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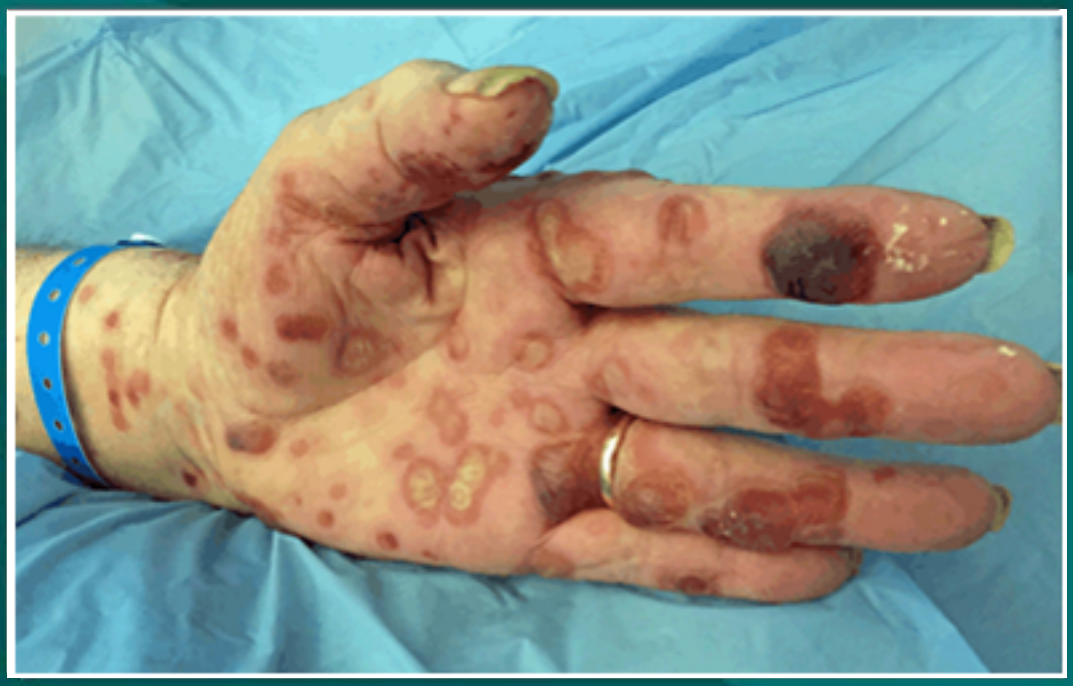

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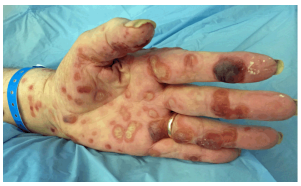

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Numerous clustered 5 mm to 2 cm clear smooth vesicles and tense bullae on the palmar surface with no nail involvement.

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CASE SERIES

PEER REVIEWED | OPEN ACCESS

Respiratory failure in adults due to foreign body aspiration: A case series

Lycke R. Woittiez, Elsbeth J. Wesselink, Marcel A. de Leeuw, Cornelis Slagt

ABSTRACT

Introduction: Foreign body aspiration (FBA) is rare in adults and its clinical presentation can be very diverse. Acute symptoms as dyspnea and choking are often immediately linked to FBA. However, mild or even asymptomatic chronic pulmonary symptoms can be presented as a result of FBA. Physical examination is usually nonspecific. Chest X-ray is often normal or shows nonspecific findings. Treatment and definite diagnosis can be accomplished using rigid or flexible bronchoscopy. Case Series: We present two cases of foreign body aspiration. The first case was the aspiration of a broken tracheostomy tube leading to acute respiratory failure and the second case was the aspiration of a medication blister which initially presented as atypical chronic pulmonary symptoms but evolved to a medical emergency of acute respiratory failure. Conclusion: These two cases show the broad range of symptoms and findings associated with FBA. When patients present with nonspecific pulmonary findings, FBA should be included in the differential diagnosis.

Keywords: Aspiration, Bronchoscopy, Chest X-ray, Foreign body, Pulmonary medicine, Respiratory failure

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INTRODUCTION

Foreign body aspiration (FBA) occurs frequently in children, but rarely in adults [1]. In different series where both children and adults with FBA were included, children represented 46–92% of the total study group [2, 3]. Most adult patients who experience FBA have predisposing conditions resulting in a decreased consciousness, such as cerebrovascular accidents, intracranial hemorrhage or septic encephalopathy. Other possible risk factors are tracheostomy handling, emergency intubation, cranioencephalic trauma, intravenous drug abuse, alcohol or sedative use and dental and medical procedures [4]. Furthermore, the occurrence of FBA is dependent on the region where

people live. In Islamic countries, aspiration of headscarf pins is quite common. How often FBA in adults occurs in the Netherlands is unknown. In this article we describe two cases of FBA in adults. The first patient presented with acute respiratory failure. The second patient presented with atypical chronic pulmonary symptoms which evolved to acute respiratory failure.

CASE SERIES

Case 1

A 42-year-old male was presented to the emergency department. His medical history revealed brain surgery for a tumor in the posterior fossa. The surgery was performed years ago, but left him with swallowing disorders for which he needed a permanent tracheostomy tube. In retrospect, it became clear that he had withdrawn himself from the medical follow-up. When presented in the emergency department he was acutely dyspneic caused by a broken and dislocated tracheostomy tube (Figure 1, left panel). He had a respiratory rate of >50/min with a peripheral oxygen saturation of 70%, a blood pressure of 115/60 mmHg and a pulse of 165 bpm.

The patient rapidly deteriorated. Therefore, an attempt was made to remove the barely visible tracheostomy tube which was trapped in the larynx under local anesthesia. During this attempt the tube dislocated to more distal airways, which caused an improvement in the vital signs. After dislocation, the patient was alert without experiencing any dyspnea. The pulmonologist performed a flexible bronchoscopy and retrieved the tube from the left main bronchus (Figure 1; right panel). After this procedure the patient remained in good condition.

Case 2

An 84-year-old male was admitted in our cardiac care unit with complaints of dyspnea and cough since several weeks. He had a history of chronic obstructive pulmonary disease (COPD), kidney and cardiac failure. At admission to the CCU he was tachypneic, his peripheral oxygen saturation with additional oxygen (>12 L/min) was 88% and bilateral crackles were heard. He had a high blood pressure of 229/103 mmHg, a rapid pulse of 102 bpm and a temperature of 38.0°C. Laboratory investigation showed high inflammatory values (CRP 120 mg/L), renal insufficiency (creatinine 210 µmol/L) and anemia (hemoglobin 7.1 mmol/L). The chest X-ray showed signs of congestive heart failure. The patient was treated with diuretics and amoxicillin/clavulanic acid. Although the patient's condition seemed to improve, five days after admission he suddenly developed respiratory failure with hemoptysis. The patient was transferred to the intensive care unit for mechanical ventilation. After hemodynamic and respiratory stabilization, a chest CT scan was performed which showed a foreign body in the right main bronchus (Figure 2). On bronchoscopy a medication blister pack surrounded by fibrin was identified. It was carefully removed and the patient's condition rapidly improved. When asked later, the patient remembered choking when taking his medication several months ago. His cough had developed afterwards.

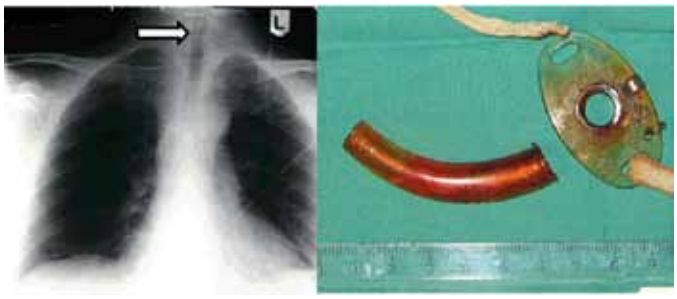

Figure 1: Left panel: Chest X-ray on admission of patient the broken tracheostomy tube. Fractured tracheostomy tube is indicated by the arrow. Right panel: broken tracheostomy tube after removal by flexible bronchoscopy.

DISCUSSION

The above mentioned cases show that FBA in the adult patient can present in very different ways and the diagnosis can be challenging.

Foreign Bodies

The types of foreign bodies (FB) that have been aspirated vary greatly among different published series. Most commonly described are bones, nutshells, metallic dentures, organic components and food particles. However, there are no limits to what can be retrieved from the airway; pen caps, needles, teeth, headscarf pins and even glass have been described [2–4]. Aspiration of medication tablets has been described in case reports, and represented up to 16% of the total of aspirated foreign

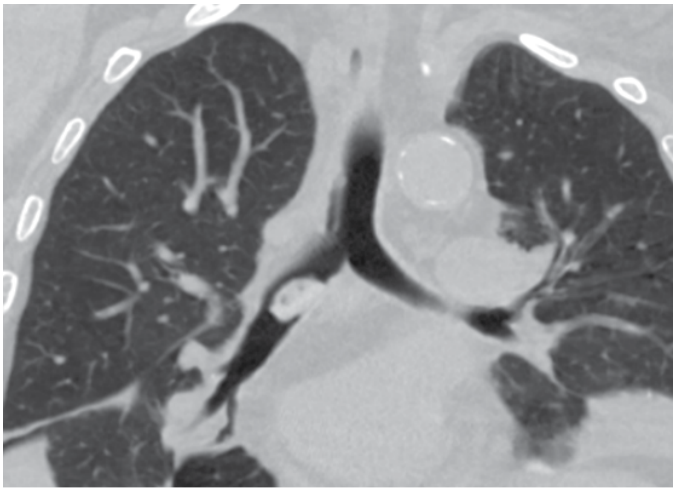

Figure 2: Computed tomography scan showing blister pack in the right main bronchus.

bodies [5]. Aspiration of a medication blister has not been described before. In a review in 1994, only 12 reports of synthetic tube fracture were found. A tracheostomy tube can fracture after prolonged use, probably due to a combination of prolonged wear, poor tracheostomy care and the formation of granulation tissue. When tracheostomy tubes fracture this usually occurs at the junction of the tube and neck plate. Patient A had not appeared at follow-up visits and his tracheostomy tube had not been changed for some time.

Symptoms

When patients are choking, FBA is often suspected. However, acute FBA is associated with choking in only 7–23% of patients [4, 6]. Symptoms are usually nonspecific and are dependent on the nature of the FB, the diameter, location and eventually the development of complications. Symptoms that are commonly seen are cough (22–94%), dyspnea (6–35%), chest pain (2–22%), hemoptysis (11–24%), fever (8–31%) and wheezing (2–28%) rare are recurrent or chronic pneumonia, cardiac arrest or no symptom at all (2–9%) [1, 2, 4, 6, 7].

There can be a significant delay in the diagnosis as symptoms can be absent or atypical. The time from aspiration to clinical presentation is determined by the severity of the symptoms. In most studies, a minority of patients (19–53%) present within one week of aspiration [4, 6]. In 58–70% of the patients the delay in diagnosis was more than 1 month [1, 6]. Much longer delays (1–40 years) after aspiration have been described [4–6]. Twenty-five percent of the patients did not remember FBA, and only 22% remembered it on clinical suspicion [4]. In the geriatric population, only 30% could remember FBA at the first visit to the doctor [7]. Again, the physical examination is non-specific, clinical signs are absent in 39–87% of patients. Decreased breath sounds were noted in 13–47% of patients and respiratory distress was seen in only 5% [2].

Both our cases show that FBA can result in respiratory failure as a result of the aspiration itself or in a later stage due to dislocation of the FB. When respiratory failure develops and a FB is expected, rapid bronchoscopic removal is indicated.

Radiology

Not only the signs and symptoms of FBA are nonspecific, chest X-ray findings are atypical as well. The chest X-ray is completely normal in 10–32% of patients. The FB was visible on the chest X-rays in a fraction of cases (11–31%). Bones or needles are relatively easy to see, whereas radiolucent FB are only “seen” due to secondary changes. Common findings on chest X-ray are atelectasis (2–50%), air trapping, emphysema (1–17%) or pneumonia (2–37%). Other, less common findings are pneumomediastinum, lung abscess, pleural effusion, lobar collapse and bronchiectasis [1, 4, 5].

The sensitivity of computed tomography for diagnosing FBA ranges from 90–100%, its specificity from 75–100% [8]. The slice thickness of the CT scan has to be taken into account [7, 8]. Atypical findings, such as atelectasis (63%), hyperlucency (44%), thickened bronchial wall adjacent to the FB (44%), bronchiectasis (31%), pleural effusion (19%) and hilar lymphadenopathy (31%) can be found [5].

Virtual bronchoscopy, in which high resolution CT scan is used to depict the bronchi from an endoscopic viewpoint, has shown high sensitivity and specificity in the diagnosis of a foreign bodies in children. However, no studies were found for this indication in adult patients [9].

Treatment

When the patient is choking and acute FBA is suspected, the initial management should focus on maintaining the patency of the airway and stabilizing the vital signs. If necessary, securing the airway by intubation (or surgical if needed) should be performed promptly to reverse hypoxia. Ventilation strategy should be used with caution to prevent pressure associated lung injury. In acute asphyxiating FBA, rigid bronchoscopy, performed under general anesthesia, is the treatment of choice [1].

Bronchoscopy

In the case of a suspected chronic presence of a FB the history, physical examination and radiology findings are often inconclusive and a bronchoscopy should be performed. It is both diagnostic and therapeutic, as it shows 85% of the inhaled FB [1]. Removal of a FB can be performed by rigid or flexible bronchoscopy.

In the past, a rigid bronchoscopy was mostly used with a high success rate of 98%. However, for chronic aspiration flexible bronchoscopy is as effective as rigid bronchoscopy and causes fewer complications. Therefore, flexible bronchoscopy is now often used as first option. Flexible bronchoscopy can be performed under local anesthesia and has a success rate of 60–97% [1, 6]. Another advantage of flexible bronchoscopy is that it visualizes segmental airways to the third generation of branching, and rigid bronchoscopy only visualizes the trachea and proximal bronchi. Therefore, when the FB is impacted in distal airways, flexible bronchoscopy is the treatment of choice. Flexible bronchoscopy is also indicated in patients with cervicofacial trauma. Computed tomography scan can help distinguishing which technique should be used first [8].

When removal with flexible bronchoscopy is unsuccessful, a repeat procedure should be performed [4]. Usually a rigid bronchoscopy under general anesthesia [1]. Reasons for failure include entrapment of the FB in the bronchial wall, serious granulation with bronchial atresia or serious hemorrhage [6]. Complications after bronchoscopy are laryngeal edema,

subcutaneous emphysema and pneumothorax [1]. The FB is usually located in the right lung, probably because the right main bronchus is more in line with the trachea [2, 5]. However, abnormalities on chest X-ray that are not right-sided should not lead to questioning the diagnosis since up to 25% of FB are located in the left bronchus and 6% in the trachea [4, 6].

Pathology

In 88% of the patients with chronic FBA of particulate matter, a cryptogenic organizing pneumonia (COP), which is a nonspecific reaction to toxic insults, was found. On biopsy, multinucleated giant cells were seen in 67%, granulomas in 33%, peribronchial fibrosis and chronic inflammation in 7%, and acute bronchiolitis in 5% [10]. When objects are aspirated, granulations form around them, which might first appear as a malignancy.

Complications

Complications associated with FBA are found in almost 80% of patients, and are often the presenting symptoms. The complications most commonly found are obstructive pneumonia (22%), bleeding (14.5%), atelectasis (10%) and endobronchial stenotic scarring (8%). Pneumonia can be complicated by empyema. The incidence of complications significantly increases from 32% when the FB is removed within three days after aspiration to 63–90% when it is removed later [1, 6]. After a difficult bronchoscopy, development of mediastinitis should be anticipated.

CONCLUSION

The presented cases show the different clinical presentations in patients presenting with foreign body aspiration (FBA), varying from chronic nonspecific to acute life-threatening clinical conditions. Importantly, chronic nonspecific presentations may evolve into acute life-threatening events. Maintaining the airway patency is essential in the acute setting. The diagnosis of FBA can be difficult, since history, physical examination and chest X-ray are often atypical. When suspicion is high, a bronchoscopy should be performed. Chest computed tomography scan can be helpful in distinguishing between flexible and rigid bronchoscopy. When patients present with nonspecific pulmonary findings, FBA should be included in the differential diagnosis.

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Author Contributions

Lycke R. Woittiez – 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
Elsbeth J. Wesselink – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published
Marcel A. de Leeuw – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published
Cornelis Slagt – 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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CASE SERIES

PEER REVIEWED | OPEN ACCESS

Wernicke's syndrome associated with hyperemesis gravidarum

Mohamed Amine Bouslama, Afraa Brahim, Ahmed Nedhir Sfeyhi, Khalil Tarmiz, Khaled Ben Jazia

ABSTRACT

Introduction: Hyperemesis gravidarum during pregnancy can be complicated by Wernicke's encephalopathy (WE). It is a neuropsychiatric syndrome due to thiamine deficiency generated by increased requirement and loss. It is a fatal but preventable complication. The management and prevention of this disorder is still unclear for lack of guidelines. Case Series: These are two cases of Wernicke's encephalopathy complicating hyperemesis gravidarum occurred in two women during the first trimester of pregnancy. Diagnosis was confirmed by MRI scan, and the treatment consisted of intravenous thiamine and parenteral nutrition with a good maternal and fetal outcome. Conclusion: Wernicke's encephalopathy must be suspected in women showing its characteristic clinical signs to avoid worse outcome, such maternofetal death or permanent neuromuscular sequelae. Medical management by thiamine is simple and efficient.

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INTRODUCTION

Hyperemesis gravidarum is uncontrollable vomiting during pregnancy that can lead to serious complications like liver damage, esophageal rupture or Wernicke's encephalopathy (WE), especially if it remains untreated.

Wernicke's encephalopathy is an acute neurological disorder related to poor thiamine absorption. It is a medical metabolic emergency which can lead to death if not managed aggressively. It was described by Carl Wernicke in 1881, in patients presenting with the triad of ocular signs, ataxia, and confusion [1].

There are currently no suitable, evidence-based guidelines for managing or preventing this disorder.

We report two clinical cases of WE in pregnancy presenting in the department of intensive care of Farhat Hached Teaching Hospital.

CASE SERIES

Case 1

A 28-year-old primigravida with no notable medical history was hospitalized in ICU at 14 weeks gestation for uncontrollable vomiting occurs six times per day during eight weeks before hospitalization. Vomiting was clear and not associated with abdominal pain.

Despite initial therapy with antiemetics and intravenous glucose, the evolution was by the persistence of vomiting and the appearance of neurological disorders involving rotatory vertigo and horizontal nystagmus. Laboratory investigations had shown severe hypokalemia 2.1 mmol/L and mild liver cytolysis: aminotransferase 185 U/L. Cerebral MRI scan had been demanded because of neurological signs. It showed periventricular and periaqueductal hyper intensities in T2 sequence affirming WE syndrome. Therapy consisted on metoclopramide 10 mg intravenously 8 hours, rehydration with 2000 ml saline solution with added potassium 20 mmol given over four hours, and thiamine supplementation 500 mg/day. Neurological signs disappeared after 48 hours of hospitalization. Total recovery was noted after 12 days. The patient delivered eutrophic baby at 40th gestation week with good outcome.

Case 2

A 24-year-old pregnant woman at 19th gestation week was admitted in the ICU with the history of excessive vomiting for several weeks followed by progressive weakness of lower limbs. Vomiting was blood-streaked with epigastric pain. At the admission, the physical examination found a drowsiness and bilateral nystagmus.

Serum biochemistry showed altered hepatic indices: aminotransferase 67 U/L, acute renal failure: creatinine clearance: 43 ml/min, and lactic acidosis: 3.6 mmol/l.

A metabolic encephalopathy was suspected. The cerebral MRI scan revealed a neuroradiologic picture compatible with WE: images of signal-intensity alterations with different intensity patterns are seen in the thalami, signal-intensity alterations in the mammillary bodies (Figures 1 and 2)

The patient was treated with 500 mg of thiamine per day associated with total parenteral nutrition. The fetal well-being was good, checked daily.

Vomiting and drowsiness had disappeared after 48 hours. Nystagmus had regressed at 13 hospital days, so thiamine supplementation was stopped. Maternofetal outcome was good.

DISCUSSION

Hyperemesis gravidarum is pathology of the first trimester of pregnancy which impact is about 0.6% [2]. It can be complicated by WE in case of thiamine deficiency.

In fact, thiamine plays a vital role in the metabolism of carbohydrates. Thiamine is a cofactor for several essential enzymes in the Krebs cycle and the pentose phosphate pathway [3].

In the setting of thiamine deficiency, thiamine-dependent cellular systems begin to fail, resulting eventually in cell death that feeds the localized vasogenic response. Thiamine-dependent enzymes play an essential role in cerebral energy utilization that's why thiamine deficiency can cause brain tissue injury most notably in regions with higher metabolic demands.

Diagnosis is based on the classic clinical triad made by encephalopathy, ataxic gait, and some variant of oculomotor dysfunction (nystagmus in 93% of cases). But symptoms may be vague and non-specific, i.e., headaches,

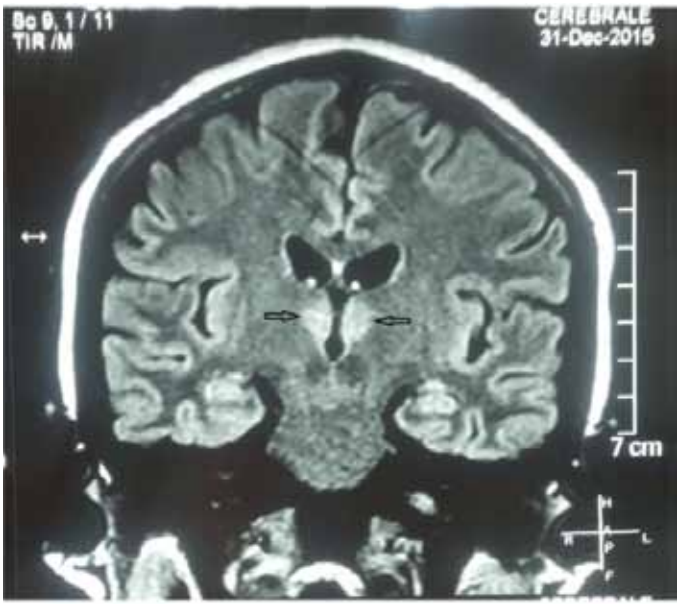

Figure 1: FLAIR coronal images signal-intensity alterations with different intensity patterns are seen in the thalami, signal-intensity alterations in the mammillary bodies.

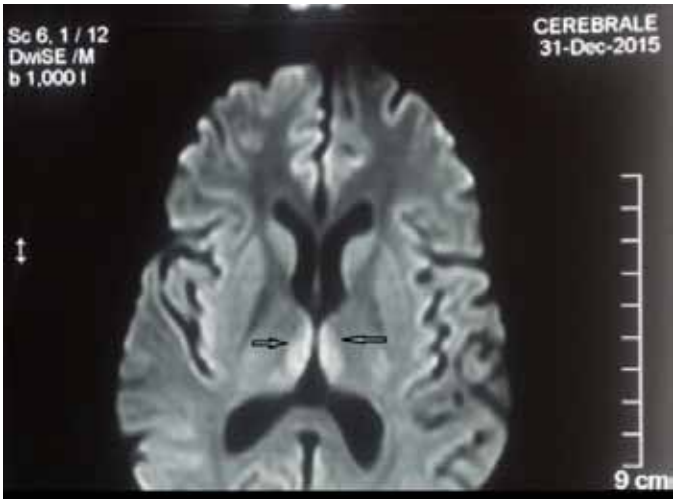

Figure 2: T2-weighted images showing the high signal intensities in the bilateral paramedian thalami with mammillary bodies.

fatigue and irritability [4].

Magnetic resonance imaging scan is the most valuable method to confirm the diagnosis with a 93% of specificity.

The typical signs of WE are increased periventricular and periaqueductal signal Intensity (FLAIR and T2) and mammillary bodies [5].

Wernicke's syndrome is fatal but reversible medical emergency. Neurologic deficits could persist despite treatment. Mortality range is about 20% [6].

If WE is suspected, parenteral thiamine should be immediately given at the dose of 500 mg/day during the two first days then 250 mg/day until the resumption of oral feeding [7].

Some authors recommend the thiamine intake until the delivery [8]. Wernicke's syndrome threaten the fetal prognosis. It is associated with an increased risk of low birth weight, neurodevelopmental disorders, intrauterine growth restriction, preterm delivery, and fetal and neonatal death. Ischemic stroke can be seen in fetuses of women affected by US with no specific explanations [9].

Early diagnosis and vitamin therapy within the first 24 hours prevent fetal worse outcome [10]. Anti-emetics are also useful as adjoining therapy to reduce the intensity of vomiting.

A chronic WE can develop in Korsakoff syndrome which consists in impairment in memory with loss of working memory. Moreover, in severe forms of WE, muscular sequelae are possible [8].

For our two patients, no incidents had been recorded, and the remission was complete.

CONCLUSION

Wernicke's syndrome is a rare but serious neurologic complication of hyperemesis gravidarum that must be suspected in pregnant women in the first trimester showing the clinical symptoms. Magnetic resonance imaging findings confirm the diagnosis. Maternofetal outcome depends on the rapidity of the treatment by thiamine.

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Author Contributions

Mohamed Amine Bouslama – 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

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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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CASE SERIES

PEER REVIEWED | OPEN ACCESS

Not safe for consumption: Synthetic cannabinoids causing fatal acute rhabdomyolysis in two young men

Anthea B. Mahesan Paul, Lary Simms, Abraham Ebenezer Paul, Christopher Schmidseder, Andrew A. Mahesan, Jojo Yorke

ABSTRACT

Introduction: Synthetic cannabinoids are a class of recreational drugs that are included in the growing epidemic of synthetic recreational drugs sweeping the United States. Synthetic cannabinoids look, feel and act like Marijuana with dangerous and potentially fatal adverse effects. Synthetic cannabinoids, in addition are not detected by routine urine toxicology screening, and thus the magnitude of its prevalence is unknown. Case Series: In this case series, we report two cases of fatal acute rhabdomyolysis associated with synthetic cannabinoid use in previously healthy young men. The strong correlation found by detailed history, clinical evaluation, and laboratory tests including the negative universal drug screen strongly suggests an association between synthetic cannabinoid use and fatal acute rhabdomyolysis in both cases. Conclusion: Synthetic recreational drug use, including synthetic cannabinoids should be included in the differential diagnosis for acute rhabdomyolysis in young people with negative universal drug screens and initiatives should be taken to educate physicians and the general public of the serious consequences of synthetic cannabinoid use.

Keywords: Synthetic cannabinoids, Spice, Rhabdomyolysis

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INTRODUCTION

Synthetic cannabinoids (SC) are a blend of herbal and chemical compounds used as a recreational drug that imitates the effects of marijuana. Synthetic cannabinoids have been sold in enticing packaging mislabeled as potpourri or herbal mixtures. The packages often labeled with, “not safe for consumption” have a variety of names including “Spice”, ”K2”, “Nice Guy”, “NBT”, “Black Mamba” and “Crazy Monkey” [1]. Due to the relatively new advent of SCs, there is limited data available on the short-term and long-term effects of synthetic cannabinoid use. To our knowledge, this is the first case series of two fatalities that can be associated with acute rhabdomyolysis and synthetic cannabinoid use in the United States.

There have not been randomized controlled trials for SC use in the US and thus the documented effects of SCs are the result of subjective patient reporting. The commonly reported adverse effects of SC use by the CDC are drowsiness or lethargy (26.3%), emesis (16.4%), confusion (4.2%), tachycardia (29.0%), and agitation (35.3%) [2]. Spaderna et al. have summarized the less common adverse effects reported in the literature affecting a large range of organ systems including neurological, cardiovascular, psychological, and gastrointestinal adverse effects [3].

Physicians and healthcare personnel should be aware of the possibility of potentially fatal rhabdomyolysis in patients that use synthetic cannabinoids.

CASE SERIES

Case 1

A 30-year-old Caucasian male presented to the emergency room unconscious and unresponsive after being on an 8-day synthetic cannabinoid binge (NBT brand). According to his fiancé, he experienced nausea and vomiting for approximately six days before hospital admission accompanied by abdominal pain and difficulty urinating. His urine was described as dark brown. In addition he complained of muscle cramps, and soreness. The patient had a history of spice use for the past two years, and was a ½ pack a day smoker. He had no history of other drug use, or any relevant medical or surgical history. On admission, He exhibited hypertension, oliguria, muscle tenderness accompanied with creatinine phosphokinase levels of 47,000 IU, hyperkalemia, BUN: 51 mg/dL, abnormal liver function tests, creatinine: 5.15 mg/dL, hypocalcaemia and hypoalbuminemia consistent with severe rhabdomyolysis. Universal drug screen was negative. Despite aggressive intravenous hydration, forced diuresis, and hemodialysis, the oliguria persisted and the patient died 5 days after admission with renal failure and ventilator associated bilateral pneumonia. (Figure 1)

Case 2

A 23-year-old African-American male with history of substance abuse, and no other significant past medical history, presented to the emergency room complaining of nausea, vomiting, chest pain, generalized weakness, generalized body aches and muscle cramps that began approximately four days before admission. He had a history of exclusive spice use for the past five days. He also complained of sharp retrosternal chest pain, muscle pain being unable to walk for three days prior, accompanied with low urine output. The patient was a former occasional smoker and occasional alcoholic, last use of both cigarettes and alcohol was 1 week prior. On admission, the patient was hypertensive with a blood pressure of 170/82 mmHg. Laboratory findings showed abnormal findings consistent with dehydration and rhabdomyolysis with creatine kinase level of 37,200 IU, CK-MB: 113.7 ng/mL, abnormal liver function tests, hyponatremia with sodium of 105 mmol/L, creatinine: 4.26 mg/dL, hypocalcaemia, hyperkalemia, BUN: 120 mg/dL, and elevated Troponins. Universal drug screen was negative. Despite aggressive intravenous hydration, forced diuresis, hemodialysis, and supplementation the patient rapidly deteriorated and died three days after admission. (Figure 2) (Table 1).

Table 1: Laboratory Values for Patient 1 and Patient 2 on Admission

Test	Patient 1		Normal Values	Patient 2	
CPK	47, 000	H	(52-336) U/L	37, 200	H
Potassium	2.5	H	(3.5-5.5) mmol/L	4.9	*
BUN	51	H	(7-18) mg/dL	120	H
SGOT/AST	1044	H	(15-37)	5053	H
SGPT/ALT	241	H	(12-78)	2228	H
Creatinine	5.15	H	(0.52-1.23) mg/dL	5.69	H
Calcium	6	L	(8.5-10.1) mg/dL	5.2	L
Albumin	3.2	L	(3.4-5.0) g/dL	3	L
Sodium	121	L	(136-145) mmol/L	105	L
Estimated GFR	18	L	mL/min	10	L
Drug screen	Negative	*		Negative	*

Abbreviations: H= High, L= Low, *= Normal

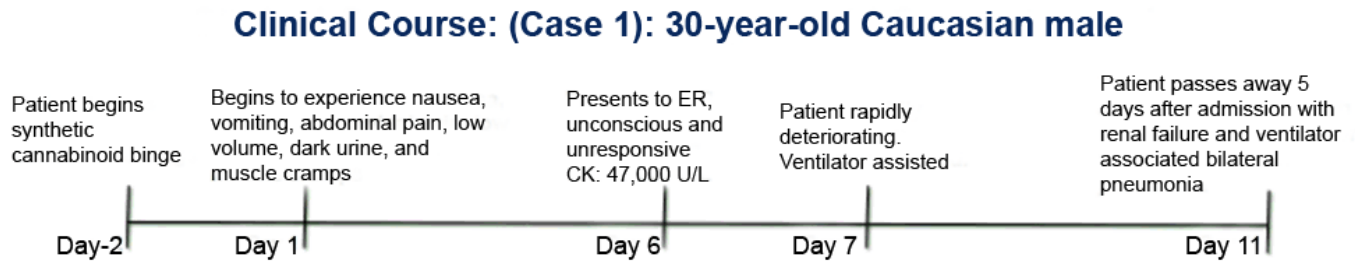

Figure 1: Clinical Course of Patient 1.

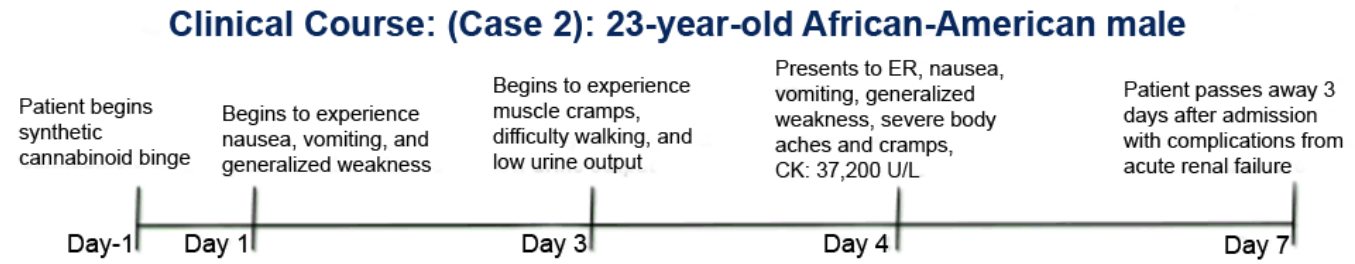

Figure 2: Clinical Course of Patient 2.

DISCUSSION

Unfortunately, young people are being victimized by the recreational synthetic drug epidemic sweeping the United States. Though the exact number of regular SC users is undetermined, according to a study at American high schools, 11% of 12th graders and 4% of 8th graders reported using SC products in the year 2013 [4]. In addition, the CDC reported a stark increase of calls regarding effects from SC use by 229% in 2015 from the same January–May period in 2014 [2]. Although it is apparent the prevalence of SC use is on the rise, Buser et al. found that young adults and adolescents appear to be unaware of the health risks associated with synthetic cannabinoids [5].

Since its debut in the United States in 2009, legislature has struggled to keep up with the growing epidemic of synthetic recreational drug use. In the summer of 2012, President Obama signed legislation to include 21 different synthetic cannabinoid compounds and five overall cannabinoid structural classes into Schedule I of the Controlled Substances Act (CSA) [6]. The legislature worldwide struggles to keep up with the synthetic manufacturing of these recreational drugs with the total number of identified SC compounds reaching over 84 as of May 2013 [7].

The major users of SCs have been suggested to be populations wishing to avoid drug detection such as parolees, students and military staff due to the inability of the drug to be detected through the standard urine drug screening [8]. Another hypothesis has been proposed that SC's may be used as an adulterant to unsuspecting consumers under the guise of marijuana, due to the relatively low price of about $6-10 USD per gram [9]. Regardless of the intentional or unintentional consumption of this drug, the dilemma remains that the general public remains ignorant of the dangerous and potentially fatal adverse effects of SC use.

A limitation of this case series is the inability to chemically confirm the presence of SCs at the time of admission. This association between SC use and acute fatal rhabdomyolysis was made after careful exclusion of other potential causes aided by a detailed history attained from the patients and patients' families, clinical evaluation, and laboratory tests including the negative universal drug screen. Through the universal drug screen it is apparent that our patients were not abusing other drugs at the time of admission and this supports our hypothesis that the history of SC use was the key contributing factor to the development of acute rhabdomyolysis. A number of recreational drugs commonly abused including benzodiazepines, ecstasy, heroin, ketamine hydrochloride, marijuana, lysergic acid diethylamide, methamphetamine, narcotics, phencyclidine, ethanol and cocaine have been associated with drug-induced acute rhabdomyolysis [10]. It is suggested that acute rhabdomyolysis could occur from muscle destruction by compression and ischemia due to muscular hyperactivity, extreme exercise, and catatonic states brought on by acute recreational drug intoxication [11].

CONCLUSION

With the legalization of medical marijuana in 23 states in the US it is easy to see how synthetic cannabinoids that feel, smell and look the same may be misleading to some individuals. Initiatives should be under taken to educate

physicians and the general public of the consequences and effects of SC use and synthetic recreational drugs as a whole.

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Guarantor
The corresponding author is the guarantor of submission.

Conflict of Interest
Authors declare no conflict of interest.

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CASE REPORT PEER REVIEWED | OPEN ACCESS

Sigmoid volvulus during second trimester of pregnancy in a primigravida: Report of a rare case with review of imaging of sigmoid volvulus

Obaid Ashraf, Sameer Peer, Mohsin Fayaz, Mohammad Saleem Dar, Inayat Illahi, Fahad Shafi

ABSTRACT

Introduction: Intestinal obstruction is a rare cause of acute abdomen in pregnancy. The incidence of intestinal obstruction in pregnancy ranges from 1:1500 to 1:66431. The differential diagnosis for intestinal obstruction in pregnancy includes congenital or postoperative adhesions, volvulus, hernia, intussusception, malignancy and appendicitis. Sigmoid volvulus is the most common cause of intestinal obstruction accounting for 25–44% of the reported cases in literature. It is extremely important to diagnose this condition early as a delay in diagnosis can lead to maternal as well as fetal complications. Case Report: A case of 20-year-old female, primigravida, in 22nd week of pregnancy who presented to our hospital with a three-day history of colicky abdominal pain, few episodes of vomiting and obstipation. Ultrasound showed a single live intrauterine fetus and a dilated gut loop extending from left hypochondrium to the left iliac fossa. T2-weighted TRUFI and HASTE MR Images in axial and coronal plane showed radiological signs highly suggestive of sigmoid volvulus. The radiological findings were confirmed on laparotomy and detorsion of the sigmoid loop with decompression followed by sigmoidopexy was performed. No maternal or fetal complications occurred in the perioperative period. Conclusion: Sigmoid volvulus is a rare non-obstetric complication of pregnancy which requires an early diagnosis and prompt intervention to prevent maternal and fetal complications. Magnetic resonance imaging scan can provide an accurate diagnosis of sigmoid volvulus and its use is safe in pregnancy with respect to the risks of radiation exposure in pregnancy.

Keywords: Crossed-fused renal ectopia, Intestinal obstruction, Pregnancy, Sigmoid volvulus

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INTRODUCTION

Intestinal obstruction is a rare cause of acute abdomen in pregnancy. The incidence of intestinal obstruction in pregnancy ranges from 1:1500 to 1:66431 [1–4]. The differential diagnosis for intestinal obstruction in pregnancy includes congenital or postoperative adhesions, volvulus, hernia, intussusception, malignancy and appendicitis [1, 2, 4, 5]. Sigmoid volvulus is the most common cause of intestinal obstruction accounting for 25–44% of the reported cases in literature [1–4, 5–7]. It is extremely important to diagnose this condition early as a delay in diagnosis can lead to maternal as well as fetal complications. Maternal complications include bowel infarction and necrosis with hypovolemia, electrolyte disturbances, septic shock, multi-organ dysfunction and death [1, 3, 5, 6]. Fetal complications include preterm delivery, intrauterine death and neonatal sepsis [5]. Maternal mortality for sigmoid volvulus depends upon the status of the bowel and it ranges from 5% in cases of viable bowel to 50% in those cases where perforation has occurred. Fetal mortality in sigmoid volvulus is estimated to be approximately 30% [1, 6]. The diagnosis of sigmoid volvulus in pregnancy is challenging as the symptoms and signs of intestinal obstruction could be masked by pregnancy itself [1, 3, 4, 6, 8]. Another problem faced by the clinicians is regarding the choice of the diagnostic imaging modality. Radiation exposure to the developing fetus increases the risk of chromosomal abnormalities, neurological malformations and hematological malignancies [5]. Ultrasound is a diagnostic modality which is free of radiation hazard. However, gaseous distension of bowel loops limits its utility in establishing an accurate preoperative diagnosis. It can be useful in the assessment of fetal status, ascites and any other alternative diagnosis of acute abdomen. Magnetic resonance imaging scan could be used as a problem solving tool in the diagnosis of the cause of intestinal obstruction. It is not only free of radiation hazard but can also provide an accurate preoperative diagnosis of the etiology [3, 4]. However, availability of MRI scan in many centers across developing countries is a limiting factor. All these factors lead to a delay in reaching an early diagnosis and thus the risk of complications is increased.

CASE REPORT

A 20-year-old female, primigravida, in 22nd week of pregnancy, presented to our hospital with a three-day history of colicky abdominal pain, few episodes of vomiting and obstipation. On examination, she was conscious, oriented in time, place and person. She looked pale and was in distress. Her pulse rate was 120/min and blood pressure was 110/70 mmHg. Her respiratory rate was 27/min and SpO2 was 96%. Her abdomen was distended and tense but non-tender. Fetal heart sounds were heard with stethoscope. She looked dehydrated. Digital rectal examination revealed an empty rectum. There was no bleeding per vaginum and the os was closed.

Initial resuscitation with IV fluids was done. The patient was catheterized and a nasogastric tube was put in. A flatus tube was inserted into the rectum but no gas came out. Her laboratory investigations were unremarkable except for a total leucocyte count of 17000/μl (N85L15). Cardiotocography was unremarkable and there was no evidence of fetal distress.

Ultrasound of the abdomen in emergency lab showed a single live intrauterine fetus in cephalic presentation at the time of scan with a fetal heart rate of 145/min. A dilated gut loop was seen in the left hypochondrium which was extending into the left iliac fossa. Gaseous distension of the bowel loops precluded the examination of rest of the abdomen.

At our institution, X-ray exposure in pregnancy is avoided unless it is absolutely necessary for the management of the patient and since MRI scan is available in our institution, so it was decided to get an MRI scan done. Scan was performed on Siemens Magnetom Avanto 1.5 Tesla scanner. T2-weighted TRUFI and HASTE sequences were obtained in coronal and axial planes. Magnetic resonance imaging scan revealed a massively distended loop of sigmoid colon in an "inverted U-shaped" configuration extending from the left iliac fossa to the left hemidiaphragm with apposition of the walls of two adjacent loops giving a "coffee bean" appearance (Figure 1). The sigmoid colon was seen to extend above the transverse colon ("northern-exposure sign") which was displaced to the right side. A transition point in the sigmoid loop was noted in the left iliac fossa with narrowing and twisting of the loop. "Split-wall sign" i.e., separation of the intestinal walls by mesenteric fat, was also noted at the transition point (Figure 2). An incidental finding of crossed-fused renal ectopia was also detected on MRI scan (Figure 3). Mild ascites was also noted. Hence a diagnosis of sigmoid volvulus was made and the patient was taken by the surgical team for laparotomy. Magnetic resonance imaging findings were

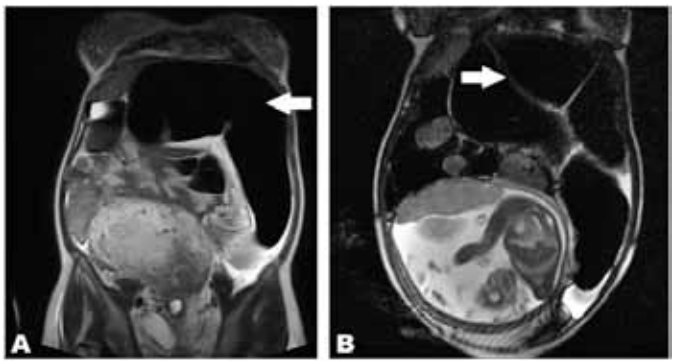

Figure 1: (A) T2-weighted coronal HASTE image showing a massively dilated sigmoid loop (arrow) in an "inverted-U" configuration extending up to the left hemidiaphragm (northern-exposure sign), (B) T2-weighted coronal TRUFI image apposition of the walls of the two adjacent dilated gut loops giving a "coffee-bean" appearance (arrow). Fetus can also be seen in the image. Mild free fluid is also seen in the pelvis.

confirmed intraoperatively. Massively dilated sigmoid colon with four anticlockwise mesenteroaxial turns was seen (Figure 4). Mild ascites was also noted. Bowel was viable and no signs of ischemia or infarction were seen. Gravid uterus, ovaries and fallopian tubes were normal. The surgical team performed detorsion of the sigmoid loop with decompression followed by sigmoidopexy. The postoperative period was uneventful. Postoperative ultrasound showed normal live fetus. No maternal or fetal complication occurred.

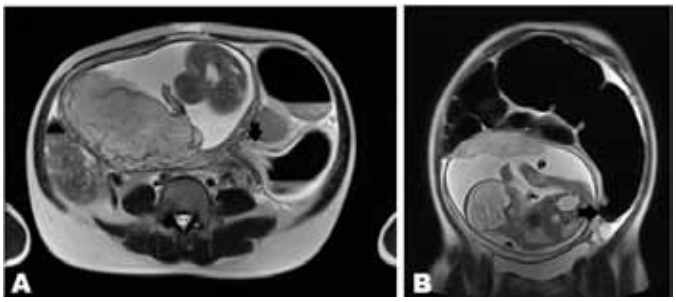

Figure 2: (A) T2-weighted axial HASTE image showing narrowing of the sigmoid colon at the transition point (arrow). The intestinal walls are seen to be separated by the mesenteric fat (split-wall sign), (B) T2-weighted coronal HASTE image showing the twisting of the sigmoid loop at the transition point (arrow).

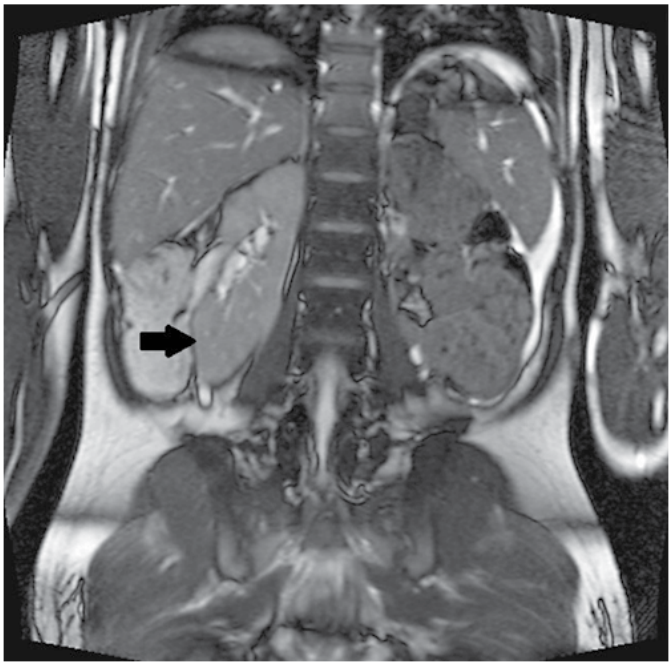

Figure 3: T2-weighted coronal TRUFI image showing maternal crossed-fused renal ectopia on the right side (arrow).

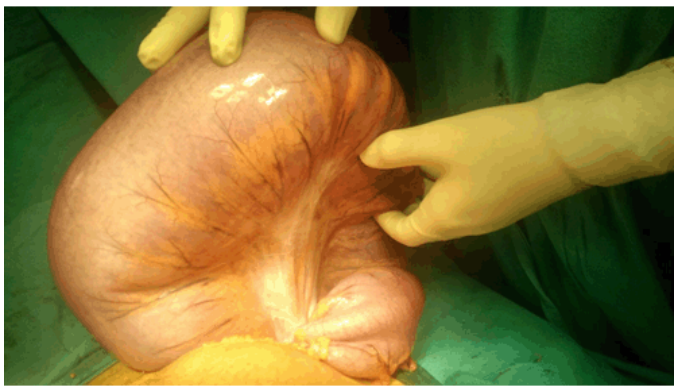

Figure 4: Intraoperative photograph showing massively dilated sigmoid colon delivered through the laparotomy incision. No evidence of gut ischemia, infarction or perforation seen.

DISCUSSION

The first case of intestinal obstruction during pregnancy was reported by Houston in 1830 [1]. The first case of sigmoid volvulus in pregnancy was documented by Braun in 1885 [4, 6]. The rarity of sigmoid volvulus in pregnancy can be judged from the fact that only 105 cases have been reported in literature till 2015 since the first case report in 1885 [6]. The sigmoid is the most common site of colonic volvulus and accounts for 60–75% of all cases of colonic volvulus [9]. It occurs when sigmoid colon undergoes a twist along its mesenteric axis leading to a closed loop obstruction which predisposes to ischemia, perforation and death [7]. Sigmoid volvulus usually occurs in chronically ill, debilitated and institutionalized patients. In India and Africa, a high-fiber diet predisposes to sigmoid volvulus while in South America, Chagas disease is a predisposing factor [4, 9]. A long redundant sigmoid colon with a narrow mesentery, also known as dolichosigmoid, predisposes to volvulus formation [8]. Pregnancy itself predisposes to sigmoid volvulus as enlarging uterus displaces an abnormally mobile sigmoid loop out of the pelvis causing it to twist around the sigmoid mesocolon leading to vascular compromise and obstruction. This could also be a possible explanation of higher frequency of sigmoid volvulus in third trimester of pregnancy [1, 2, 4–6, 8]. Among the reported cases of sigmoid volvulus in pregnancy, most of the patients are in the age group of 15–35 years with 75% of the cases being multiparous and 66% cases occurring in third trimester [3].

The presenting features of sigmoid volvulus in pregnancy include abdominal pain, distension and obstipation, the so-called sigmoid volvulus triad. Additionally, nausea and vomiting could also be the presenting complaints [1–9]. On examination, abdominal tenderness, hyperkinetic or absent/hypokinetic bowel sounds and empty rectum are the usual signs [3]. The mean duration of symptoms has been reported to range from 1 hour to 6 days with a mean duration of 40.6 hours [1]. Among laboratory investigations, leukocytosis could be considered as a consistent sign but in early phase of the disease, counts could be normal or slightly elevated and furthermore, in pregnancy counts could be normally elevated [1].

On a plain radiograph of abdomen, a large air-filled loop of sigmoid colon arising from the pelvis and extending cranially beyond the level of the transverse colon (the “northern exposure” sign) may be considered

diagnostic of sigmoid volvulus. Other features include the "coffee bean" sign, which refers to the coffee bean–like shape assumed by the dilated sigmoid colon [9].

In cases where diagnosis is uncertain, a contrast enema or CT scan may be done. Contrast enema shows the "beak sign" at the level of the twist beyond which no contrast passes. Contrast enema may help to reduce the volvulus. On CT scan, swirling of the mesentery may be seen at the site of the volvulus [9]. Levsky et al. described two imaging signs of volvulus: X-marks-the spot-sign in which crossing sigmoid transition is seen and the split wall sign in which the intestinal walls are separated by adjacent mesenteric fat planes due to partial twisting of the sigmoid loop [7, 8].

The major concern regarding the use of radiography and CT in pregnancy is that of radiation exposure to the developing fetus. The recommended cumulative radiation dose to the fetus during pregnancy is 5–10 rads. The radiation dose to the fetus for a plain X-ray on average is 0.1–0.3 rads. In general no single examination, including CT, exceeds the recommended safe dose range [1, 5, 6]. Considering the harmful effects of radiation on organogenesis and the risk of hematological malignancies, some authors are of the opinion that the use of X-rays should be avoided during pregnancy [1]. However, when the benefit to the mother clearly outweighs the risk to the fetus, radiation may be used to establish an early diagnosis [5, 6].

Ultrasonography does not use ionizing radiations and hence is safe in pregnancy [1]. It provides information regarding the fetus and can help to exclude an alternative diagnosis for acute abdomen. Transition point may also be seen on ultrasound [8].

Magnetic resonance imaging can be used as a problem solving tool in the diagnosis of sigmoid volvulus. There is no risk of radiation exposure to the fetus. High contrast resolution in MRI scan can help in establishing the diagnosis accurately [8]. All the signs suggestive of sigmoid volvulus which are seen on radiograph or CT scan can be equally demonstrated on MRI as well [8]. However, availability of MRI scan in developing countries may be a limiting factor for its use in establishing a preoperative diagnosis in such cases.

A multidisciplinary approach is used in the management of sigmoid volvulus in pregnancy which involves obstetricians, neonatologists and general surgeons [3, 6]. Initial resuscitation includes administration of IV fluids, nasogastric decompression and correction of electrolyte imbalance if any [6]. Tocolytics may be used for uterine stability and steroids may be used in cases where fetal maturity is a concern [3, 6]. The surgical approach depends upon the status of the bowel. In cases with good colonic vascularization, no peritoneal signs and no signs of bowel ischemia, decompression of volvulus is an acceptable treatment with the aim of restoration of the colonic blood supply. Detorsion of the volvulus with a soft rectal tube placed endoscopically or during laparotomy could be attempted. If bowel gangrene or perforation is suspected or is present, due to prolonged compromise of vascular supply, urgent resection is required [6, 8]. Aftab et al. reported a case of sigmoid volvulus in pregnancy which was managed by endoscopic reduction [1].

This case report highlights the need for establishing an early diagnosis and prompt intervention in cases of sigmoid volvulus presenting during pregnancy to prevent catastrophic complications. This case also emphasizes the role of MRI scan as a safe and accurate diagnostic tool for management of these cases.

CONCLUSION

Sigmoid volvulus is a rare non-obstetric complication of pregnancy which requires an early diagnosis and prompt intervention to prevent maternal and fetal complications. Magnetic resonance imaging can provide an accurate diagnosis of sigmoid volvulus and its use is safe in pregnancy with respect to the risks of radiation exposure in pregnancy.

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Author Contributions

Obaid Ashraf – 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
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Guarantor

The corresponding author is the guarantor of submission.

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CASE REPORT PEER REVIEWED | OPEN ACCESS

Giant gastric lithobezoar in an adult: A case report

Alana Costa Borges, Marco Augusto Sobreira Rocha Filho, Vitor de Vasconcelos Muniz

ABSTRACT

Introduction: A bezoar is a conglomeration of partially digested or undigestible foreign material in the gastrointestinal tract, classified according to its composition. Inorganic matter bezoars are very rare, especially lithobezoars, i.e., stone-composed, in the upper digestive system. Case Report: A case of 49-year-old male, schizophrenic patient, with a prior history of pica presenting with non-specific symptoms and a 10-kg weight loss. Upon physical examination, an enormous palpable left mass was noted and when mobilized, it produced a peculiar sound. Diagnostic workup comprised exclusively plain radiographs, with pathognomonic findings and subsequent indication of surgery. Exploratory laparotomy revealed a 3.5-kg gastric bezoar encompassing diverse objects, but predominantly stones. There was no evidence of their presence in other organs and no complications. The patient had uneventful recovery and is currently engaged in outpatient follow-up. Conclusion: Pica with resulting bezoar should be suspected in psychiatric adult patients with non-specific symptoms. Giant upper gastrointestinal lithobezoars are very rare and may produce a gastric "crunch sign" on physical examination, which is a clue for the correct diagnosis. Open surgery is effective and can be the treatment of choice.

Keywords: Bezoar, Crunch sign, Gastric, Lithobezoar, Mass, Pica

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INTRODUCTION

A bezoar is a conglomeration of partially digested or undigestible foreign material in the gastrointestinal tract [1]. The term derives from Arabic "bazahr" or "badzehr", which means antidote, due to the fact that until the 19th century, bezoars obtained from sacrificed animals were utilized to cure several illnesses [2]. The earliest record of human trichobezoar was credited to Baudamant in 1779 and the first preoperative diagnosis to Stelzner in 1896 [3].

Bezoars are classified according to their composition. The most common are phytobezoars (vegetable matter) and trichobezoars (hair). Other less frequent are lactobezoars (concentrated milk formulas) and inorganic

material bezoars [1, 3, 4]. Clinical manifestations depend on its location, ranging from no symptoms to acute abdominal syndromes. The stomach is the most frequently affected organ [1, 3]. We report a unique case of a giant gastric bezoar in an adult patient.

CASE REPORT

A 49-year-old male, schizophrenic patient, was presented to the emergency department. His relatives informed a progressive hyporexia over a four-month period associated with post-prandial fullness and a 10-kg weight loss. His past medical history was remarkable for metal pica (coins) two years before.

Physical examination revealed mild pallor, epigastric bulging which was dull to percussion and an enormous hard, irregular, palpable mass, extending from the epigastrium to the left iliac fossa. Upon mobilization of the mass, a peculiar sound was produced, which was a clue for the diagnosis.

Plain abdominal radiograph showed no signs of intestinal obstruction and a dilated stomach with a giant radiopaque bezoar containing different-sized grouped smaller images. This finding was another important evidence for the diagnosis (Figure 1).

The patient was taken to open surgery, with evidence of a grossly dilated stomach (Figure 2) and performance of a longitudinal gastrotomy (Figure 3), manual removal of the foreign bodies (Figure 4) and a two-layer suture. At closer examination of the gastric mucosa, two superficial ulcers were identified, at the lesser and greater curvature, most likely due to contact with the sharp objects. There was neither any evidence of foreign bodies in other organs nor any complications.

A 3.5-kg bezoar (Figure 5) comprised diverse objects: one plastic bag, two batteries, three lighters, six toothbrushes, some shoelaces, pens, coins, keys, spoons and necklaces, but predominantly stones in countless number.

The patient's postoperative evolution was uneventful, with multidisciplinary care. Additionaly, he was cared for under close vigilance, with physical and chemical restraints to avoid pica. The diet was resumed in the third postoperative day and he was transferred on the seventh day to a referral psychiatric hospital.

DISCUSSION

Lithobezoars (stones) are quite rare, with very few cases described, as are metal bezoars and plastic bezoars [1, 2, 4, 5]. Most of the published data consist of case reports of colonic lithobezoars in children and adults [4, 6]. To the best of the our knowledge, there is only another article detailing a gastric lithobezoar which presented as dyspepsia, however, with much less stones compared to the current case [7].

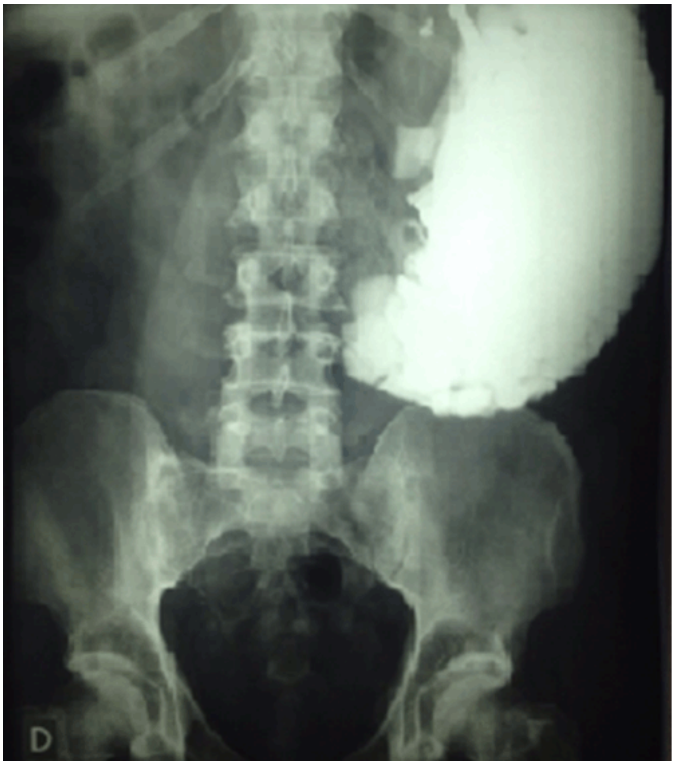

Figure 1: Abdominal radiograph with radiopaque gastric bezoars.

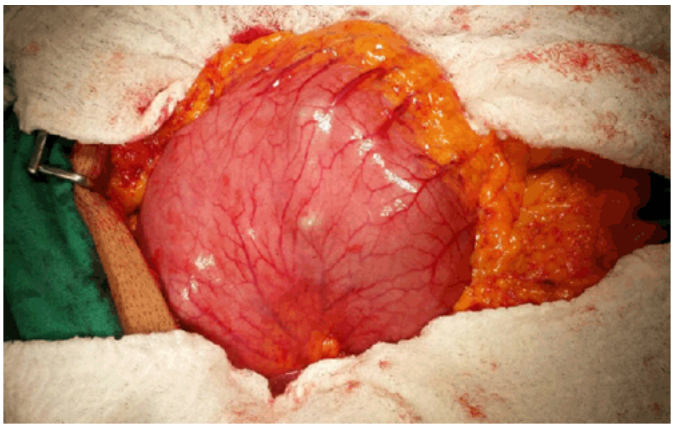

Figure 2: Grossly dilated stomach.

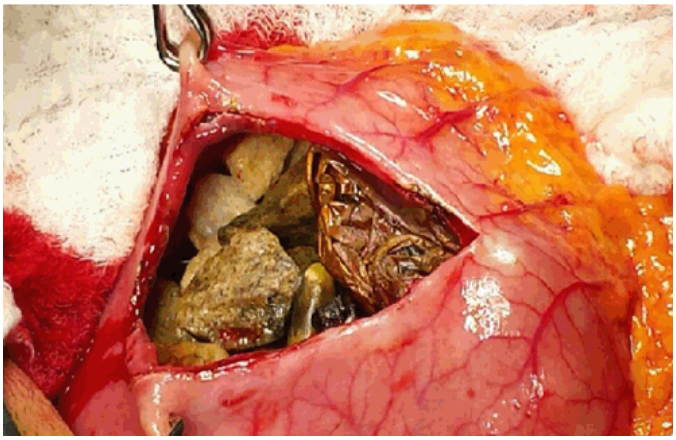

Figure 3: Longitudinal gastrotomy exposing the bezoar.

Risk factors for bezoars in general include previous gastric surgery, conditions of reduced acidity or delayed motility, poor mastication, mental retardation, psychiatric disorders, and pica, i.e., an abnormal eating behavior characterized by ingestion of nonnutritive substances and classified according to the consumed matter: trichophagia (hair), acuphagia (sharp objects), lithophagia, etc. [3, 6, 8]. In this case, the patient was schizophrenic and had previous metal pica.

Usually, there are no symptoms until the bezoar reaches a substantial size but many patients remain oligosymptomatic. Gastric bezoars ordinarily cause dyspepsia (80%), distention, nausea, vomiting, hyporexia and halitosis. Complications as ulcers and perforation can occur [3, 5].

On physical examination, sometimes, it is possible to palpate the organ filled with the stones, with a characteristic "crunch sign", which usually applies to the colon [4, 9, 10]. In the present case, given the palpation of the gastric bezoars with resulting singular sound, we strongly believe the concept also fits the stomach. Hence, we would like to propose the "gastric crunch sign".

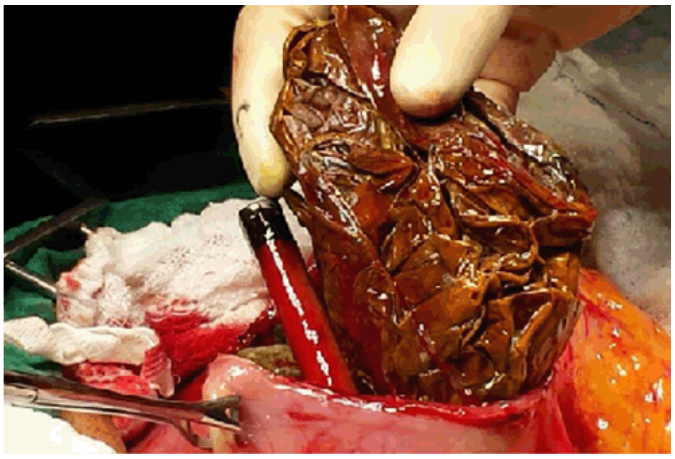

Figure 4 Manual removal of the foreign bodies.

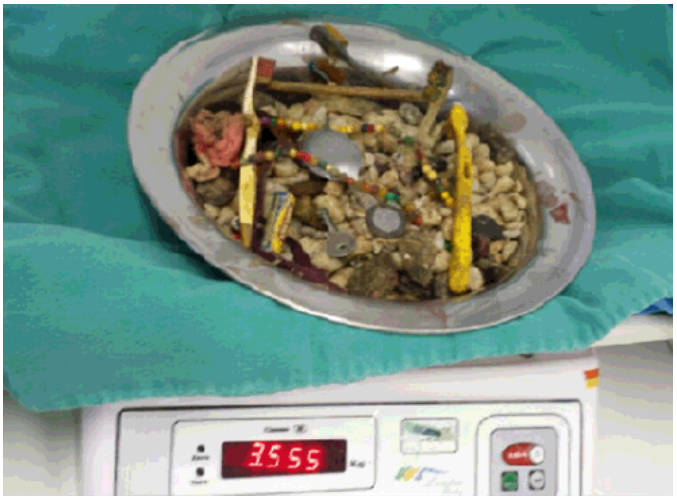

Figure 5 Conjoint weight of the bezoar.

Radiographical diagnosis is especially valuable in litho bezoars, showing typically conglomerated radiopaque images within the organ. This unique appearance, "corn on the cob" sign, is considered pathognomonic. In this particular type of bezoar there is no need for barium studies or other image methods [4, 9, 10].

Upper endoscopy can be utilized in the diagnostic approach [3]. Nevertheless, the authors did not perform it given the large amount of foreign material in the stomach, making air insufflation particularly challenging, with risk of perforation.

Gastrointestinal bezoars can be effectively treated by endoscopy or surgery. Endoscopic extraction is the choice in little or fragmentable bezoars, utilizing snares, rat-tooths, Dormia baskets and lithotripsy methods [1, 3, 5]. Nevertheless, endoscopic management carries the risk of distal migration of fragments and iatrogenic injuries due to the manifold introductions/retrievals of the endoscope [3].

Open surgery is indicated in cases of failure of conservative attempts, large dimension bezoars composed of unfragmentable matter, complications such as intestinal obstruction, perforation and hemorrhage. The preferred approach is longitudinal gastrotomy in the anterior wall with removal of the bezoars followed by gastrorrhaphy [3]. We are successfully resorted to surgery and the patient had a favorable outcome.

CONCLUSION

In conclusion, pica with resulting bezoar should be suspected in psychiatric patients presenting with nonspecific symptoms. Plain abdominal radiograph, a simple and widely available test, should be performed in those patients, confirming the diagnosis in case of radiopaque objects. Giant upper lithobezoars are very rare, can produce a gastric "crunch sign" and can be treated effectively by open surgery.

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Author Contributions

Alana Costa Borges – 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

Marco Augusto Sobreira Rocha Filho – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published

Vitor de Vasconcelos Muniz – 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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CASE REPORT PEER REVIEWED | OPEN ACCESS

Tumor lysis syndrome in metastatic colon cancer after single FOLFOX cycle

Akanksha Agrawal, Deepanshu Jain, Mark Morginstin

ABSTRACT

Introduction: Tumor lysis syndrome (TLS) is a life-threatening oncological complication, often described in patients with a large tumor burden, more commonly among hematological malignancies. Case Report: We present a case report of 55-year-old male who presented to the emergency department with worsening abdominal pain, oliguria, nausea, vomiting and diarrhea of one-day duration. One-month prior, the patient was diagnosed with metastatic colon cancer. He was started on FOLFOX regimen the day prior. Patient was found to be in acute kidney injury with hyperkalemia, hyperuricemia and hyperphosphatemia. Patient was admitted with a diagnosis of TLS (Cairo-Bishop grade II) and managed with aggressive intravenous hydration, furosemide and single dose of rasburicase. In two days, symptoms resolved with improvement in laboratory parameters and patient was discharged after four days. Conclusion: There have been so far six published cases, reporting TLS in metastatic colon adenocarcinoma. Our case is the only one demonstrating that TLS can occur in metastatic colon cancer patient after a single cycle of FOLFOX therapy even in the absence of any pretreatment. Our patient had all known risk factors for developing TLS like large tumor burden, liver metastases, elevated pretreatment lactate dehydrogenase (LDH), use of combination chemotherapy drugs and dehydration. Unlike previously reported six cases where TLS resulted in death, our patient survived. Therefore, a clinician should maintain high index of suspicion for TLS among metastatic colon cancer patients and should do prompt intervention to prevent potentially life-threatening complications like cardiac arrhythmias, acute renal failure, seizures, or death.

Keywords: Abdominal pain, FOLFOX, Metastatic colon cancer, Tumor lysis syndrome

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INTRODUCTION

Tumor lysis syndrome is a life-threatening oncologic emergency characterized by hyperuricemia, hyperkalemia, hyperphosphatemia and secondary hypocalcemia leading to complications like cardiac arrhythmias, acute renal failure, seizures, or death. It is

the end result of high cell turnover, commonly seen among hematological malignancies with high proliferative rate or those with high sensitivity to treatment [1]. It is rarely observed in patients with solid tumors as a result of therapy or even spontaneously [2].

There have been so far six published cases reporting TLS in metastatic colon adenocarcinoma, four after FOLFIRI (5-flourouracil, leucovorin, and irinotecan) or irinotecan chemotherapy, one after cetuximab therapy and one after FOLFOX (5-flourouracil, leucovorin and oxaliplatin) therapy in a patient who had received pretreatment with FOLFIRI [3–8].

We report a case of TLS after first dose of FOLFOX chemotherapy in a patient with metastatic colon carcinoma, managed successfully with intravenous hydration and rasburicase.

CASE REPORT

A 55-year-old African-American male with past medical history of hypertension, emphysema, pneumothorax, prostate cancer status post prostatectomy presented to the emergency department with epigastric and right upper quadrant stabbing pain, anorexia and weight loss for two weeks.

On arrival, patient had a blood pressure of 138/82 mmHg, pulse rate of 85 beats per minute, afebrile at 36.4°C and respiratory rate of 20/min. Physical examination was significant for right upper quadrant tenderness and hepatomegaly. Blood work was significant for acute kidney injury, hyperkalemia, leukocytosis and abnormal liver function tests and negative for HIV, hepatitis B and hepatitis C.

Ultrasound abdomen was significant for multiple hypo-densities in liver. Further imaging with CT scan showed multiple hypo-enhancing liver masses, 1.3 cm nodule in right adrenal gland, multiple cysts in kidney and a 2-cm round soft tissue mass in posterior cecum with adjacent posterior mesentery and mesenteric lymphadenopathy.

Colonoscopy was positive for edematous ileocecal valve but biopsy was negative for any malignant features. No other mass lesions were identifiable on colonoscopy. The liver biopsy was positive for malignant cells, adenocarcinoma type compatible with colonic primary. Further staining was negative for K-ras mutation and CK7 but positive for CD20. Patient also had an elevated CEA and CA 19-9 but normal AFP. Other labs were significant for CEA-9918.7 μg/L, CA 19-9- 520 U/ml, AFP-1.9 μg/L, uric acid 9.7 mg/dL and LDH 1196 U/L. Patient was given a diagnosis of stage IV colon cancer. Patient improved with supportive treatment and was discharged with oncology follow-up.

After about 20 days, patient was started on FOLFOX regimen, which consisted of leucovorin (772 mg IV), 5-fluorouracil (4632 mg IV) and oxaliplatin (164 mg IV). Next day, patient presented to the emergency department with abdominal pain, nausea, vomiting and diarrhea. Patient was found to have acute kidney injury with creatinine of 1.9 mg/dl. In addition, patient was hyperkalemic (6.4 mEq/L), hyperuricemic (20.3 mg/dl) and hyperphosphatemic (5.5 mg/dl) with normal calcium level (Table 1). There was no seizure activity or cardiac arrhythmia on telemetry monitor (Cairo-Bishop grade II TLS). Patient was admitted to the step down unit with a diagnosis of Tumor lysis syndrome and managed with aggressive intravenous hydration, furosemide and single dose of rasburicase (6 mg). In two days, symptoms resolved with improvement in potassium (4.6 mEq/L) and uric acid (5.6 mg/dl) creatinine (1.4 mg/dl) (Table 1). Patient was transferred to the general oncology floor and was discharged four days later with oncology follow-up.

DISCUSSION

There have been reports of about 100 cases of TLS in patients with solid tumors [9]. Irrespective of the cancer type, there is a 20–50% increase in mortality for undiagnosed or late-diagnosed cases of TLS in solid tumors [9]. The fatality rate for these 100 reported cases was 41% [10].

Tumor lysis syndrome is characterized by constellation of electrolyte abnormalities. Cellular material rich in potassium, phosphorous and uric acid is released as a consequence to cellular death, either mediated by cancer therapy or spontaneously in rapidly dividing tumors. This leads to hyperkalemia, hyperuricemia and hyperphosphatemia. Due to binding of calcium with phosphorous, hypocalcemia is observed. This in turn may lead to acute impairment of renal function, cardiac arrhythmogenicity, central nervous system toxicity, and eventually death [11].

Large tumor burden, liver metastates, elevated pretreatment LDH, use of combination chemotherapy drugs and dehydration are major risk factors, which place a patient at higher risk of developing TLS [2]. Hence a watchful monitoring is essential to prevent, diagnose and manage the metabolic complications associated with TLS.

There have been so far six published cases, reporting TLS in metastatic colon adenocarcinoma. Four of these were after FOLFIRI (5-flourouracil, leucovorin, and irinotecan) or irinotecan chemotherapy, one after cetuximab therapy and one after FOLFOX therapy in the setting of pretreatment with FOLFIRI therapy. Our case is the first of its type to demonstrate that TLS can occur in a metastatic colon cancer patient after a single cycle of FOLFOX therapy even in the absence of any pretreatment. Our patient had all known risk factors for developing TLS like large tumor burden, liver metastases, elevated pretreatment LDH, use of combination chemotherapy drugs and dehydration. Unlike previously reported six cases where TLS resulted in death, our patient survived. Therefore, a clinician should maintain high index of suspicion for TLS among metastatic colon cancer patients

Table 1: Comparison of laboratory parameters at initial visit, after single FOLFOX cycle and after TLS treatment

Laboratory Parameters	Initial visit (Before FOLFOX chemotherapy)	Second Visit	
		After FOLFOX chemotherapy	After rasburicase and intravenous hydration
Total Bilirubin (mg/dL)	2.3	10.1	6.0
Direct Bilirubin (mg/dL)	1.7	7.7	4.8
Alkaline Phosphatase (U/L)	646	732	415
AST (U/L)	205	344	136
ALT (U/L)	119	95	56
Uric Acid (mg/dL)	9.7	20.3	5.6
Potassium (mEq/L)	5.1	6.4	4.6
Blood Urea Nitrogen (mg/dL)	36	61	30
Creatinine (mg/dL)	1.3	1.9	1.4

and should do prompt intervention to prevent potentially life threatening complications like cardiac arrhythmias, acute renal failure, seizures, or death.

CONCLUSION

Tumor lysis syndrome (TLS) has been rarely described in metastatic colon cancer. This is the first case describing TLS in a patient with metastatic colon cancer after single cycle of FOLFOX chemotherapy. Henceforth, it is important to maintain high suspicion for TLS while treating metastatic colon cancer with chemotherapy like FOLFOX to prevent potentially life-threatening complications like cardiac arrhythmia, acute renal failure, seizure or death.

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Author Contributions

Akanksha Agrawal – 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

Deepanshu Jain – 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

Mark Morginstin – 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

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

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CASE REPORT PEER REVIEWED | OPEN ACCESS

Metastatic prostate cancer to the testis as an initial presentation: Management review and outcome

Mathew Yamoah Kyei, Verna Vanderpuye, Lawrence Edusei, James Edward Mensah, Joel Yarney

ABSTRACT

Introduction: Metastatic prostate cancer to the testis as an initial presentation is a rare condition. Though it has been noted to be associated with poor prognosis, no treatment guideline is currently available aimed at improving the outcome. This report reviews the management of a case and its outcome. Case Report: A 47-year-old male presented with a painless swelling of the left testis and rectal discomfort of three weeks duration. He had a strong family history of prostate cancer. Examination of the scrotum revealed a left non-tender testicular mass 7x5 cm that was hard with irregular surface. A digital rectal examination revealed an enlarged and nodular prostate and seminal vesicle involvement. A histological and immunohistochemical analysis of the prostatic biopsy and specimen from an inguinal orchidectomy of the left testis confirmed a metastatic prostate cancer to the testis. After three months of combined androgen deprivation therapy, he developed progressive obstructive urinary symptoms, severe pain in the anus, and a rapid increase in size of the prostate gland being castrate resistant. He underwent three dimensional conformal radiotherapy to the whole pelvis followed by seven cycles of docetaxel based chemotherapy. Nineteen months following diagnosis, his PSA was 1.86 ng/ml. Despite normal tPSA levels, there was clinical failure with development of bilateral lower limb swelling from para-aortic lymphadenopathy at two years. The patient died after two additional cycles of docetaxel chemotherapy. Conclusion: In a young patient with prostate cancer presenting initially with testicular metastasis, aggressive management strategies as pertains for castrate resistant cases need to be initiated early to improve outcomes.

Keywords: Androgen deprivation, Chemotherapy, Metastasis, Prostate cancer, Radiation, Testis

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INTRODUCTION

Metastasis of prostate cancer to the testis is a rare condition with incidence of 2–4%. [1] Most reports are incidental findings after bilateral orchidectomy for androgen deprivation therapy with the majority of the patients aged above 60 years [1, 2]. Considering tumors of solid organs, prostate cancer has been noted to be the most common cause of metastases to the testis. [3] The mechanism for metastasis of prostate cancer to the testis has been proposed to include retrograde venous extension or embolism, arterial embolization, lymphatic extension and endocanalicular spread [2, 4]. The overall prognosis is poor as the patients have high Gleason score of 8 or 9, exhibiting atypical neoplasm behavior becoming hormone refractory 6 to 8 months following diagnosis resulting in survival of between 6 to 12 months after diagnosis [2]. A challenge in diagnosis is encountered when the patient is relatively young and there is a need to rule out a primary testicular tumor as the cause of the testicular enlargement in a resource poor setting. Choosing appropriate treatment path to achieve an increase survival can be difficult as there are no established guidelines.

CASE REPORT

A 47-year-old male presented with painless swelling of the left testis of three weeks duration. There was no prior history of trauma or radiation exposure, and the left testis had descended normally into the scrotum at birth. There were no associated lower urinary tract symptoms or bowel symptoms except a mild rectal discomfort. He had no smoking history and no co-morbidities. He, however, had a strong family history of prostate cancer. His father and elder brother had been diagnosed with prostate cancer at 70 years and 55 years of age, respectively.

Examination findings revealed a healthy-looking middle aged man. He was not pale and not jaundiced. He had no palpable peripheral lymphadenopathy. No masses were palpated at abdominal examination. The examination of the left scrotum revealed a non-tender left testicular mass, 7x5 cm that was hard with irregular surface. There was an associated secondary hydrocele.

A digital rectal examination revealed an enlarged prostate with hard nodules bilaterally and seminal vesicle involvement (Clinical stage T3b).

Blood Investigations done showed a total serum prostate specific antigen (tPSA) of 25.25 ng/ml, hemoglobin level of 15.1 g/dl, total white cell count of $6.2x10^9$ /L, and erythrocyte sedimentation rate (ESR) of 10 mm/Hr. There was normal serum level of alphafetoprotein (8.6 ng/ml) (≤ 40 ng/ml) and β-hCG (0.0 mIU/ml) (<5 mIU/ml). Lactate dehydrogenase (LDH) was however elevated being 241U/L [normal range 135–225]. No abnormality was detected in the urinalysis.

A transrectal ultrasound guided biopsy of the prostate was done and reported as:

Specimen 1: Left lobe of prostate

Four of the six cores of tissue submitted contain invasive adenocarcinoma with Gleason's score of 9 (4+5), accounting for 15–90% of the volumes of the individual cores and with perineural infiltration. There was accompanying high grade prostatic intraepithelial neoplasia.

Specimen 2: Right lobe of prostate

All of the six cores submitted contain an infiltrating invasive adenocarcinoma with Gleason's score 9 (4+5), accounting for 30–100% of the volumes of the individual cores and with perineural infiltration.

A panel of immunohistochemical stains performed on the prostate needle biopsies excluded neuroendocrine differentiation (Table 1).

A computed tomography scan of the abdomen and pelvis revealed prostate tumor infiltrating the seminal vesicles, bladder base, rectum and a left iliac lymph node (Figure 1).

Radioisotope bone scintigraphy was negative for bone metastasis.

The patient had a left inguinal orchidectomy, the histology of which confirmed an adenocarcinoma tumor infiltrate of the left testis with vascular as well as intra ductal extension of the neoplastic cells (Figure 2).

Further immunohistochemical stain reactivity of the left inguinal orchidectomy specimen revealed a positive staining for α-methyl CoA racemase (P504S) and PSA confirming the diagnosis of metastatic prostate cancer to the left testis (Table 2, Figure 3).

Additional immunohistochemical stains for chromogranin, calretinin, CAM 5.2, Spalt-like transcription factor 4 (SALL4) and S-100 protein were negative. A few cells were positive for epithelial membrane antigen (EMA) but there was diffuse nuclear positivity for NKX3.1. The appearances were those of metastatic poorly differentiated carcinoma, consistent with spread from the prostatic primary.

Table 1: Immunohistochemical stain reactivity on prostate needle biopsy specimens

Panel	Stain Reactivity
Cytokeratin AE1/AE3	Positive tumor cell staining
Prostate Specific Antigen (PSA)	Focal tumor cell staining
Neural cell adhesion molecule (CD-56)	Negative tumor cell staining
Chromogranin-A	Negative tumor cell staining
Synaptophysin	Negative tumor cell staining
Thyroid transcription factor-1 (TTF-1)	Negative tumor cell staining

Table 2: Immunohistochemical stain reactivity on left inguinal orchidectomy specimen

Panel	Stain Reactivity
Cytokeratin AE1/AE3	Positive tumor cell staining
Prostate Specific Antigen (PSA)	Focal tumor cell staining
A-methylacyl CoA racemase (P504S)	Positive tumor cell staining
c-kit gene prot-oncogene (CD-117)	Positive tumor cell staining
Placental alkaline phosphatase (PLAP)	Negative tumor cell staining
CD-30	Negative tumor cell staining
Alphafetoprotein (AFP)	Negative tumor cell staining
Alpha-inhibin	Negative tumor cell staining

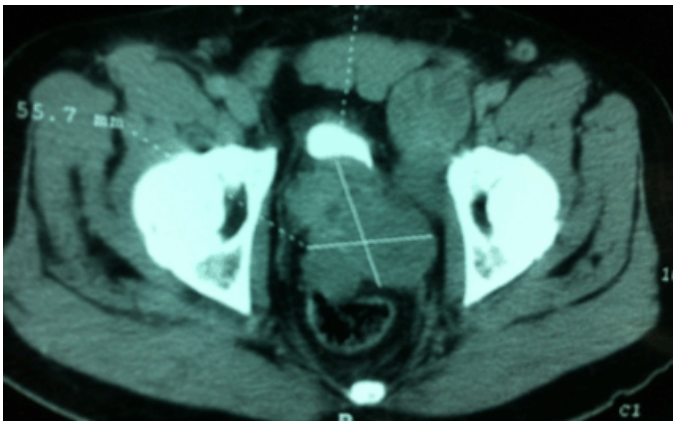

Figure 1: A pelvic computed tomography scan showing the prostate tumor infiltrating the seminal vesicles, bladder base and rectum with associated left internal iliac lymphadenopathy.

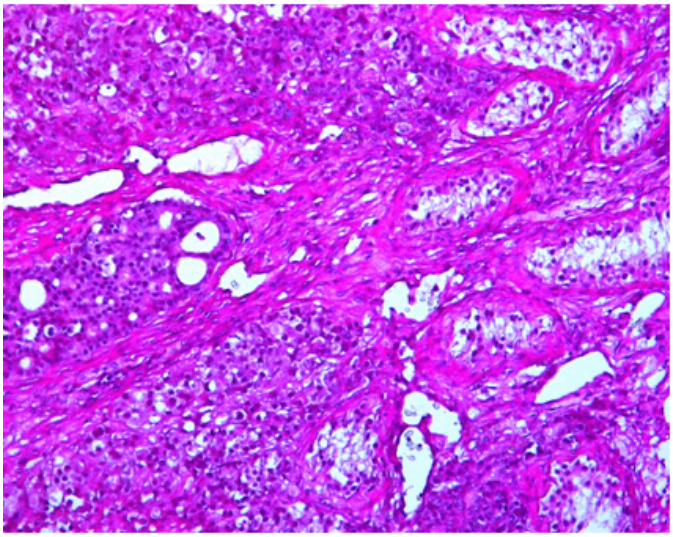

Figure 2: Residual seminiferous tubules with interspersed islands of the metastatic adenocarcinoma (H&E stain, x100).

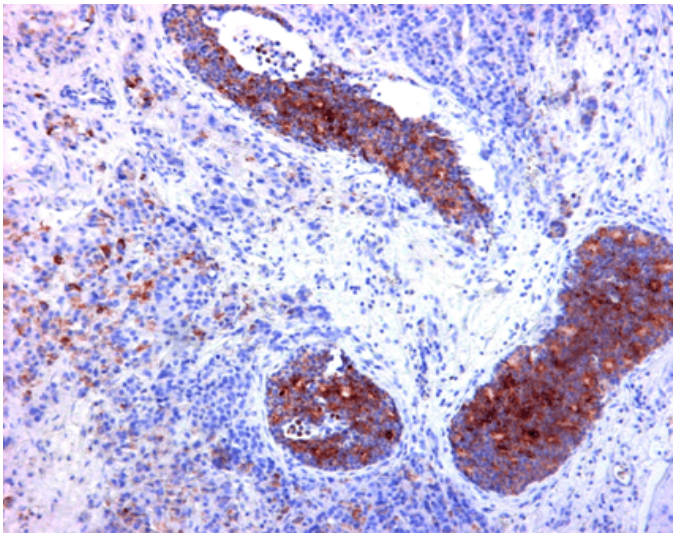

Figure 3: Focal positive staining for prostate specific antigen.

The patient was initially started with androgen deprivation therapy with bicalutamide 50 mg daily followed by subcutaneous injection of goserelin acetate 10.8 mg depot two weeks into starting bicalutamide. However, the serum total prostate specific antigen rose to 73.15 ng/ml after three months from the initial 25.25 ng/ml. The trans rectal prostate volume increased from 39.48 g to 219.84 g over the same period despite the serum testosterone level remaining castrate (<1.0 ng/ml). He developed pain in his rectum, difficulty passing stools and obstructive urinary symptoms. This indicated disease progression being castrate resistant. In order to control the local disease, he received palliative radiotherapy using 3D conformal technique to a dose of 50 Gy to the prostate and seminal vesicles and 45 Gy to the pelvic nodes over five weeks. Post radiotherapy PSA was 5.72 ng/ml and his symptoms abated. He subsequently received chemotherapy with Docetaxel at 75 mg/m^2 every three weeks with prednisolone 5 mg twice daily for 7 cycles, which he tolerated well. His PSA decreased further to 1.86 ng/ml 3 months after the last dose of chemotherapy and 11 months following diagnosis. The patient's general condition and PSA remained stable with a good performance status and improved quality of life 19 months after initial diagnosis. Despite normal tPSA levels, there was clinical failure with development of gross bilateral lower limb swelling from para-aortic lymphadenopathy at two years. The patient died after two additional cycles of docetaxal chemotherapy.

DISCUSSION

Metastasis of prostate cancer to the test presenting clinically is a rare event as most are incidental findings at orchiedectomy for advanced prostate cancer or at autopsy

[1, 4]. There have been reports of patients presenting with metastasis to the testis long after treatment for the primary condition with the period between diagnosis and metastasis to the testis found to be between 2.5–15 years. [1, 5]. It has been noted that symptomatic metastasis to the testis tended to be solitary, unilateral and may simulate primary neoplasms including rete adenocarcinoma and sertoli cell tumor [3] thus in this middle aged man of 47 years a primary testicular tumor was of concern. Without a prior diagnosis of prostate cancer, a thorough physical examination including a digital rectal examination revealed the possible association between the testicular mass and the prostatic lesion. A panel of immunohistochemical tests showing positive staining for α-methyl CoA racemase (P504S), PSA and NKX3.1, achieved through collaboration, was required to arrive at the diagnosis. Bilateral total orchidectomy was not performed as the initial intent was for diagnostic purposes as the patient was relatively young and for cultural reasons most men prefer chemical castration. Despite the maximum androgen blockade, there was early evidence of castrate resistance resulting in the initiation of docetaxal based chemotherapy. This supports findings by other researchers of an early onset of castrate resistance in patients presenting with testicular metastasis. [2]

The drastic drop in PSA from 73 ng/ml to 5.72 ng/ml with an associated improved quality of life following local radiotherapy indicate the usefulness of radiotherapy in controlling localized hormone refractory disease and has been supported in some studies [6]. We choose to continue androgen deprivation in spite of the refractory state in addition to the chemotherapy as it has been found to delay progression and improve survival. [7] We were, however, limited to the use of docetaxal based chemotherapy which resulted in an improved survival of 2 years as we had no access to other second line agents such as enzalutamide, abiraterone, or sipuleucel-T.

As have been noted earlier, no established treatment lines have been stated. In the reports in literature which were incidental findings after orchidectomy, as a part of primary treatment, no further treatments are proposed with the patients noticed to do well [1, 2]. For the cases with metastasis to the testis detected long after initiation of treatment for the prostate cancer, androgen deprivation had been continued even though they had the potential to be aggressive [5]. Manikandan et al. had improved survival (four years at time of their report) in their case with the patient receiving 8 cycles of chemotherapy followed by atrasentan 10 mg/day [8]. The use of multimodal therapy in our patient presenting with testicular metastasis without prior treatment for his prostate cancer led to an improved survival of two years as against a survival of less than one year observed in other cases/ series [2].

As more cases of prostate cancer with metastasis to the testis are being reported, there is the need for evidenced based treatment guidelines to improve outcomes. Early initiation of docetaxel based chemotherapy together with local control of the prostate cancer using radiation therapy improved the outcome in this relatively young patient.

CONCLUSION

Aggressive management strategies including androgen deprivation, early initiation of docetaxel based chemotherapy and radiotherapy therapy for local disease control are warranted in patients with prostate cancer presenting clinically with testicular metastases to improve quality of life and overall survival. Evidenced based treatment guidelines are needed as more cases are being reported.

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Author Contributions
Mathew Yamoah Kyei – 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
Verna Vanderpuye – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published
Lawrence Edusei – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published
James Edward Mensah – Substantial contributions to conception and design, Revising it critically for important intellectual content, Final approval of the version to be published
Joel Yarney – Substantial contributions to conception and design, Acquisition of data, Revising it critically for important intellectual content, Final approval of the version to be published

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CASE REPORT PEER REVIEWED | OPEN ACCESS

Emphysematous gastritis after bone marrow transplant: Successful conservative management

Hammad Liaquat, Imad Jaafar, Matt Bojanowski, Endashaw Omer

ABSTRACT

Introduction: Emphysematous gastritis (EG) has a nonspecific presentation, so radiography is an essential diagnostic tool. There is usually a high mortality rate and poor prognosis associated with EG. Case Report: A 38-year-old male with a history of recurrent seminoma who received chemotherapy and underwent stem cell transplantation before he was diagnosed with EG. During the hospital course, the patient developed gastrointestinal symptoms, neutropenic fever and, subsequently, multi-organ failure. Medical imaging reported findings consistent with EG. The patient was managed conservatively with subsequent recovery and resolution of EG on repeat medical imaging. Conclusion: Emphysematous gastritis in the setting of immunosuppression and multi-organ failure can be managed successfully with prompt diagnosis and conservative management.

Keywords: Bone marrow transplant, Conservative management, Emphysematous gastritis, Multi-organ failure

How to cite this article

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INTRODUCTION

Emphysematous gastritis (EG) is the presence of gas within the stomach wall in association with clinical sepsis [1]. Initially reported in 1889 by Fraenkel, EG is a rare illness diagnosed with the support of imaging and is associated with mortality rate reported to be as high as 62%. Approximately 70 cases of EG have been reported in literature so far. There are some reports of successful conservative management in uncomplicated EG while surgery is usually required in the presence of complications like perforation or peritonitis [2]. We present a case of a man with testicular cancer who developed EG after recent chemotherapy and stem cell transplantation and was managed successfully with conservative treatment.

CASE REPORT

A 38-year-old male with a history of relapsed seminoma was admitted to the University Hospital for autologous stem cell transplantation after he had received several cycles of neoadjuvant chemotherapy. One day after successful bone marrow transplant he developed nausea, vomiting, abdominal pain and diarrhea. Subsequently, he developed fever with daily temperature spikes of 102–105°F. He became progressively pancytopenic and, three days after stem cell transplant, his absolute neutrophil count reached nadir of 100 cells per microliter. Patient continued to deteriorate clinically and developed multi-organ failure. He was put on mechanical ventilation, given intravenous fluid resuscitation and broad spectrum antibiotics empirically (meropenem, vancomycin, micafungin and acyclovir). His renal function continued to worsen and hemodialysis was initiated. All infectious workup was negative except for a positive cytomegalovirus IgM antibody. Plain film of the chest did not show pneumonia. His gastrointestinal symptoms continued to worsen. A CT scan of the abdomen without contrast revealed diffuse pneumatosis of the stomach consistent with a diagnosis of EG (Figure 1). No portal venous gas or free intraperitoneal air was seen on CT scan.

The surgery consult service was consulted and they recommended medical management. He received intravenous hydration, total parenteral nutrition, bowel rest and gastric decompression with nasogastric tube. The patient was monitored with daily plain films of the abdomen. Antiviral therapy was switched to ganciclovir.

After three weeks of conservative therapy, the patient's clinical status began to improve. His abdominal pain and diarrhea resolved, and gastric output was negligible. Serial plain films of the abdomen showed decreasing distension of the stomach and small bowel. He was successfully extubated, and the nasogastric tube was removed. The patient continued to recover well clinically and lab abnormalities resolved. His diet was advanced slowly, which was well tolerated. A repeat abdominal CT scan reported resolution of EG (Figure 2). He was later discharged from the hospital with instructions for outpatient oncology, nephrology and gastroenterology follow-up.

The patient was seen in gastroenterology clinic two weeks after discharge from the hospital. He had no gastrointestinal complaints, had normal bowel function and was eating a normal diet. His blood electrolytes and cell counts continued to be within normal range.

DISCUSSION

Emphysematous gastritis is often associated with conditions that damage the gastric mucosal barrier and thus enable bacteria to establish themselves in this unusual location. Known associated conditions include ingestion of corrosives, alcohol abuse, renal failure, diabetes, non-steroidal anti-inflammatory drug usage, pancreatitis, gastroenteritis and recent abdominal surgery [2, 3]. There have been cases of associated bacterial infection with *Clostridium welchii*, *Escherichia coli*, *Streptococcus*, *Bacillus subtilis,* proteus, *Pseudomonas* and *Enterobacter* [1, 4]. Our patient was

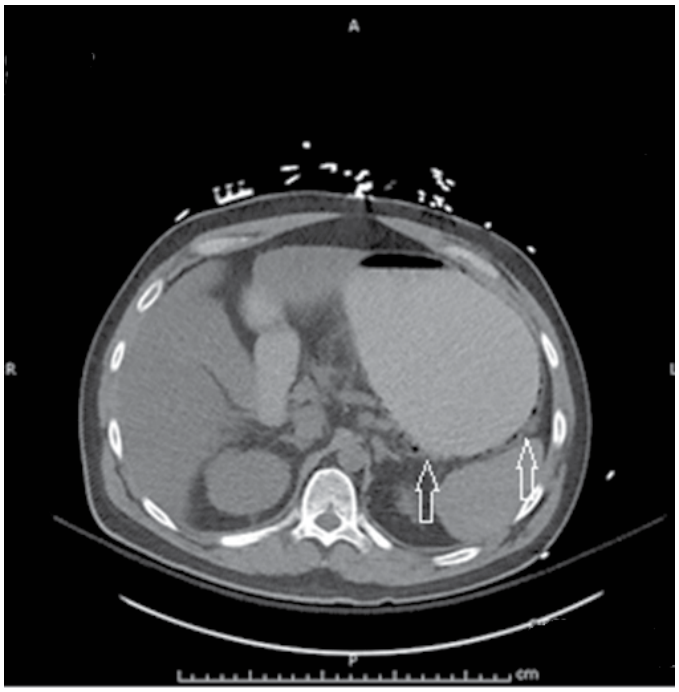

Figure 1: Axial image of the non-contrast computed tomography scan of the abdomen with the arrows showing areas of pneumatosis in the wall of the stomach supporting diagnosis of emphysematous gastritis.

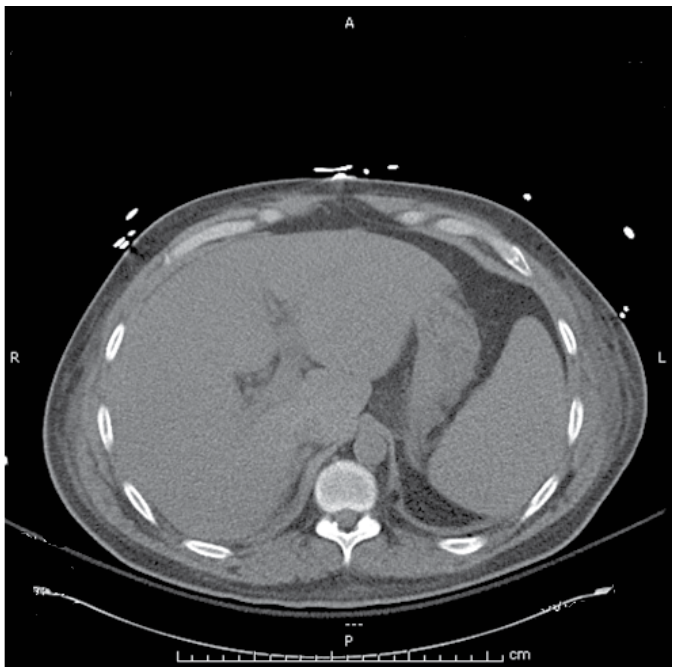

Figure 2: A second axial image on a non-contrast computed tomography scan of abdomen one week later showing resolution of emphysematous gastritis.

immunosuppressed, developed renal failure and tested negative on all bacterial cultures.

There is no established diagnostic criteria for EG [2]. Previous studies have suggested severe symptoms, characteristic medical imaging and bacterial infection as diagnostic markers. The clinical course and medical imaging in our patient was characteristic of EG.

Typically, gas is unusual within the stomach due to its mucosal barrier, acidity, and strong blood supply [3, 4]. It can be a sign of underlying disease and is found in EG as well as gastric emphysema. Gastric emphysema (GE) is usually associated with an asymptomatic clinical course without co-existent microbial infection and early spontaneous resolution [5, 6]. Severe clinical illness and co-morbidity, microbial infection usually with gas-forming bacteria, and high mortality rate are hallmarks of EG. There is also a greater likelihood for the need of surgery in a patient with EG [7]. Computed tomography scan of the abdomen is the best imaging modality to differentiate between GE and EG. Characteristic circular air bubbles are found in GE, while thickened gastric folds along with linear streaks of air are found in EG. The presence of portal venous gas can also be found with EG and is usually concerning for development of mesenteric ischemia [3].

Our patient underwent stem cell transplant prior to his presentation of EG. This may be the first reported case of a patient who developed EG after autologous stem cell transplant. A case in Korea has developed gastric pneumatosis mimicking an intestinal perforation after hematopoietic stem cell transplantation, but he did not develop EG [8]. The most closely related report has been a case of a patient with aplastic anemia developing emphysematous gastritis [4]. Our patient also uniquely presented with a positive IgM result for cytomegalovirus (CMV). The CMV is the most commonly recognized viral infection of the stomach [9] but a past literature search does not show a case of emphysematous gastritis with CMV infection. Our patient may be the first reported case in this regard as well.

There is no consensus about the optimal management of EG. Conservative medical management for EG has been successful in several cases [2, 4, 10]. The management in these cases included bowel rest, hyperalimentation via central venous nutrition, broad-spectrum antibiotics, proton pump inhibitors and a nasogastric tube. Despite immunosuppression and multi-organ failure, our patient recovered with standard medical management. Some patients with similar co-morbidity have not survived similar management in the past [11–13]. Surgical intervention has been successful for cases of EG complicated by gastric or intestinal perforation, ischemia, and unsuccessful medical management [4, 14, 15].

CONCLUSION

Emphysematous gastritis is a rare and critical illness. It has non-specific presentation and has high mortality rate. Prompt diagnosis and conservative management can prevent complications and adverse outcome as well as result in complete recovery of patient.

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Author Contributions

Hammad Liaquat – 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
Imad Jaafar – Acquisition of data, Analysis and interpretation of data, Drafting the article, Final approval of the version to be published
Matt Bojanowski – Acquisition of data, Analysis and interpretation of data, Drafting the article, Final approval of the version to be published
Endashaw Omer – Substantial contributions to conception and design, 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 PEER REVIEWED | OPEN ACCESS

Medullary thyroid carcinoma associated with multiple myeloma: A case report

Seyyed Mohammadreza Mortazavizadeh, Yasaman Ayoughi, Fariba Binesh, Moein Karbalaeian

ABSTRACT

Introduction: The co-existence multiple myeloma (MM) and another cancer have been reported severally in literature, but medullary thyroid carcinoma (MTC) as a second cancer with multiple myeloma is an extremely rare case, to our knowledge, there are no prior reports of these concurrent cancers. Case Report: A 54-year-old man was admitted with chief complain of bone pain, clinical investigation revealed, lytic bone lesions and infiltration of plasma cells in bone marrow and patient with diagnosis of multiple myeloma received chemotherapy, he was in partial remission, but nine months later a solitary nodule was founded in his thyroid, with high levels of calcitonin and fine needle aspiration (FNA) reported the medullary thyroid carcinoma, after total thyroidectomy which confirmed this cancer the patient recovered and now he is followed-up for these cancers. Conclusion: Diagnosis of secondary cancer is important because: first to distinguish secondary malignancy from metastatic cancer and then try to investigate the etiology of this association.

Keywords: Medullary thyroid cancer, Multiple Myeloma, Secondary malignancy, Thyroid carcinoma

How to cite this article

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INTRODUCTION

Multiple myeloma (MM) is a hematologic cancer which usually manifested by bone pain, hypercalcemia, anemia, lytic bone lesions and monoclonal gammopathy. The co-existence multiple myeloma and another cancer such as renal cell carcinoma, lung, gastric carcinoma, colon, prostate, breast, bladder, uterus, and liver have been reported severally in literature [1–5], but medullary thyroid carcinoma (MTC) as a second malignancy with multiple myeloma is an extremely rare case which seems has not been reported before, here we are presenting a case with these two types of cancer.

CASE REPORT

A 54-year-old male with diabetes history presented on 17th November 2013 with complaints of severe bone pain on his shoulder and vomiting. The physical examination was unremarkable but a primary photo X-ray in emergency room showed abnormal lytic lesions in the bone and pathologic fracture in clavicle (Figure 1) , so he was referred to the oncologist with high suspicious for a malignancy, the primary laboratory investigation revealed ESR:77 mm/h , the skull X-ray showed multiple punched out lesions (Figure 2)

And the whole body scan reported multiple irregular and abnormal uptake in anterior and posterior rib-skull-right shoulder and highly suggestive multiple bone metastasis (Figure 3) in the next laboratory investigation yielded the following findings: β_2-microglobulin :9.9(1.1-2.4) mg /L, α_2:17.6 (7.1-11.8) g/L , in his urine electrophoresis, Lambda light chain was 88 (5.9)mg/L, (detailed results laboratory tests are given in Table 1), the aspiration of bone marrow found 25% plasma cells which some were multiple nucleus and others looking plasma blast (detailed reports of bone marrow aspiration is given in Table 2) and biopsy of bone marrow reported that hematopoietic cells have been replaced by proliferation of plasma cells and finally with diagnosis of the multiple myeloma, chemotherapy was initiated for patient with the regiment of VAD (vincristine, adriablastina, , dexamethasone) and zometa.

After four cycles chemotherapy, once more bone marrow aspiration revealed incomplete response to cure with 15 % plasma cells and evaluation of free lambda light chain was 124 (up to 5.9) mg/L so the chemotherapy changed and continued with velzomib, and once more bone marrow aspiration showed mild plasmacytosis 7% and the patient reached a partial remission, about after nine months later, the laboratory tests changed and ESR elevated for second time, during investigation we found a palpable solitary nodule in the thyroid of patient, the thyroid function test showed TSH: 2.6 mlU/L, T4: 8.5 µg/dL and T3:142 ng/dL and ultrasonography of thyroid reported a well-defined hypoechoic solitary nodule in middle of right lobe of thyroid with numerous and small zones of calcification with no adenopathy in neck and suggested for fine needle aspiration (FNA) because it was suspicious for malignancy , in that time some differential diagnosis were made: metastasis of multiple myeloma to thyroid, or primary thyroid malignancy metastasis to bone and our first diagnosis of multiple myeloma could be rejected by this assumption and the last one diagnosis was thyroid malignancy as a secondary cancer. Therefore, the patient underwent FNA which reported medullary thyroid carcinoma with the laboratory test investigation of calcitonin: 268 ng/L, CEA: 78 µg/L, ESR: 63 mm/h and with these evidences the probability of metastatic of primary multiple myeloma ruled out. And with diagnosis of MTC, patient was referred to surgeon and after total thyroidectomy, the histopathology after surgery confirmed medullary thyroid carcinoma (Figure 4).

After surgery the patient was recovered and the laboratory tests return to nearly normal levels. The patient remained in good health and he is followed-up for these malignancies.

DISCUSSION

Second primary cancer is one of the effective prognostic factors among cancer survivors and it is estimated to be the sixth most common form of malignancy in the world [6], although secondary solid neoplasms in patients with multiple myeloma are rare, but some studies have reported the increasing risk of that. In a study by Stegeman et al., multiple myeloma and solid tumor co-existence was found to be 3% [7] and Kyle at al. reported that, in 1027 patients diagnosed with multiple myeloma, the proportion of secondary hematologic malignity was 0.7% and the proportion secondary solid malignancy was 1.9% [8]. As we mentioned earlier, malignancies synchronized with

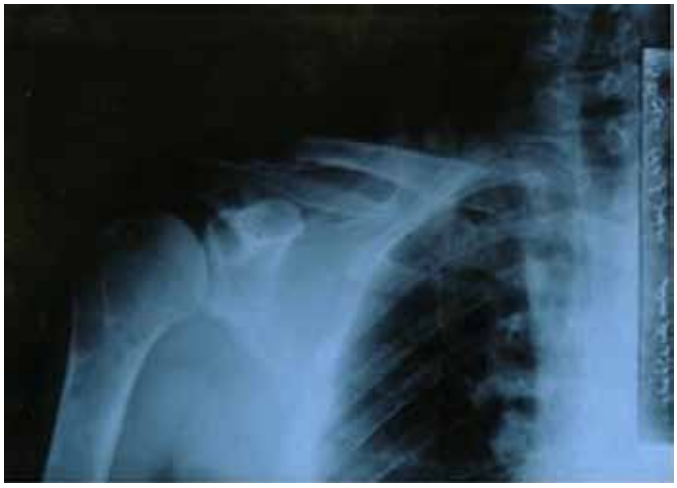

Figure 1: Lytic bone lesions and pathologic fracture in shoulder.

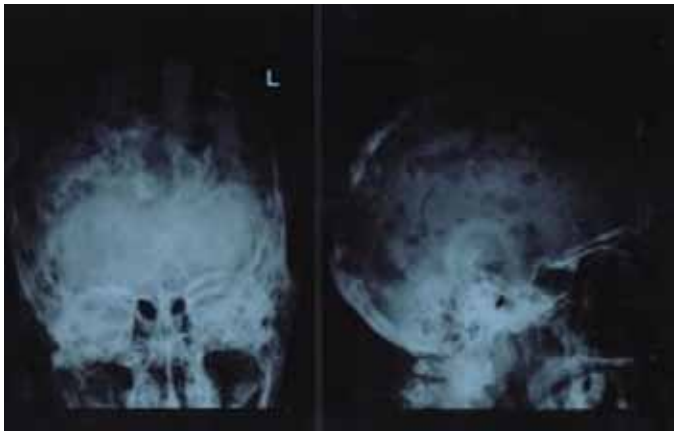

Figure 2: Numerous punched lesions in skull.

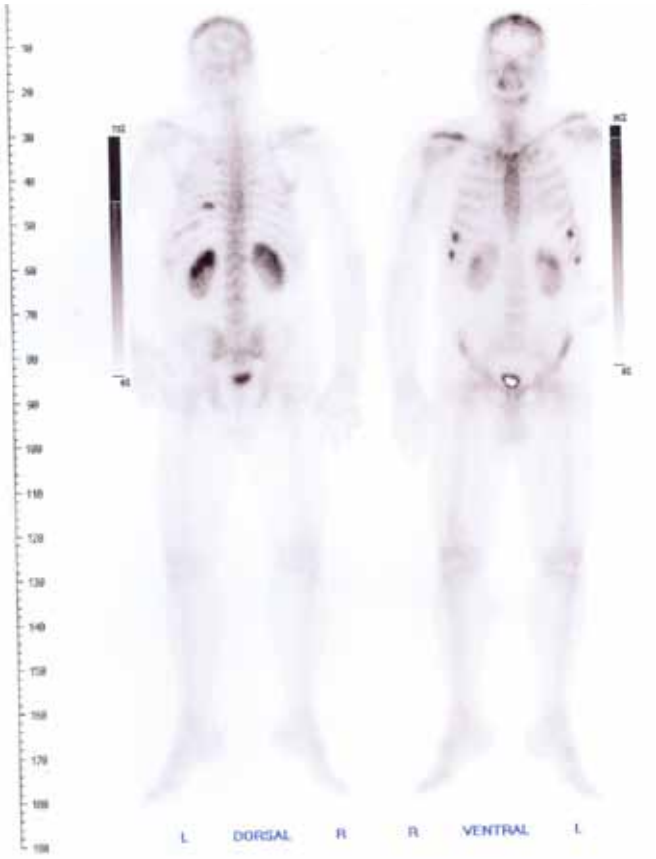

Figure 3: Bone scan revealed multiple metastasis.

multiple myeloma have been reported before [1–5]. But it was not included medullary thyroid carcinoma which occurs extremely rare, to our knowledge there are no prior reports of this case. The etiology of this correlation in our case is not clear but according to some hypothesis, secondary malignancies associated with MM can be because of behaviors (such as smoking, harmful levels of alcohol consumption and poor diet), inherited susceptibilities and/or the medical treatment

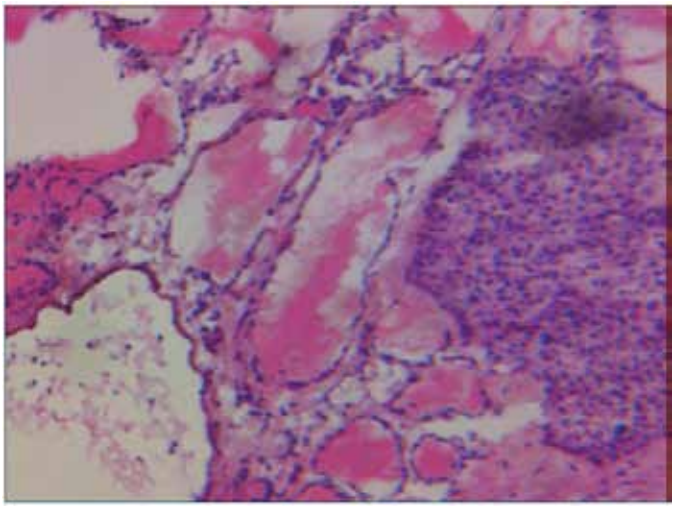

Figure 4: Tumor cells with circle nucleus and granular cytoplasm and there are amount of eosinophilic amorphous between them (H&E stain, x200).

Table 1: Laboratory results of the patients

		Protein fractions (g/L)	
White blood cell	8600 mm^3	Albumin	54.4 9
Hemoglobin	13.9 g/dL	α 1	4.6
Platelet	282000 mm^3	α 2	17.6
ESR	77 mm/h	β 1	4.7
BUN	10 mg/dL	β 2	4.5
Creatinine	1 mg/dL	γ	14.2
LDH	408 U/L		
Alkaline Phosphatase	279 U/L		
SGPT	15 U/L		
SGOT	20 U/L		
Total Bilirubin	1.1 mg/dL		
β 2 microglubulin	9.9 mg/L		
IgG	1577 mg/dL		
IgM	81.6 mg/dL		
IgA	108 mg/dL		

ESR: erythrocyte sedimentation rate, BUN: blood urea nitrogen, LDH: lactate dehydrogenase, SGPT: serum glutamate-pyruvate transaminase, SGOT: serum glutamic oxaloacetic transaminase. Ig: Immunoglobulin

Table 2: Bone marrow biopsy results

Cell count in bone marrow (%)	First time (BMA) Before starting chemotherapy	Second time(BMA) After four cycle chemotherapy	Third time (BMA) After changing chemotherapy to velzomib
Blast	4	4	3
Promyelocyte	7	7	9
Metamyelocyte	8	1	6
Myelocyte	6	3	11
Band cell	12	3	4
Neutrophil	11	17	17
Eosinophil	2	5	3
Plasma cell	25	15	7
Erythroid cells different	14	46	35
	Megakaryocytes sporadically seen	Megakaryocytes sporadically seen	Megakaryocytes seen enough
	Diagnosis: Plasma cell myeloma	Diagnosis: Incomplete response	Diagnosis: Mild plasmacytosis

that cancer survivors have received [9–10]. Further investigations would be done in order to understand of whether this secondary malignancy is only accidental or whether there is any cause and effect relationship, another point of this case is about the differential diagnosis of thyroid nodule which was discovered some months after diagnosis multiple myeloma that could be mistaken by metastasis of multiple myeloma to thyroid gland which has been reported severally [11–13] or the primary cancer was MTC which could metastasis to bone and mimicked the multiple myeloma and it could change the way of treatment. But pathology samples and the elevation of laboratory markers such as calcitonin level excluded it and responding to cure and remission after total thyroidectomy, ruled out the second suspicious diagnosis. And another things which should be kept in mind in these kind of cases, try to investigate and pay attention to the new signs and symptoms which set during disease and may not be related to the primary cancer or complications of it, and another associated disease should be considered even if it is too unusual. In this case, patient reached to remission after chemotherapy but once more elevated ESR caused to investigate more.

CONCLUSION

A rare correlation between multiple myeloma and medullary thyroid carcinoma case is reported, diagnosis of secondary cancer is important because: first: to distinguish secondary malignancy from metastatic cancer then try to investigate the etiology of this association and finally to determine to choose the best treatment because of its good prognosis.

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Author Contributions

Seyyed Mohammad Reza Mortazavizadeh – 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
Yasaman Ayoughi – 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
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Moein Karbalaeian – Acquisition of data, or 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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CASE REPORT PEER REVIEWED | OPEN ACCESS

Hernia of the broad ligament of the uterus

Satoshi Tokuda, Hajime Orita, Tomoaki Ito, Mutsumi Sakurada, Tomoyuki Kushida, Hiroshi Maekawa, Miki Yamano, Ryo Wada, Koichi Sato

ABSTRACT

Introduction: Intestinal obstruction is commonly seen at emergency services but is not usually caused by internal hernias (organs or intestinal tract invaginated in the abdominal cavity, purse and hiatus). Herniation through defects of the broad ligament is especially rare. In this study, we report a case that was difficult to diagnose due to a lack of useful information; a 49-year-old woman with intestinal obstruction because of hernia of the broad ligament of the uterus. Case Report: A 49-year-old female presented with intermittent abdominal pain. Laboratory findings showed only slight leukocytosis (white blood cell count 12,500/μl). Four days later, her condition had worsened. Computed tomography (CT) scan showed distended loops and fluid in the Douglas' pouch. We thought that the cause was internal hernia and decided to operate. During surgery, the hernia of the broad ligament of the uterus was discovered. We resected the fallopian tube and proper ligament of the ovary and released the hernia. The postoperative course was uneventful and the patient was discharged on the day-8. Conclusion: We treated a case of an internal broad ligament hernia which was difficult to diagnose.

Keywords: Broad ligament, Ileus, Internal hernia, Intestinal obstruction, Uterus

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INTRODUCTION

Internal hernias are very rare accounting for about 1% of all hernias [1]. Add to that, herniation through defects of the broad ligament of the uterus is even rarer (5% of internal hernias) [2]. The broad ligament of the uterus is one of the structures which fix the position of the uterus (along with the round ligament of the uterus, transverse cervical ligament and uterosacral ligament). This hernia can be caused by congenital abnormality, pregnancy, pelvic inflammatory disease, aging and so on.

We report a difficult case to diagnose the hernia from the clinical features and were anxious about the surgical indications.

CASE REPORT

A 49-year-old woman presented with intermittent abdominal pain around her umbilicus and nausea. There was no rebound tenderness and abdominal guarding. She had the symptoms for a half day before coming to our hospital. She had myoma of the uterus and cavernous hemangioma but had never undergone surgery. Laboratory findings showed only slight leukocytosis, and X-ray showed a few dilated small bowel loops in the abdomen. We found the same distended small bowel loops in the CT scan (Figure 1).

The patient was admitted to Department of Surgery, Juntendo Shizuoka Hospital, Shizuoka, Japan with ileus and treated with fast transfusion (fluid resuscitation) and nasogastric tube. From the tube, we found drainage of about 400–500 ml/day.

Four days later, her stomachache had worsened, so we did another X-ray and CT scan (Figure 2).

Laboratory results had not changed (only leukocytosis was found) and there was no indication of intestinal disorders. We found an increase in the size of the small bowel loops with air on the X-ray. The CT scan showed distended loops and fluid in the Douglas' pouch. There was large volume ascites in the abdomen, so we decided to operate for the ileus.

For her safety, we selected a laparotomy. At surgery, we saw the ascites (serous) about 700 ml. The small intestines were edematous, the mesentery was strangulated by the fallopian tube and the blood flow to the intestine was decreased.

We resected the fallopian tube and proper ligament of the ovary and released the hernia. We washed the intraperitoneal with warm saline solution. We did not need to resect the intestine because of the recovery of the blood flow (Figure 2C–E).

The postoperative course was uneventful and the patient was discharged on day-8.

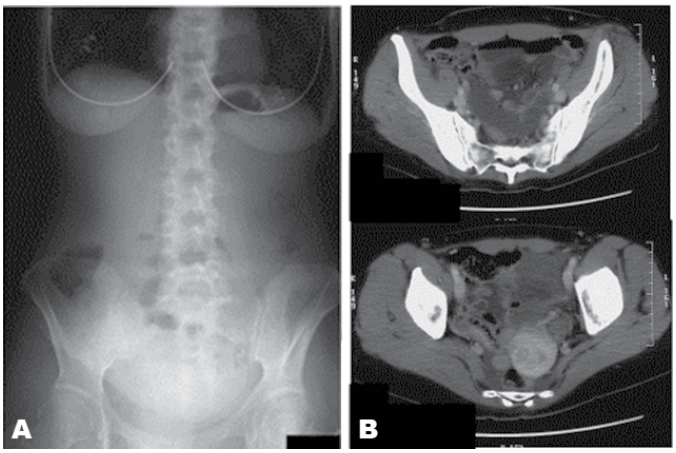

Figure 1: (A, B) The X-ray and computed tomography (CT) scan were taken on admission. The X-ray showing no abnormal gas. Axial CT scan of the abdomen with intravenous contrast administration showing only distended small intestine.

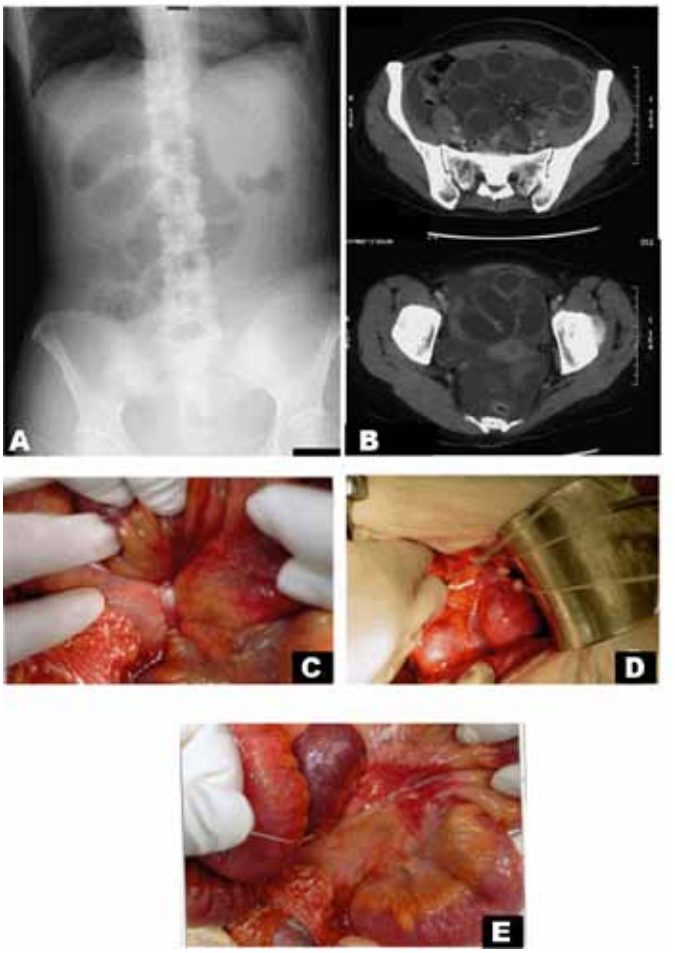

Figure 2: (A, B) The X-ray and computed tomography (CT) scan were taken four days later. X-ray showing an increase in the small intestinal gas. Computed tomography scan showing the distended loops with air-fluid level in the Douglas' pouch. Mesenteries and blood vessels concentrate in the vicinity of uterus. The distended loops push the uterus to contralateral and ventral side. In addition, the loops push the sigmoid colon and rectum to the back side, (C) The mesentery was tightened by the Fallopian tube, (D) The situation after dissection of the fallopian tube and ligament of ovary, (E) The mesentery congested with blood.

DISCUSSION

Internal hernias are rare and are classified into seven groups; paraduodenal, foramen of Winslow, pericecal, intersigmoid, transmesenteric, transomental and retroanastomotic [3]. Of internal hernias, the paraduodenal type is the most common (30–53%) [4]. An internal hernia of the broad ligament is an extremely rare cause of small bowel obstruction [1, 2].

The causes of the type of hernia can be roughly divided into two types; congenital (ruptured cystic structures reminiscent of the mesonephric or mullerian ducts) and acquired (operative trauma, pregnancy and birth trauma, or prior pelvic inflammatory disease) [5]. In addition, the type of hernia often occurs on the left side because of the defect of the broad ligament [6].

Hunt classified hernias into two types; fenestra and pouch [7]. The fenestra type is an internal hernia that

goes through the broad ligament, on the other hand the pouch type is formed by a defect in the anterior or posterior aperture. Our case was a fenestra type hernia.

We found cases in the literature illustrating the effectiveness of CT scan for the diagnosis of internal hernia of the broad ligament. Symptom included (i) distended loops with air-fluid level in the Douglas' pouch, (ii) distended loops pushing against uterus, rectum and sigmoid colon, and (iii) convergence of the mesentery and blood vessels for the uterus [8]. In our case, the patient had no history of surgery.

We saw the leukocytosis in her laboratory report, but were not able to get other useful information from it.

On admission, CT scan showed only enlargement of a portion of the small intestine, so there was no indication of internal broad ligament hernia. We retrospectively checked the CT scan which had been performed four days after admission and noticed the distended bowel loops pushed against the uterus, rectum and sigmoid colon. We could see the convergence of the mesentery and blood vessels on the uterus. It is said that the certain features of the CT scan are very useful for the diagnosis [8]. However, the CT scan which had been performed on admission did not have the features.

It was very important to check the chronological change after hospitalization. We found the change in her condition and consequently were able to treat her.

Internal hernia of the broad ligament has no characteristic physical symptoms, so a CT scan is very valuable in diagnosis. We should not hesitate to take CT scans repeatedly, even if the first image does not have specific features. If the condition is diagnosed in advance, the operation can be performed using laparoscope.

We can see the effectiveness of laparoscopy for this type of hernia in the literatures [9, 10]. Laparoscopic surgeries result in a better postoperative course and shorter hospitalization than open surgeries [11].

CONCLUSION

We treated a case of an internal broad ligament hernia which was difficult to diagnose. Computed tomography (CT) is very useful for diagnosing of such cases. The CT scan which was performed in our case showed the features of the hernia retrospectively. We should keep this condition in mind when we treat women with ileus.

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Author Contributions

Satoshi Tokuda – Substantial contributions to conception and design, Acquisition of data, Drafting the article, critical revision of the article, Final approval of the version to be published
Hajime Orita – Substantial contributions to conception and design, Acquisition of data, Drafting the article, critical revision of the article, Final approval of the version to be published
Tomoaki Ito – Acquisition of data, Drafting the article, Final approval of the version to be published
Mutsumi Sakurada – Acquisition of data, Drafting the article, Final approval of the version to be published
Tomoyuki Kushida – Acquisition of data, Drafting the article, Final approval of the version to be published
Hiroshi Maekawa – Acquisition of data, Drafting the article, Final approval of the version to be published
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Ryo Wada – Acquisition of data, Drafting the article, Final approval of the version to be published
Koichi Sato – Acquisition of data, Drafting 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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CASE REPORT PEER REVIEWED | OPEN ACCESS

Congenital myotonic dystrophy with persistent pulmonary hypertension in the newborn

Vladan Milovanov, Yanko Petkov

ABSTRACT

Introduction: Congenital myotonic dystrophy type 1 (CDM1) is a trinucleotide repeat disorder with early onset of symptoms and high neonatal mortality. Most patients with CDM1 have >1000 CTG repeats; a high number of CTG repeats generally indicate severe disease. CDM1 complicated by persistent pulmonary hypertension in the newborn (PPHN) has seldom been reported, and all previously reported cases have resulted in neonatal death. Case Report: We present a neonate with CDM1 complicated by PPHN with early onset of symptoms and severe course of disease as a result of anticipation by maternal transmission. A triplet repeat primed polymerase chain reaction (TP-PCR) analysis showed CDM1 with only 800 CTG repeats. The patient was successfully treated by inotropic support and mechanical ventilation. Conclusion: This is the first reported case of CDM1 with PPHN that did not result in neonatal death. More information on association between PPHN and number of CTG repeats in neonates with CDM1 is needed.

Keywords: Congenital myotonic dystrophy, Newborn, PPHN, TP-PCR analysis, Neonatal death

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INTRODUCTION

Congenital myotonic dystrophy type 1 (CDM1) is a multisystem muscle disorder that can present prenatally with polyhydramnios and reduced fetal movements, or in the neonatal period with hypotonia, respiratory distress, facial muscle weakness and clubfoot. The estimated prevalence of CDM1 ranges from 1:10 000 to 1:100 000, depending on the geographic region [1]. CDM1 is caused by expansion of a CTG trinucleotide repeat in the non-coding region of the myotonic dystrophy protein kinase (DMPK) gene, located on chromosome 19q13.3 [2]. The normal CTG repeat size is 5–34, [1, 2] and most patients with CDM1 have more than 1000 CTG repeats [2]. There are no reported cases of CDM1 with PPHN, with a CTG repeats number <1000. We present a case of severe CDM1 complicated by PPHN, with only 800 CTG repeats.

CASE REPORT

The case concerns a male born by vaginal delivery with vacuum extraction at 38+3 weeks of gestation

after a pregnancy complicated by polyhydramnios. His birth weight was 3292 g. His immediate Apgar score was 7, but after three minutes he became limp and cyanotic with feeble respiratory efforts. He was resuscitated by intubation and administration of 100% oxygen and surfactant in the delivery room. The initial examination showed profound hypotonia, facial diplegia and a triangular-shaped mouth. Laboratory evaluation revealed hydrogen ion (pH) 7.10, partial pressure of carbon dioxide (pCO_2) 11.7 kPa, hydrogen bicarbonate 19 mmol/l, base excess (BE) 0.6 mmol/l. Complete blood count, serum glucose and electrolyte, and infection parameters were normal. Chest radiography showed thin ribs and an elevated right hemidiaphragm, which are typical signs of CDM1 (Figure 1). Arterial mean blood pressure was 30 mmHg. Pre-ductal and post-ductal pulse oxygen saturation monitoring showed a difference of 10%. He developed PPHN, confirmed by echocardiographic examination. The PPHN was effectively treated by inotropic support and mechanical ventilation. He was extubated on day-four, and subsequently required intermittent administration of continuous positive airways pressure (CPAP) support during period of the infancy. Initial brain ultrasound showed mild bilateral ventricular dilation. The ventricular dilation gradually progressed to hydrocephalus (Figure 2), and he underwent ventriculoperitoneal shunt placement at age of four months. He had congenital bilateral clubfoot that was successfully treated by splinting. The ophthalmologic examination was normal. Muscle biopsy showed increased numbers of fibrous septa and ring fibers. The creatine kinase level was normal. A triplet repeat primed polymerase chain reaction (TP-PCR) analysis showed a CDM1 with 800 CTG repeats. Genetic testing of the parents showed that the mother had 125 CTG repeats.

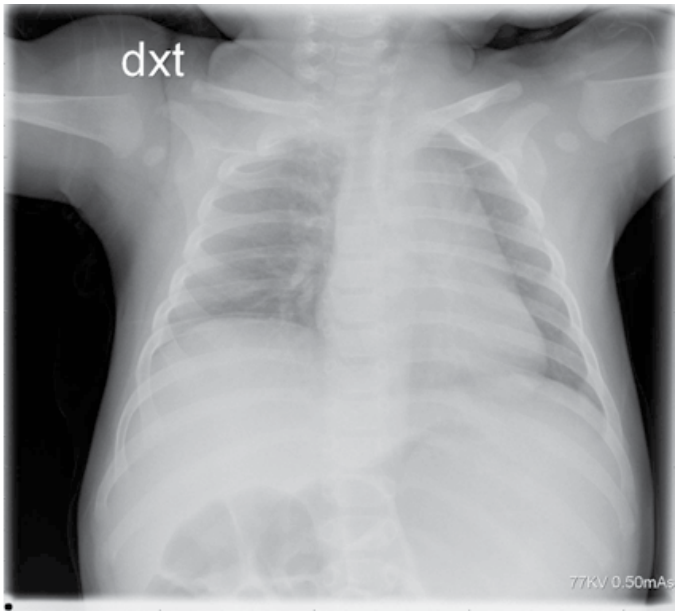

Figure 1: Chest X-ray on day-1 showing thin ribs and a raised right hemidiaphragm.

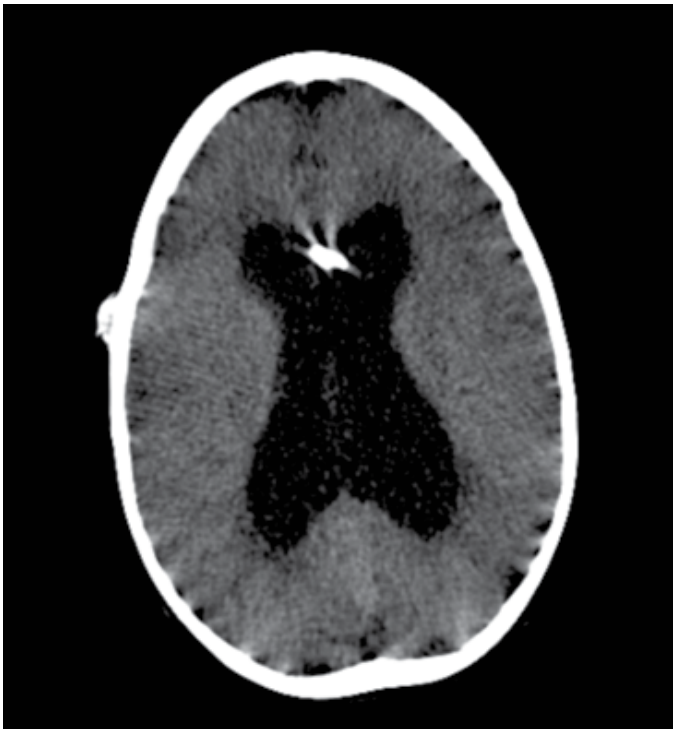
Figure 2: Computed tomography scan of the head without contrast enhancement demonstrating hydrocephalus.

The mother had muscle weakness and fatigue, but was unaware of her disease. The parents had a healthy older daughter. Genetic counseling was recommended before family planning. The mother's older sister also had mild symptoms, and TP-PCR showed 64 CTG repeats. She has a 12-year-old son without any symptoms. No other family member were tested.

Our patient was discharged from hospital at fifth month after birth. Intermittent CPAP support by night at home was continued until the age of 14 moths. Follow- up at age of 18 months showed neurodevelopmental delay. He started to crawl and was able to walk with support shortly. Physiotherapy and neurological follow-up have been proceeded.

DISCUSSION

CDM1 is associated with a poor prognosis, with an overall mortality rate of up to 50% in severely affected children [3]. The most commonly reported cause of death is respiratory failure, which occurs secondary to hypotonia and muscle weakness. Other factors associated with a lethal outcome are prematurity, low birth weight, prolonged mechanical ventilation and chylothorax [1, 4]. There is a well-established link between mechanical ventilation for more than four weeks and a lethal outcome [1, 4]. As with most severely affected neonates, our patient presented with generalized hypotonia and respiratory failure. He was resuscitated at birth and treated with mechanical ventilation for four days. This is significantly less than in previously reported cases [1, 4]. We have

speculated that the use of new techniques of mechanical ventilation resulted in fewer days on a ventilator and survival in our case.

PPHN has seldom been reported in neonates with CDM1, but contributes to the high mortality rate when present. Only three cases have been described, all with lethal outcomes during the neonatal period [5, 6]. Cantagrel et al. [6] reported a case of CDM1 with PPHN complicated by pneumothorax, pneumomediastinum and pneumopericardium, which resulted in death in the first day of life. In our case, there were no air trapping complications, which may explain respiratory stabilization and survival. Reis-Bahrami et al. [5] described two cases of CDM1 complicated by PPHN in premature neonates who were born at gestational ages of 35 and 36 weeks, respectively. They died at fourth day and ninth day, respectively, despite aggressive ventilator and pharmacological support. Neonatal death was probably determined by both CDM1 with PPHN and surfactant deficit due to prematurity. Neither of these two infants was treated by surfactant, in contrast to our case. To the best of our knowledge, this is the first reported non- fatal case of CDM1 complicated by PPHN.

Besides PPHN and respiratory compromise during infancy, our patient showed neurodevelopmental delay. He started to crawl at the age of 18 months. According to literature, surviving infants experience improvement in motor function and are usually able to walk [7]. However, affected children may develop weakness, myotonia, cardiac problems, impaired attention and autism spectrum disorder [7–9].

The diagnosis of CDM1 was confirmed by TP-PCR analysis. Molecular genetic testing of our patient showed 800 CTG repeats. On the other hand, his mother had 125 repeats and she presented with mild clinical signs. This case illustrates the well-reported phenomenon of anticipation by maternal transmission, in which the child has earlier onset of disease and more severe disease than the mother [2, 6]

All patients with CDM1 have an increased CTG repeat size in the DMPK gene. Most patients with CDM1 have > 1000 CTG repeats [2]. The severity of disease generally correlates well with the CTG repeat size and patients with a higher number of CTG repeats generally have more severe disease. Redman at al. [10] reported a few patients with CDM1 who had repeat sizes of between 730 and 1000. However, there is no data on severity of disease in these patients. Further investigation into the severity of CDM1 and CTG repeat sizes could be valuable for predicting outcomes. It is rare to find cases of CDM1 complicated by PPHN with only 800 CTG repeats in the DMPK gene.

CONCLUSION

We presented a case of congenital myotonic dystrophy type 1 (CDM1) complicated by PPHN that did not result in death during the neonatal period. Further research is needed to elucidate the relationship between PPHN and the number of CTG repeats in patients with CDM1.

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Author Contributions

Vladan Milovanov – 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

Yanko Petkov – 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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CASE REPORT PEER REVIEWED | OPEN ACCESS

A giant omental cyst mimicking ascites in a three-year-old Congolese child

Léon Tshilolo, Rudy Lukamba, Alphonse Simbi, Valentin Kazadi, Christian Mayemba, Bienvenu Lebwaze, Raphael Kalengayi

ABSTRACT

Introduction: Omental and mesenteric cysts are rare intra-abdominal lesions. They can present as an abdominal distension and make differential diagnosis with ascites. Case Report: A three-year-old boy with fever, under nutrition and abdominal distension previously diagnosed as ascites was submitted to laparotomy. We founded a large multi lobed cyst from the large omentum filling almost the entire abdominal cavity and containing sero-hemorrhagic to very dark liquid. The histopathologic examination showed a cystic formation lined by cubic mesothelium cells, with abundant inflammatory infiltrate rich in lymphocytes and histiocytes, mixed with a few giant cells of foreign body of melanotic origin. Conclusion: The diagnostic role of sonography in abdominal distension of unknown etiology is emphasized.

Keywords: Ascites, Child, Melanotic pigments, Omental cyst

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INTRODUCTION

Omental and mesenteric cysts are rare intra-abdominal lesions which may be congenital, traumatic, neoplastic or infectious in origin [1, 2]. The similarity of his clinic with abdominal masses, intra-peritoneal as extra peritoneal, as well as intra-abdominal effusions (ascites), makes the diagnosis somewhat difficult [3]. The preoperative diagnosis can be made by the combined modalities of ultrasound and CT scan [2, 4].

We report the case of a three-year-old child admitted for abdominal ascites and under nutrition who underwent to laparotomy. The role of multidisciplinary approach and diagnostic tools is emphasised.

CASE REPORT

A three-year-old boy was first admitted three months before to another hospital in Kinshasa and then referred to Monkole Hospital Centre for evaluation of abdominal distension and fever lasting for two weeks. The patient was a neglected child who was welcomed in a charity

residence in Kinshasa. No available data was known about his previous medical history. In the previous admission, he was submitted to a malaria treatment and a paracentesis was performed revealing an apparent exsudative fluid. No biological data were available.

On physical examination, the child was undernourished, pale and febrile (39°C). The heart beats were rapid at 140 beats/min without abnormal murmur; respiratory rate varied from 35–40 cycles/min and the pulmonary sounds were reduced at the bottom. The abdomen was very protuberant and scarcely tender, with a positive fluid thrill and a vaguely "no well-defined" mass in the right hypochondria and pubic region. The other abdominal organs (liver and spleen) were not easy to appreciate. A venous collateral circulation and tattoos were noted on the abdominal wall. The abdomen circumference measured 57 cm (Figure 1).

Significant biological parameters at admission were: white blood cell count 13,000/mm^3 (N49 L49 M0 E2), hemoglobin 9.8 g/dl, hematocrit 26.9%, MCV 70 fl; platelets 579,000/mm^3. ESR 118/h. Quick time: 80% and INR1.2. Rapid HIV test and tuberculosis test were negative. Blood urea, electrolytes and liver and kidney function studies were within normal limits.

Chest and abdominal X-ray showed no lesions in the lung parenchyma, but more horizontally ribs and more upper right diaphragm. Abdomen was fulfilled by an opaque homogenous mass with concomitant displacement of the intestines to posterior and upper area. No calcification was observed (Figure 2).

Abdominal ultrasound revealed the presence of a large cystic mass with multiple septa and mobile internal echoes (brown movements) from epigastrium to pelvis (Figure 3).

All the solid organs (liver, spleen, kidneys and pancreas) were normal but displaced by the mass. There was no ascites. Diagnosis of "mesothelium tumoral mass" or "peritoneal myxoma" was evoked.

A selective laparotomy was then indicated. The exploration of the abdominal cavity revealed a solid and multiseptated cystic mass originated from the large omentum, adjacent to the liver (Figure 4). The content of the mass was a liquid variable from sera aspect in one cavity and to very dark aspect in the others.

The mass was completely excised and the postoperative course was uneventful. There has been no recurrence till now after five years of the laparotomy.

Histological examination showed a fibrous tissue having the shape of the wall of a tortuous cystic. Some fragments were the seat of a nonspecific chronic inflammation with the presence of giant cells of foreign body. There was no presence of cartilage tissue in the fragments or malignancy or any neoplasm. The overall look was pleading for an inflamed and remodeled cystic mass whose nature could be an inflamed cyst enterogene and reworked or other congenital hamartoma cyst kind overhauled.

Proofreading blades noted cystic formation lined by cubic mesothelium cells, sometimes flattened, and supported by a fibro-fatty tissue that was the seat of vascular congestion with hemorrhagic suffusion. No significant lymphatic vessels proliferation has been observed. Abundant inflammatory infiltrate rich in lymphocytes and histiocytes was also present, mixed with a few giant cells of foreign body, next to brown-black deposits reminiscent of melanin pigments. The special coloration Fontana confirmed the nature of the melanin pigments (Figure 5). The Perls coloration was negative, excluding the ferric deposits. Diagnosis of inflamed melanotic peritoneal cyst was selected.

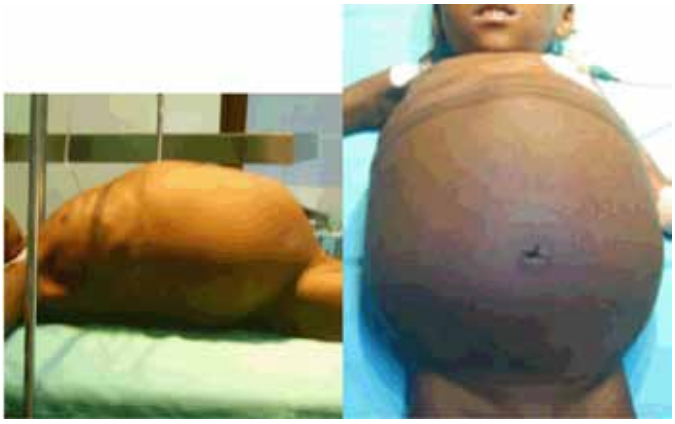

Figure 1: Important abdominal distension (profile and face view) in a three-year-old child with a general state altered by emaciation.

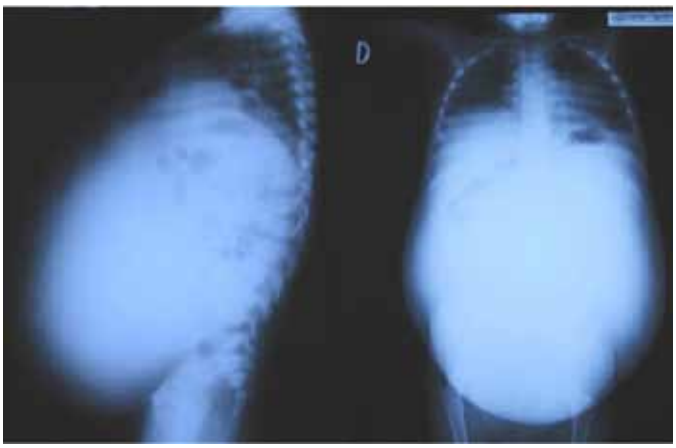

Figure 2: Chest and abdominal X-rays revealed an opaque homogenous mass with displacement of the intestines to posterior and upper area.

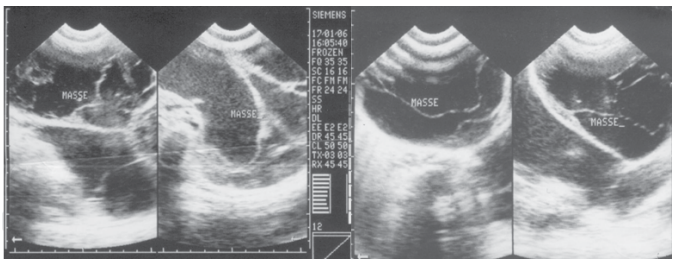

Figure 3: Abdominal Ultrasound dysplaied the presence of a large cystic mass with multiple septa and liquid with brown movements inside.

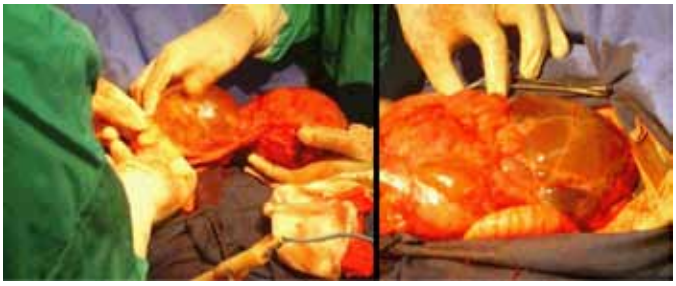

Figure 4: Multicystic mass removed from large omentum and containing liquid varying from sera to dark aspect.

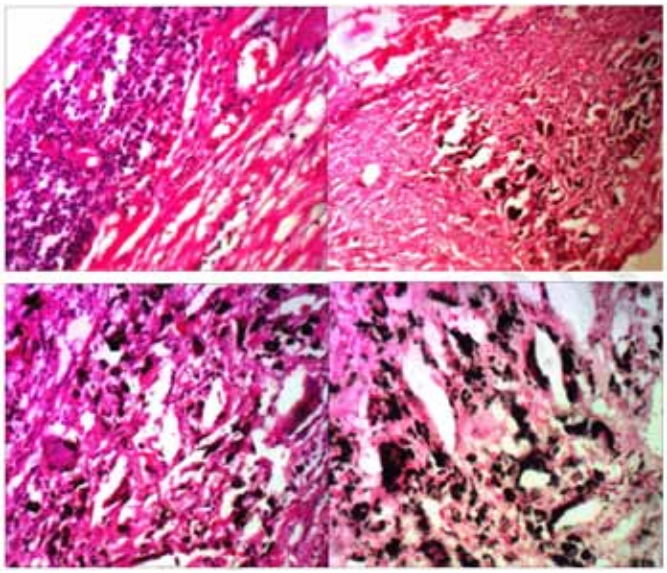

Figure 5: (A) Fibrous cystic wall lined with mesothelium and invested by an inflammatory infiltrate polymorphic (H&E stain, x100), (B) Brown-blackish deposits recalling melanin (H&E stain, x100), (C) Foreign body giant cell (H&E stain, x400), (D) Dark staining of melanin pigments (Fontana, x40).

DISCUSSION

Mesenteric, omental, and retroperitoneal cysts are unusual causes of intra-abdominal masses in childhood. Mesenteric cysts in children are mostly lymphatic malformations of mesentery or omentum origin and clinical presentation varies from abdominal distension, chronic abdominal pain, subocclusion to sudden onset of peritonitis or volvulus [2, 5].

Different developmental theories for these cysts are discussed, including the benign proliferation of the ectopic mesenteric lymphatics that do not communicate with the rest of the lymphatic system, the failure to join the embryonic lymphatic spaces to the venous system, the failure of the fusion of mesenteric leaves, the deficiency of the normal lymphaticovenous shunts, trauma, neoplasm, and the localized degeneration of lymph nodes [2, 5].

Histologically, the nature of the cyst is determined according to the type of epithelium that lines the inner layer of the wall. Lymphangiomas are the most common cause of mesenteric and omental cysts comprising 90% of cases and the remaining cysts are mesothelial cysts.

Thus, the peritoneal cysts are divided into several types and their origin is not currently well understood [6]. Melanotic peritoneal cyst multilocular has been described as a particular entity of peritoneal cysts which likely represent a peritoneal inclusion cyst related to chronic inflammation [7].

The pseudomelanosis coli is another possibility that could explain the presence of melanin pigments in the cyst wall. It is a benign condition due to the accumulation of blackish-brown pigment lipofuscinic kind in the lamina propria of the colon, with the possibility of migration [8]. It is often due to the abuse of laxatives such as anthraquinone derivatives. The brown-black deposits correspond to cellular apoptotic bodies phagocytized by intraepithelial macrophages migrate to the lamina propria after being transformed into lipofuscinic pigments by lysosomes [9].

Our patient had a giant cyst compressing the intestines with a possible constipation, but with no history of abuse of laxatives. The presence of hemorrhagic fluid has been described in omental cysts but only few cases reported the histological description. In this case report, the Perls coloration was negative for the ferric deposits and the presence of melanin pigments in the cyst wall has no clear explanation.

The presence of lipofuscin pigments would be an event that would complicate a cyst whose genesis is not clearly defined.

The clinical history of abdominal distension with absence of any well demarcated mass can easily make the diagnosis of ascites as described in previous case reports [3, 5, 10].

CONCLUSION

A multidisciplinary approach involving pediatricians, radiologists and surgeons will be encouraged in order to avoid abusive paracentesis in children with abdominal distention mimicking ascites. Clinical diagnosis of omental cyst is not evident and required the ultrasound exploration before the laparoscopic excision.

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Author Contributions

Léon Tshilolo – 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

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

Alphonse Simbi – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published
Valentin Kazadi – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published
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CASE REPORT PEER REVIEWED | OPEN ACCESS

Vancomycin-induced bullous dermatosis

Martin Minwoo Kim, Katherine Baquerizo, Pranay Srivastava, Deepthi Lankalapalli, Asmat Ullah

ABSTRACT

Introduction: Linear IgA bullous dermatosis (LABD) is a rare mucocutaneous immune mediated blistering skin disease seen in various countries that have ranged from less than 0.5 to 2.3 cases per million individuals per year. The presentation can be similar to other bullous dermatoses, yet it has distinctive clinicopathologic and immunologic features that allow prompt recognition and treatment with complete resolution. Case Report: A 54-year-old obese Caucasian male with past medical history of atrial fibrillation on warfarin, hypertension, gastroesophageal reflux disease, benign prostatic hyperplasia, and dyslipidemia presented to the emergency department complaining of a generalized blistering rash that initially surrounded the genitalia a week after being discharged from the hospital following a mechanical fall. All medications were reviewed and skin biopsy was taken. He developed the drug-induced variant of LABD to vancomycin with mucosal involvement and compare the resemblance to other autoimmune blistering diseases such as toxic epidermal necrolysis (TEN) and Stevens–Johnson syndrome. This case demonstrates complete resolution of the disease with prompt identification of the underlying disease process based on the clinical and immunohistochemistry findings. Conclusion: Linear IgA bullous dermatosis can be difficult to diagnose as it presents similar to other bullous dermatoses. The problem of differential diagnosis coupled with clinicopathologic and immunologic features of LABD are emphasized to recognize this distinct disease.

Keywords: Bullous, Vancomycin, Dermatosis, Linear IgA, Nikolsky's sign

How to cite this article

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INTRODUCTION

Linear IgA bullous dermatosis (LABD) is a rare mucocutaneous immune mediated blistering skin disease that is characterized by subepidermal blistering and a homogenous linear deposition of IgA basement membrane antibodies along the cutaneous basement membrane [1]. The disease may occur spontaneously or arise from a drug-induced reaction, most commonly to vancomycin.

Reports of disease incidence from various countries have ranged from less than 0.5 to 2.3 cases per million individuals per year, with the first case of drug-induced LABD being described in 1981 [2, 3].

We present the case of a 54-year-old male who developed the drug-induced variant of LABD to Vancomycin with mucosal involvement and compare the resemblance to other autoimmune blistering diseases such as toxic epidermal necrolysis (TEN) and Stevens–Johnson syndrome. This case demonstrates complete resolution of the disease with prompt identification of the underlying disease process based on the clinical and immunohistochemistry findings.

CASE REPORT

A 54-year-old obese Caucasian male with past medical history of atrial fibrillation on warfarin, hypertension, gastroesophageal reflux disease, benign prostatic hyperplasia, and dyslipidemia presented to the emergency department complaining of a generalized blistering rash that initially surrounded the genitalia a week after being discharged from the hospital following a mechanical fall. During the prior hospitalization, lower leg cellulitis and an infected stage 3 sacral decubitus ulcer were noted, and the patient was started on intravenous vancomycin twice daily as well as silvadene wound dressings. After stabilization, the patient was discharged after a 10-day hospitalization course with a prescription for a course of oral clindamycin.

On readmission to the emergency department seven days after discharge, the patient complained of a sudden cutaneous blistering rash that began 48 hours ago. The patient stated that he noticed blisters and erythema on his lower abdomen that spread to both palms seven his oral cavity. Lesions were both pruritic and painful. The patient denied any systemic symptoms and review of symptoms was non-contributory. The patient also denied any travel, insect bites, or changes in any skin care products, detergents, soaps, and shampoos.

Physical examination revealed general toxic appearance, dry mucous membranes with 1–2 cm erosions in the oral cavity as well as crusted lesions on his lips with bilateral conjunctival injections. Numerous symmetric clustered 5 mm to 2 cm clear, smooth vesicles and tense bullae were widespread over the trunk, perineum, upper and lower extremities, and genitalia with Nikolsky's sign. Annular macules, and papules with surrounding erythematous base with urticarial plaques and excoriation on the chest seen (Figure 1). Most of the bullae had a surrounding erythematous base, and on the trunk were associated with annular macules, and papules; the hands showed additionally widespread crusted erosions. The borders of the lesions were well demarcated. Some lesions had a targetoid appearance. Erythematous, clear, smooth vesicles and tense bullae were widespread over the genitalia region (Figure 2). Nails and hair were unremarkable. Vital signs were within normal range. Gram stain and culture of bullae fluid showed no growth. Complete blood count (CBC) and complete metabolic panel (CMP) were unremarkable, except for chronic anemia with hemoglobin of 6.7 g/dL (baseline 8.1–8.3 g/dL) in which he received blood transfusions, and is unrelated to the disease.

Two 6-mm punch biopsy were obtained from 2 separate lesions that demonstrated mixed superficial perivascular and interstitial dermatitis including many neutrophils. The dermoepidermal junction showed vacuolization of basal keratinocytes. Numerous clustered 5 mm to 2 cm clear smooth vesicles and tense bullae on the palmar surface with no nail involvement appreciated (Figure 3). The dermoepidermal junction of the lesion showed vacuolization of basal keratinocytes (Figure 4). Visualization under direct immunofluorescence (DIF) showed linear IgA deposit in the basement membrane zone (Figure 5).

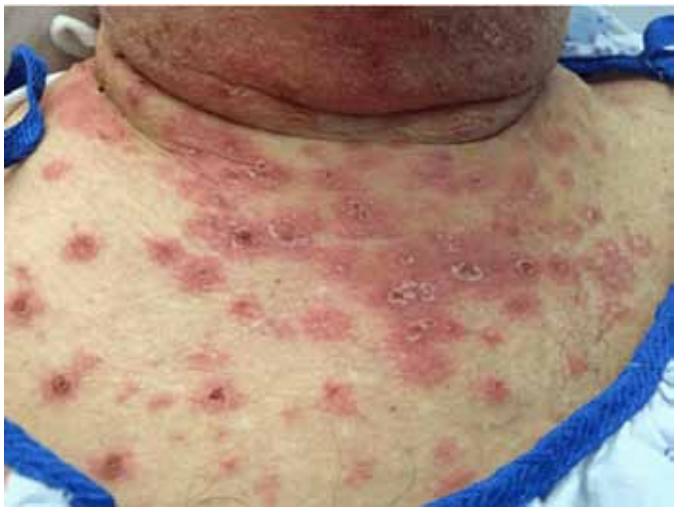

Figure 1: Annular macules, and papules with surrounding erythematous base urticarial plaques and excoriation on the chest.

Figure 2: Erythematous, clear, smooth vesicles and tense bullae were widespread over the genitalia region.

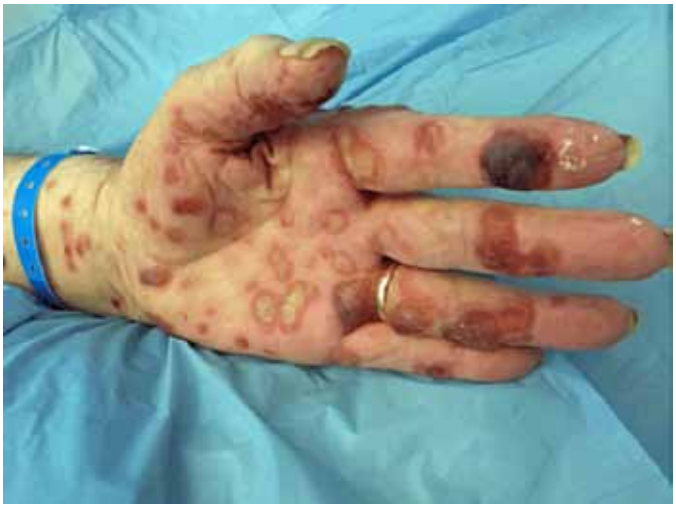

Figure 3: Numerous clustered 5 mm to 2 cm clear smooth vesicles and tense bullae on the palmar surface with no nail involvement.

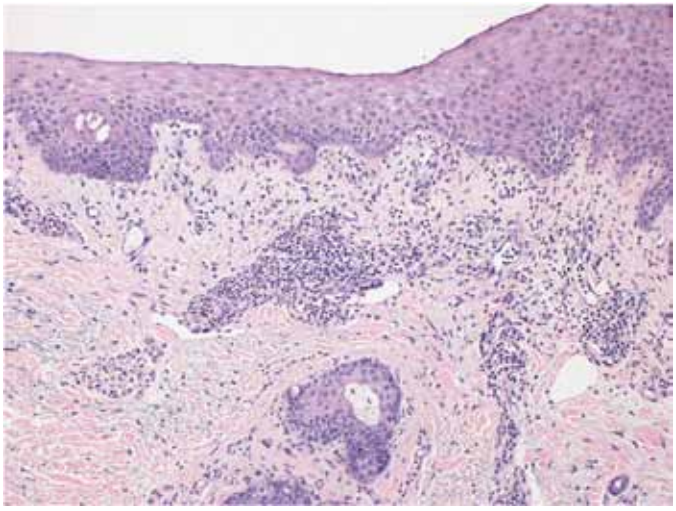

Figure 4: The dermo-epidermal junction of the lesion showing vacuolization of basal keratinocytes (H&E stain, x100).

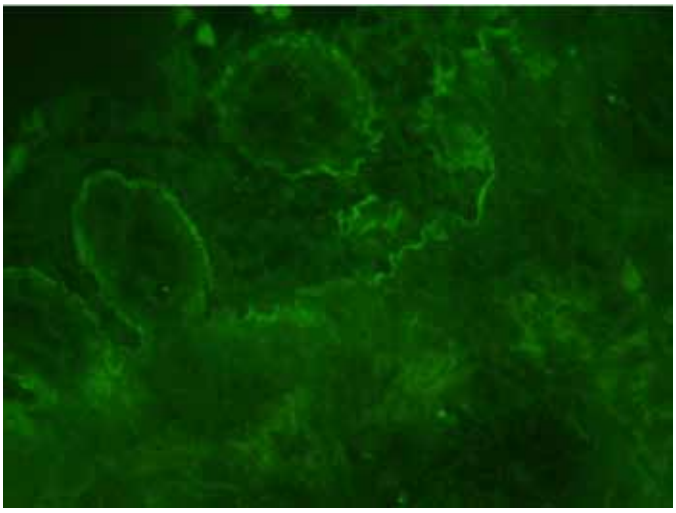

Figure 5: Visualization under direct immunofluorescent microscopy (DIF) showed linear IgA deposit in the basement membrane zone.

The constellation of patient history, physical findings and histological features pointed towards a diagnosis of Vancomycin-induced Linear IgA Bullous Dermatosis from the IV vancomycin he received during his previous hospitalization. At 72 hours after the onset, the extent of the rash peaked covering approximately 50% of his body. The patient was in good general condition with frequent monitoring of his vital signs and metabolic panel. Four days after the onset of the rash no new lesions had developed and the lesions gradually resolved spontaneously after two weeks without scarring. The patient was instructed to avoid vancomycin in the future.

DISCUSSION

The interest of this case report is to highlight the similarities and differences of LABD to other autoimmune blistering diseases. Furthermore, the correct diagnosis is made using the clinical presentation as well as immune histological features.

Linear IgA Bullous Dermatosis (LABD) is a rare yet distinct mucocutaneous immune mediated blistering disease that is characterized by a homogenous linear deposition of IgA antibodies along the cutaneous basement membrane. It has a bimodal peak of onset, the first in early childhood and the second in older individuals [1]. In the past LABD was considered as a variant presentation of dermatitis herpetiformis, however it is now differentiated as a separate condition [4, 5]. Commonly the disease arises spontaneously, but a drug-induced variant, most classically from vancomycin is frequently implicated [6, 7]. Other drugs that may be linked to LABD that have been reported include a variety of antibiotics, non-steroidal anti-inflammatory agents (e.g., diclofenac, naproxen, piroxicam), lithium, captopril, amiodarone, phenytoin, cyclosporine, furosemide, interferon alpha, and somatostatin [8–12]. As in our patient, vancomycin was the most likely culprit, though direct cause was not determined. Although multiple case reports have documented drug exposure as a precipitating factor, formal studies validating the existence of drug-induced LABD are lacking [13]. Vancomycin and phenytoin have both been reported to induce LABD with vancomycin being the pharmacologic agent most frequently reported as a potential inciting factor [13]. Vancomycin has also been reported to cause localized LABD confined to the palms at supratherapeutic levels [14].

In adults, LABD can present with a variety of skin manifestations ranging from vesicles resembling dermatitis herpetiformis (DH) to bullae mimicking bullous pemphigoid and rarely toxic epidermal necrolysis (TEN) [2]. In our case, the differential diagnosis for this patient was erythema multiforme, Stevens Johnson, due to the targetoid features and mucosal involvement. Other conditions considered were toxic epidermal necrolysis (TEN) syndrome, bullous impetigo, bullous pemphigoid,

and pemphigus vulgaris. Clinical findings in LABD patients may be difficult to differentiate from those with vesiculobullous dermatitis. Onset of the primary lesions is frequently accompanied with pruritus or a burning sensation. Grouped vesicles, bulla and papules appear in combinations over the trunk, limbs and buttocks as in our patient. Some patients with LABD may have larger sized bulla and maybe mistaken for bullous pemphigoid [14]. A distinctive annular and "string of pearls" grouping of blisters are common. Drug induced LABD may have a findings similar to that of erythema multiforme and TEN [14]. Mucosal involvement is another manifestation seen in patients with LABD. A large majority, as many as 70% [6] have varying degrees of oropharyngeal ulcerations and erosions. Conjunctival involvement has also been noted [6]. Our patient showed genital and oral erosions which can be distinguished with DH. Furthermore, the majority of patients with LABD lacked the villous atrophy and the antibodies against tissue transglutaminase seen in DH [15, 16]. Oral lesions are seen in 10–30% of patients with bullous pemphigoid. Toxic epidermal necrolysis (TEN) involves detachment of >30% of the body surface area with mucous membranes are also involved in over 90% of cases. Stevens–Johnson syndrome characterized by skin detachment in <10% of the body surface. Mucous membranes are affected in over 90% of patients [8]. Also, drug-induced LABD had a more severe presentation than the spontaneous form with Nikolsky sign and large erosions mimicking toxic epidermal necrolysis and other bullous diseases making Nikolsky sign insignificant. DIF assay was recommended for all patients with Nikolsky sign and large erosions [13].

The defining feature of LAD is the presence of homogenous linear band of IgA at the dermal-epidermal junction, however there may also be deposits IgG, IgM, and the third constituent of complement (C3) [6]. Histopathology shows subepidermal bulla containing neutrophils along the basement membrane and near the tips of the papilla where they sometimes form micro-abscesses [7]. Lymphocytes and eosinophils may also be present, however the major component is the neutrophils [13]. Blister formation is usually seen in the lamina lucida or the sublamina densa locations [14, 15].

The majority of patients with classic linear IgA disease respond to oral dapsone or sulfapyridine. Oral prednisone may also be used in order to decrease formation of blisters [6]. Other medications that have been reported successful are: trimethoprim/sulfamethoxazole, mycophenolate mofetil, dicloxacillin and erythromycin [17–19]. When dapsone is unsuccessful or steroid sparing agents are needed mycophenolate mofetil, IVIG, and azathioprine can be used. Unlike classic LAD, which is chronic and recurring, the drug-induced variant is self-limited and typically resolves after discontinuation of the offending agent, most of the time without adjuvant treatment as was seen in our case.

CONCLUSION

Epidemiologically, reports of disease incidence from various countries have ranged from less than 0.5 to 2.3 cases per million individuals per year. Despite the broad use of vancomycin in the hospital setting, clinicians are mostly unaware of potential severe skin reactions, such is the case of vancomycin-induced bullous dermatosis. It is critical to keep linear IgA bullous dermatosis (LABD) in the differential diagnosis of patients presenting with vesiculobullous dermatitis.

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Martin Minwoo Kim – 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
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Conflict of Interest

Authors declare no conflict of interest.

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CASE REPORT PEER REVIEWED | OPEN ACCESS

Onlay mesh repair for spontaneous lumbar hernia: A case report

Biswal Jayanta Kumar, Mahapatra Tanmaya, Guria Sourabh, Supreet Kumar, Meher Dibyasingh, Kanhat Karesh Samu

ABSTRACT

Introduction: Lumbar hernia is a rare hernia. It herniates through the superior lumbar triangle (Grynfeltt-Lesshaft triangle) or inferior lumbar triangle (Petit triangle). It can be classified as congenital or acquired, which may be primary or secondary. Case Report: A 70-year-old male with a reducible left sided superior lumbar hernia. Intraoperatively, there was a small defect and it was repaired with primary closure and an onlay meshplasty. The patient was absolutely asymptomatic during the follow-up. Conclusion: Though there are many techniques for repair of lumbar hernia, in case of a small defect, a primary repair with a tension free onlay meshplasty can be a quick procedure with good result.

Keywords: Flank swelling, Grynfeltt-Lesshaft triangle, Inferior lumbar triangle, Lumbar hernia, Onlay mesh repair, Superior lumbar triangle

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INTRODUCTION

Lumbar hernia is one of the rarest forms of abdominal wall hernias with only about 300 cases of primary lumbar hernias being reported over last four centuries [1]. First described by Barbette in 1672, the existence of this variant of hernia is known for four centuries and the first case was reported by Garangoet in 1731 [2].

CASE REPORT

A 70-year-old male patient presented with gradually increasing left flank swelling of duration one and half years (Figure 1A). Over the past six months there was local discomfort and dull pain. There was no history of trauma, previous surgery, hematuria or altered bowel habits. On examination there was a globular, soft, non-tender, reducible, non-pulsatile swelling of size 7×5 cm located in the left superior lumbar triangle with an expansile cough impulse. The swelling was more prominent with straining or a standard Valsalva maneuver and disappearing with

prone position The examination of rest of the abdomen, right flank, back & hernia orifices were normal.

Ultrasonography revealed dehiscence of abdominal wall of size 15 mm in the left lumbar region. So a diagnosis of left sided reducible lumbar hernia was made on the basis of clinical and radiological finding and the patient was taken up for surgery.

The patient was placed in modified right lateral decubitus position. Oblique left lumbar incision was given. The hernia sac was identified and opened. The content being the extra peritoneal fat was partially excised and the rest was reduced (Figure 2). The size of the defect was about 1.5 cm (Figure 3) which was primarily repaired with (1-0) polypropylene interrupted suture (Figure 4) and then an onlay polypropylene mesh was given over the defect (Figure 5). The skin was closed with a negative suction drain. The postoperative periods were uneventful and the patient was discharged on 4th postoperative day on analgesics.

During the follow-up visit after five months the operation scar was found to have healed well and the patient was absolutely asymptomatic (Figure 1B).

DISCUSSION

The lumbar region is bordered by the twelfth rib superiorly, the iliac crest inferiorly, the erector spinae muscles of the back posteriorly, and a vertical line between the anterior tip of the twelfth rib and the iliac crest anteriorly. The region contains two anatomic triangles, through which the rare lumbar hernia can form. The inferior lumbar triangle of Petit is the more common of the two. Its anterior border is the posterior edge of the external oblique muscle, the posterior border is the anterior extent of the latissimus dorsi muscle, and the inferior border is the iliac crest. Lumbar triangle of Grynfeltt, also known as the superior lumbar triangle is bounded by the twelfth rib and the serratus posterior inferior muscle, the posterior border of the internal oblique muscle, and by the quadratus lumborum and erector spinae muscles posteriorly. The floor of the superior triangle is composed of transversalis fascia and the entire triangular space is covered posteriorly by the latissimus dorsi muscle [3].

Lumbar hernias have been classified as congenital (20%) or acquired (80%). An acquired hernia may be primary or secondary. Secondary lumbar hernias are of traumatic or post-surgical (flank incisions, renal surgery, iliac bone harvesting) etiology comprising about 25% of acquired hernias [4]. Another way of classifying lumbar hernia is on the basis of content, they are of two types: extraperitoneal hernia with no sac, containing only fat or sliding retroperitoneal organs (paraperitoneal), and peritoneal hernia that may include intraperitoneal organs such as small bowel, omentum, ovary and stomach [5].

In most of the times the patients are usually asymptomatic, but sometimes may complain of backache, flank pain or a dragging sensation. These hernias are known to have painless progressive enlargement in size [6]. The differential diagnosis includes lipoma, soft tissue tumors, hematoma or an abscess. In obese patients detection of a mass is usually difficult. It is observed that incidences of bowel incarceration may occur in 25% but strangulation is rare because of wide hernial neck [7].

Ultrasonography or computed tomography (CT) imaging are usually obtained in patients with suspected

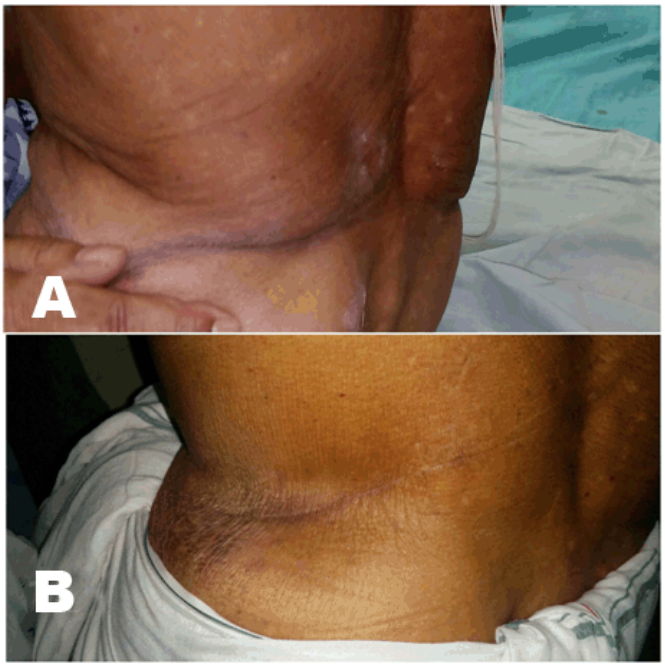

Figure 1: (A) Preoperative photo of the patient with a swelling in the left lumbar area, (B) Postoperative photograph of the patient showing healed surgical scar.

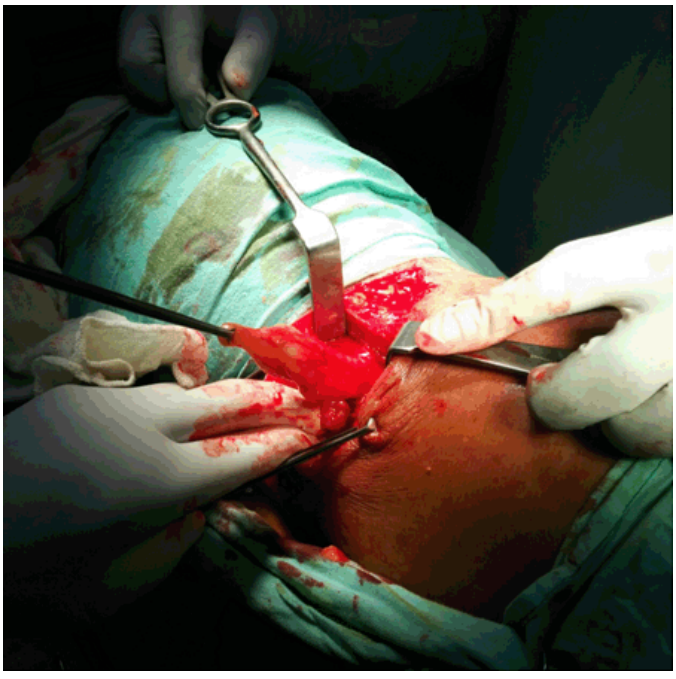

Figure 2: Herniated extra peritoneal fat through the parietal defect.

lumbar hernia to confirm the diagnosis [3]. Computed tomography scan is the diagnostic modality of choice. Computed tomography scan is able to delineate muscular and fascial layers, a defect in one or more of these layers, and the presence of herniated fat and/or viscera [4].

Surgical repair is the treatment of choice. Repair can be done either open or endoscopically. In open technique, after reduction of the contents, if the sac is found to be narrow, exploration with ligation of the sac is warranted.

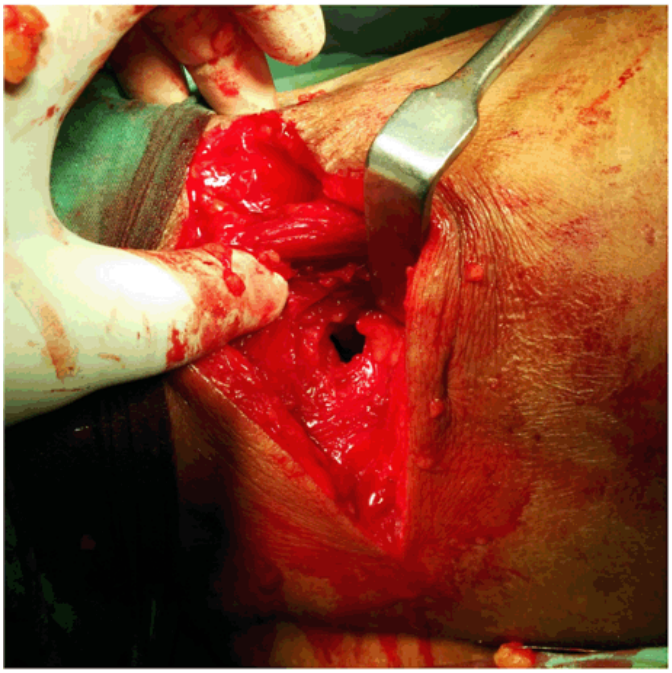

Figure 3: The primary defect of size 1.5 cm in the left lumbar region.

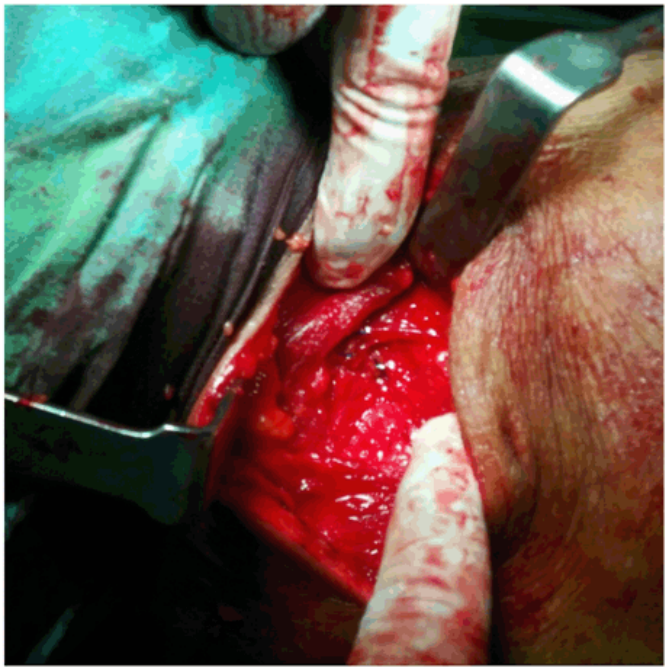

Figure 4: Primary closure of the defect with polypropylene interrupted suture.

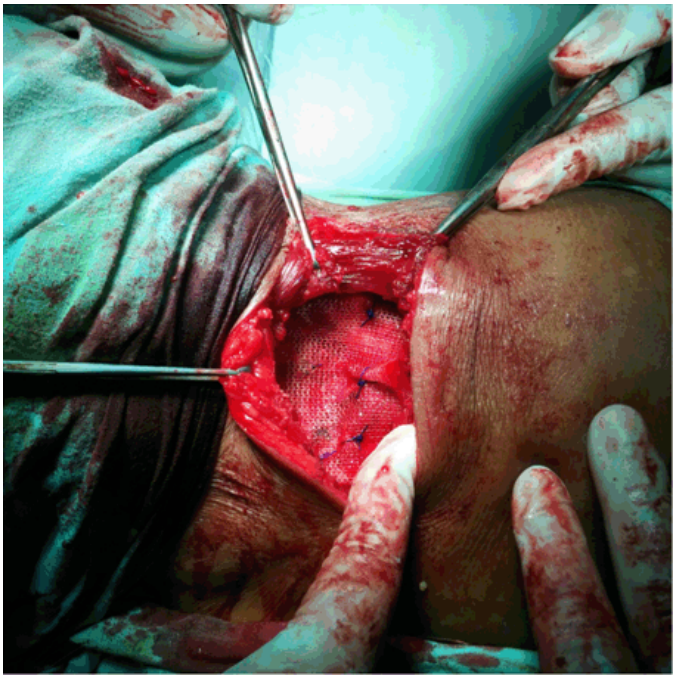

Figure 5: Onlay meshplasty after closure of the primary defect.

A sac with wide neck can be inverted and plicated. The defect is managed according to its size and the status of tissues around it: one of the strategies is to suture it primarily with interrupted heavy non-absorbable sutures. For larger defects some authors postulate the use of tensor fascia lata rotational flaps or free fascial grafts. Nowadays in the era of meshplasty a non-absorbable mesh is usually preferred for reconstruction. Depending on the size of the defect, it can be placed as an onlay, inlay or underlay [8]. The onlay technique involves primary closure of the fascia defect and placement of a mesh over the anterior fascia. Inlay repair involves securing the mesh to the fascial edge without overlap, whereas underlay repair involves placing the mesh below the fascial components. In our case, as the defect was small, it was primarily repaired with non-absorbable interrupted suture and to strengthen it an onlay tension free meshplasty was done with a polypropylene mesh. Recently, minimally invasive approaches to repair of lumbar hernias have been reported. These involve either intraperitoneal laparoscopy necessitating takedown of the lateral peritoneal reflection of the colon to facilitate exposure of the hernia defect [9], or retroperitoneoscopy in which the lateral retroperitoneal space is entered and insufflated [10].

CONCLUSION

Primary lumbar hernia is a rare clinical entity and needs a high index of suspicion during day to day practice. A good history, general physical and radiological

examination can rule out most of the differential diagnoses. Strengthening the defect can be done with synthetic mesh either open or endoscopic approach. With a small defect, open approach with primary closure and onlay tension free mesh repair can be a quick procedure with good result.

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Author Contributions

Jayanta Kumar Biswal – 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
Tanmaya Mahapatra – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published
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Kanhat Karesh Samu – Acquisition of data, Drafting 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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LETTERS TO THE EDITOR PEER REVIEWED | OPEN ACCESS

Oral amoebiasis with acute myeloblastic leukemia

Eiji Mitate, Kazunari Oobu, Tamotsu Kiyoshima, Seiji Nakamura

To the Editors,

Oral manifestations are frequent seen in immunocompromised patients. In some cases, *Entamoeba gingivalis, was* isolated and described by Gros G. [1], is found in periodontal disease [2], under chemotherapy [3], HIV(+)/AIDS patients [4] and acute osteomyelitis [5]. To our knowledge, this is the first case report of oral amoebiasis in immunocompromised patients with acute myeloblastic leukemia (AML). A 63-year-old male had painful refractory palatal and lower gingival ulcers (Figure 1A–B). Each ulcer was filled with necrotic tissue and had a 5-mm induration. Some large and enhanced cervical lymph nodes were detected on computed tomography (Figure C) scan. Brush cytological examination revealed oral amoebiasis without malignancy (Figure D). Vitamin B12 and folic acid were administrated for anemia. Blood examination revealed the following values: white blood cells 990/μl, red blood cells $1.32x10^6$/μl; and hemoglobin 4.8 g/dl, with higher hemoglobin F level. The patient was referred to the hematology department, where he was diagnosed as having AML (M2) after bone marrow biopsy. Remission induction treatment (idarubicin hydrochloride and cytarabine) was initiated. Aspergillus pneumonia, bacteremia in trichosporonosis, and tuberculous cervical lymphadenitis were also treated. After two months of remission, agranulocytosis and recurring AML were detected. During chemotherapy for AML, the patient died of trichosporonosis-related pneumonia. *E. gingivalis* is rare in healthy gingival pockets [5]. The presence of *E. gingivalis* may suggest some immune deficiency. In this case, detection of *E. gingivalis* by brush cytology and blood examination revealed immune deficiency. If malignancy or immune deficiency is expected, these two methods are considerable in early stage.

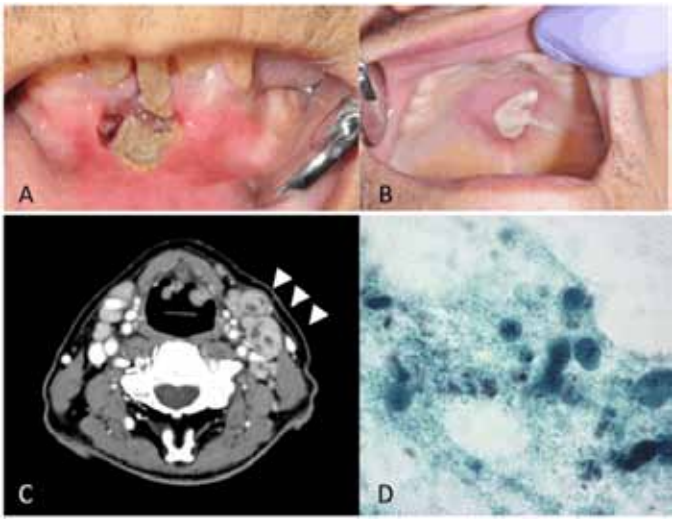

Figure 1: Oral ulcers are seen in lower gingiva (A) and palate (B) at first visit, (C) Cervical lymph nodes with rim enhance are seen (C, arrowheads) in computed tomography, (D) Brush cytological examination of ulcers showing amoebiasis.

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Conflict of Interest

Authors declare no conflict of interest.

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