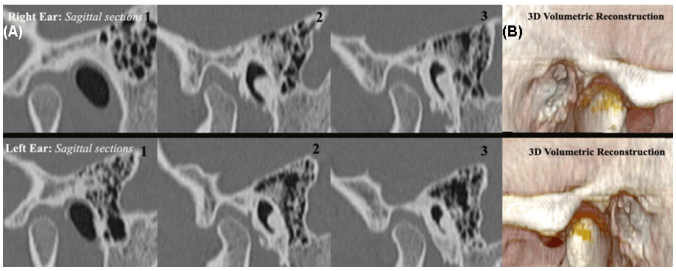

Figure 2: (A) High resolution computed tomography image showing multiple sagittal sections through the external auditory canals showing bony outgrowth, (B) 3D volumetric reconstruction.

DISCUSSION

Conductive hearing loss is commonly secondary to impaction by cerumen, foreign body, inflammation, neoplasm, or bony outgrowth such as exostoses. Exostoses are benign bony protuberances, arising from the osseous portions of the external auditory canal [1]. They are mostly seen in individuals engaging in aquatic activities like frequent surfing [2]. Their growth is believed to be due to many years of repeated exposure to cold water and

wind. The cold stimulates bone growth and the ear canal gradually narrows thus avoiding water from reaching the eardrums.

Such exostoses is believed to be worse in the ear that faces the ocean at the time of "catching the wave." The condition is commonly called 'Surfer's Ear' due to its high prevalence amongst surfers [3, 4]. It is usually bilateral, and located close to the tympanic annulus at the tympanomastoid and tympanosquamous sutures [1, 5].

The new bone that grows is more sensitive than the original bone [6]. Hyperplasia continues painlessly as the ear is continuously hit by the cold waves during aquatic activities. Water and debris can get trapped behind the bony growth and ears can get easily blocked and infected. The bony growth can completely fill the canal. Thus, the patient may present with conductive hearing loss and recurrent episodes of external otitis with otalgia and cerumen impaction. When otoscopic examination is performed to determine patency of external auditory canal, one or more broad based elevated lesions that protrude into the external auditory canal will be evident.

The HRCT of the temporal bone is the examination of choice [7, 4] as it provides detailed osseous anatomy of the temporal bone. Intravenous contrast is not required to make the diagnosis. Radiographic differential diagnosis includes external auditory canal osteoma [1]. Clinically, it needs to be differentiated from medial canal fibrosis, necrotizing external otitis, external auditory canal cholesteatoma and keratosis obturans [1]. On imaging, these entities are visible as soft tissue lesions with or without bone destruction.

Osteomas differ from exostoses as they are usually unilateral, pedunculated benign tumors [7]. They are composed of densely sclerotic, well-formed bone jutting out from the cortical surface. External auditory exostosis lesion is found as solitary sessile bony growths and usually lateral to the isthmus. On histopathologic examination, they are seen as dense concentric layers of subperiosteal bone originating from near tympanic ring [3].

A history of cold water and wind exposure facilitates the diagnosis Surfer's ear. It is believed that there is a positive association between the amount of time spent by an individual in surfing and the presence and severity of exostoses of the external auditory canal. In 1937, Van Gilse postulated a thermal cause for the development of external auditory exostoses after observing a higher frequency of this pathological condition among cold water versus warm water swimmers [3]. Some in the past have considered salt-water exposure to be the cause but this was proved untrue by a study, which showed evidence of external auditory canal exostosis even in freshwater, seawater and non-swimmers. Water temperature has been found to be a major cause rather than water salinity as it causes meatal erythema [3].

Treatment

When the ear canal is significantly narrowed a surgical procedure may be required to re-open the canal [4, 5]. It is important to stay out of the water until the ear canal is completely healed. Hearing testing is performed before and after the surgery. Surgery complications include canal stenosis, temporomandibular joint prolapse, sensorineural loss, persistent deep bony lip, and persistent tympanic membrane proliferation, facial nerve injury [6].

CONCLUSION

Regular surfers and divers should be advised to avoid very cold water. They should be advised to use earplugs or wetsuit hood. The custom plugs may be fabricated which have the advantage of staying in very well. An alcohol-based swimmers eardrop can help dry any residual moisture in the canal after water exposure. Surgery for exostoses should be performed carefully to prevent complications.

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CLINICAL IMAGES OPEN ACCESS | PEER REVIEWED

Small bowel obstruction due to rice cake

Hideki Katagiri, Tetsuo Nakata, Toshikazu Matsuo

CASE REPORT

A 67-year-old male with no significant past medical history was referred to the hospital complaining of abdominal pain. Eighteen hours prior to admission, he ate four pieces of rice cake (mochi in Japanese) and after six hours, he ate four pieces of rice cake again. Nine hours prior to admission, he noted intermittent abdominal pain with one vomitus. He visited his general practitioner. Physical examination at the general practitioner suggested small bowel obstruction. He was referred to the hospital for further investigation. On physical examination, he had mild tenderness in upper abdomen without peritoneal signs. A small bowel obstruction due to rice cake was suspected. The abdominal computed tomography (CT) scan, revealed high density materials in the stomach and the small intestine with proximal intestinal dilation (Figure 1A–B). This typical appearance made the final diagnosis of small bowel obstruction due to rice cake and he admitted to the hospital for observation. His symptoms disappeared without specific treatment and he discharged from hospital on the third day from admission.

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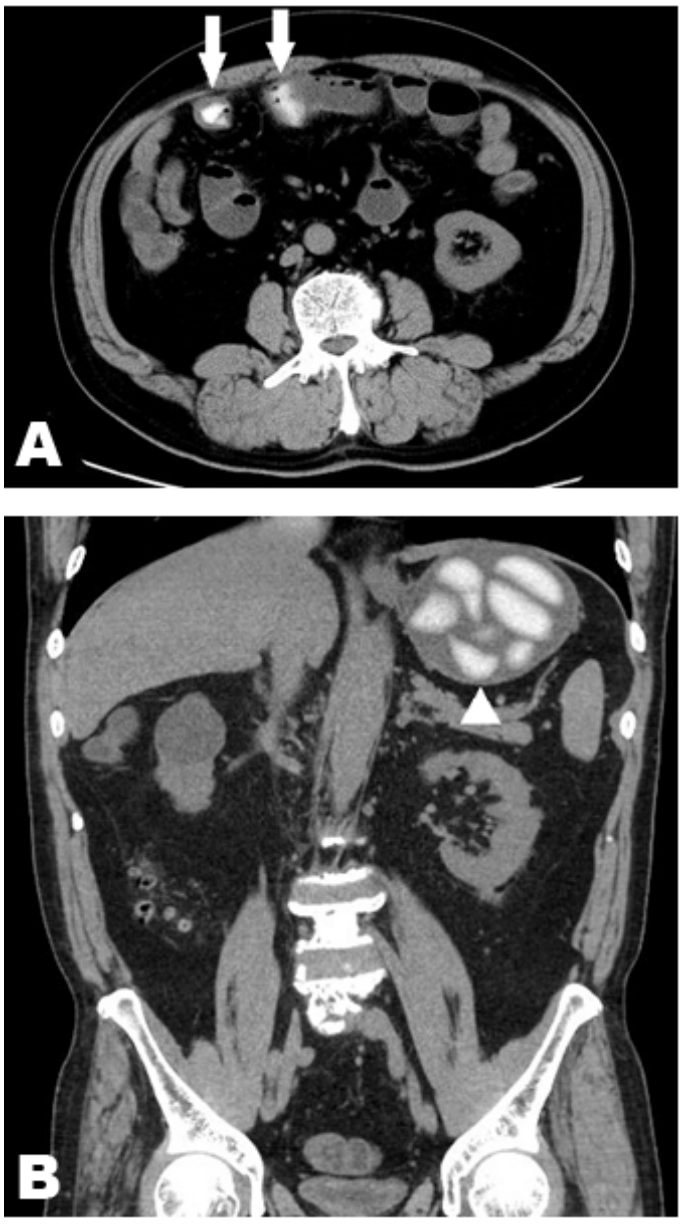

Figure 1: (A, B) Computed tomography scan of axial and coronal views revealing high density materials in the stomach (arrow head) and in the small intestine (arrows) with proximal intestinal dilation.

DISCUSSION

Small bowel obstruction (SBO) is a common disease encountered in daily practice. The causes of SBO can vary. In the review of unusual causes of SBOs, 60% of them are caused by adhesions, followed by hernias 15%, neoplasms 6%, inflammatory causes 5%, mesenteric vascular occlusion 5%, intussusception 3%, and unusual etiology 6%. [1]. Matsuzaki et.al. reviewed 193 patients of SBOs induced by food in Japan [2]. The most frequent causative food is seeds or stones fruit, followed by rice cakes, seaweed, agar or gelatin food made from devil's tongue starch. The patients are often associated with previous abdominal surgery, especially gastrectomy. Dental disorders such as problems with chewing or artificial teeth, unusual eating habits like swallowing without chewing, and irradiation to abdominal cavity can also be contributing factors.

Rice cakes are often eaten in Japan. Especially in the New Year's Holidays, because Japanese people have a tradition to eat rice cakes in those holidays. Miura et al. reviewed 14 patients of SBO caused by rice cake [3]. Ten out of fourteen patients have a history of previous abdominal surgery and all patients had ingested rice cake by swallowing without chewing. In this patient, interestingly, he had no contributing factors like previous abdominal surgery or unusual eating habits.

The diagnosis of SBO induced by food is generally difficult. However, the diagnosis of SBO due to rice cake can easily be obtained by CT scan. The rice cakes are typically visualized as high density materials in the alimentary tract [3].

Treatment of rice cake induced SBO should be non-operative management, with occasional decompression by a nasogastric tube and fluid supply. All patients can be successfully treated by non-operative management in the recent review, as in the present patient [3]. However, chronic symptom could possibly cause ulcer or intestinal perforation so careful following-up should be considered.

CONCLUSION

Small bowel obstruction due to rice cake is a relatively rare disease. Careful history taking is essential and computed tomography scan demonstrates typical appearance of high density materials.

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Author Contributions

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Tetsuo Nakata – Analysis and interpretation of data, Critical revision of the article, Final approval of the version to be published
Toshikazu Matsuo – Conception and design, analysis and interpretation of data, Critical revision of the article, Final approval of the version to be published

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The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

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Acute organoaxial gastric volvulus

James Heaton, Andrew Gilliam

CASE REPORT

A 73-year-old male presented to the emergency department with a one-day history of severe epigastric pain with vomiting progressing to dry retching. He admitted to a long history of esophageal reflux symptoms treated with oral omeprazole. His initial examination revealed a tachycardia and a swollen, tender epigastrium but no other signs of note. A nasogastric tube was passed with difficulty, he was made nil by mouth and treated with intravenous crystalloids. Laboratory blood results were unremarkable including normal amylase and liver function tests.

A chest X-ray showed a large retrocardiac viscus reported as a large hiatus hernia while his abdominal film demonstrated a paucity of bowel gas (Figure 1). An abdominal and thoracic computed tomography (CT) scan revealed a moderate hiatus hernia without obvious perforation and the possible appearance of a rotational component of the stomach with dilatation to the pylorus and no fluid beyond this point. A subsequent upper gastrointestinal contrast study clearly demonstrated an 'upside-down stomach' sign and established the diagnosis of an obstructing organoaxial volvulus secondary to a paraesophageal hiatus hernia (Figure 2) [1]. On transfer to our facility the patient went on to have definitive laparoscopic gastropexy surgery involving reduction of

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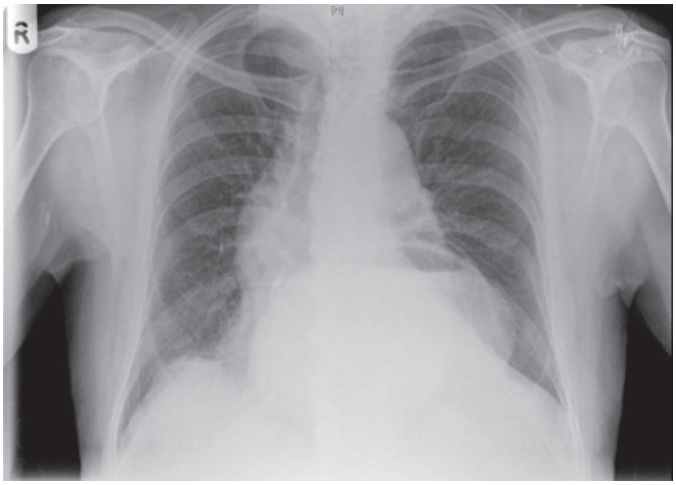

Figure 1: Chest X-ray showing a retrocardiac viscus with an air/fluid level.

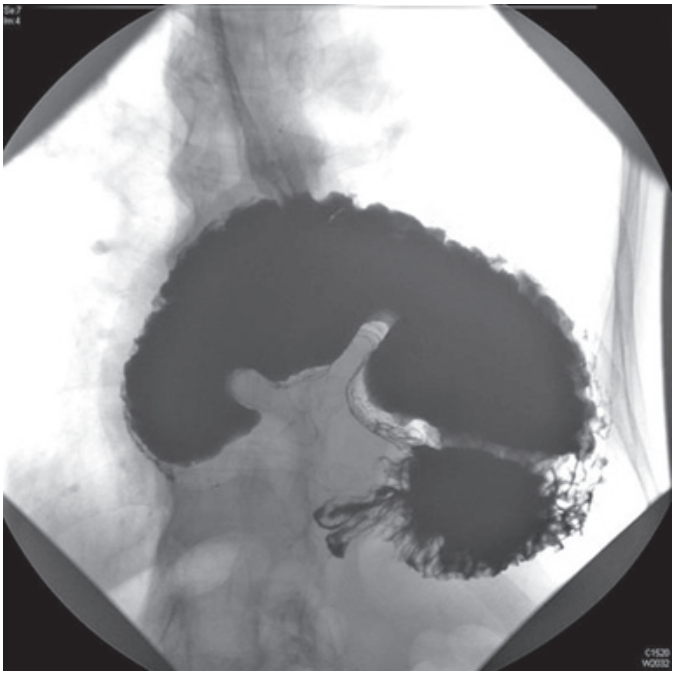

Figure 2: Upper gastrointestinal contrast study clearly demonstrating the 'upside-down stomach' sign of organoaxial volvulus.

the volvulus, excision of the hernia sac, re-approximation of the diaphragmatic crura then placement of four sutures anchoring the greater curvature of the stomach to the abdominal wall. He has had no recurrence of the volvulus and was symptom free when followed-up in clinic for six months.

DISCUSSION

Gastric volvulus is an abnormal rotation of the stomach through more than 180 degrees, first described by Berti in 1866 [2]. This can lead to ulceration, perforation, hemorrhage, ischemia or necrosis [1]. The non-operative mortality rate is as high as 80% [3].

Adults with acute gastric volvulus typically present with epigastric pain and distension, unproductive vomiting and difficulty with nasogastric tube insertion. A constellation known as Borchardt's triad [4]. About 10–20% of cases occur in children, in adults it can occur at any age but is more common after the fourth decade of life [1, 5].

Gastric volvulus can be classified according to the axis around which the stomach rotates. In organoaxial volvulus, the stomach rotates around an axis connecting the gastroesophageal junction with the pylorus. This is the most common type of gastric volvulus occurring in approximately 60% of cases and commonly leads to strangulation and necrosis [6]. In mesenteroaxial volvulus, there is a transverse axis and the antrum rotates antero-superiorly so that the posterior surface of the stomach lies anteriorly. It is also possible to have a combined type volvulus. The most common causes of gastric volvulus in adults are diaphragmatic defects. In the case of paraesophageal hernia related volvulus, as we report, the gastroesophageal junction remains in the abdomen, whereas the stomach ascends adjacent to the esophagus, resulting in a horizontally lying, upside down stomach [2].

X-ray appearances include a retrocardiac gas/fluid filled viscus on chest film if the stomach is in the thorax and a paucity of distal gas on plain abdominal film [7]. Several authors recommend computed tomography imaging as the diagnostic method of choice, this may show a torted bi-lobular stomach with a transition line [2, 8, 9]. However, the diagnosis of gastric volvulus is classically based on upper gastrointestinal contrast studies using barium or Gastrografin. These studies are both sensitive and specific if performed in the twisted state and classically show an 'upside-down stomach' sign as well as illustrating the degree of obstruction [8].

Endoscopic reduction of gastric volvuli is possible but recurrence rates are high if this is performed as an isolated procedure [2]. Surgical repair was traditionally based on an open approach but this has been superseded by modern minimally invasive techniques. Laparoscopic suture gastropexy, as described in our case, is safe and effective for both acute and chronic gastric volvulus [1-3].

CONCLUSION

Acute gastric volvulus is a rare surgical emergency with high rates of non-operative mortality. Prompt diagnosis and urgent surgery is crucial to avoid life-threatening complications associated with this condition. A purely clinical diagnosis is challenging but the condition should be suspected in a patient who presents with abdominal pain and distension, unproductive vomiting and a difficult to place nasogastric tube. Although a computed tomography scan may prove useful our case report clearly demonstrates the power of upper gastrointestinal contrast studies in establishing a definitive diagnosis.

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James Heaton – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Final approval of the version to be published

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