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Contents

Case Report

Journal of Emergency Medicine Case Reports

Case Report: Fournier Gangrene in a Diabetic Patient Using Dapagliflozin

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Abstract

Fournier gangrene is a rare but severe necrotizing infection of the perineal region, requiring immediate medical attention. Sodium-glucose co-transporter 2 (SGLT-2) inhibitors, while effective in managing diabetes and providing cardiovascular benefits, have been linked to rare but serious adverse effects. In 2018, regulatory warnings highlighted a potential connection between these drugs and Fournier gangrene, though reported cases remain scarce. This report details the case of a 59-year-old diabetic female who developed Fournier gangrene during dapagliflozin treatment. The patient presented with extensive perineal necrosis necessitating urgent surgical debridement and initiation of broad-spectrum antibiotics. Microbiological analysis identified Escherichia coli as causative pathogen. Intensive wound management and insulin adjustments led to significant clinical improvement, and the patient was discharged after 24 days. The possible link between SGLT-2 inhibitors and Fournier gangrene remains under investigation. While glucosuria induced by these medications may heighten the risk of infections, the presence of comorbidities such as diabetes and immunosuppression are likely contributing factors. This case emphasizes the complexity of determining causality. Clinicians prescribing SGLT-2 inhibitors should be vigilant for signs of rare infections like Fournier gangrene, especially in patients with multiple risk factors. Prompt diagnosis and aggressive treatment are critical for patient recovery.

Keywords: Case report, dapagliflozin, fournier gangrene, sodium-glucose cotransporter-2 inhibitors

Introduction

Sodium-glucose co-transporter 2 (SGLT-2) inhibitors, including dapagliflozin, have emerged as a cornerstone in the management of type 2 diabetes mellitus (T2DM), offering a range of benefits that extend beyond glycemic control. These agents act by inhibiting the sodium-glucose co-transporter 2 proteins located in the proximal renal tubules, which are responsible for the reabsorption of filtered glucose back into the bloodstream. By blocking this pathway, SGLT-2 inhibitors enhance the urinary excretion of glucose, resulting in reduced plasma glucose levels (1). This multifaceted action not only lowers blood glucose levels but also contributes to significant cardiorenal protection, making SGLT2i valuable in treating both diabetic and non-diabetic patients (2, 3)

Despite their benefits, SGLT-2 inhibitors are not without risks. Rare but serious adverse effects have been reported, including euglycemic diabetic ketoacidosis (DKA), urinary tract infections, and necrotizing fasciitis of the perineum, known as Fournier's gangrene (FG) (4). FG is a rare but life-threatening form of necrotizing fasciitis that affects the external genitalia and perineal region. It is most commonly associated with diabetes mellitus, immunosuppressive conditions, and poor hygiene, all of which may predispose individuals to severe soft tissue infections (1, 5).

The link between SGLT-2 inhibitors and FG was first noted in post-marketing surveillance and case reports, prompting regulatory bodies to issue warnings about this potential association. Although the absolute risk is low, the condition's severity necessitates prompt recognition and management. This case report presents the clinical course of a 59-year-old diabetic patient who developed Fournier's gangrene while on dapagliflozin therapy. The report aims to explore the potential contributory role of SGLT-2 inhibitors in the pathogenesis of FG and to discuss clinical outcomes, emphasizing the importance of vigilance in patients receiving these medications.

Case Report

A 59-year-old female with a history of type 2 diabetes mellitus (T2DM), hypertension (HT), chronic kidney disease (CKD), and coronary artery disease (CAD) presented to the emergency department with complaints of discharge and swelling near the perianal area, painful pubic swelling, and chills. One week prior, the patient had undergone drainage of a perianal abscess,

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followed by the formation of a pubic abscess in the hair-bearing region. The patient attempted self-drainage, which led to worsening symptoms, including fever and chills. Her past history was significant for obesity (BMI 36 kg/m^2). There is no history of smoking in the patient's medical background.

The patient was using the following medications for diabetes at home: insulin aspart (22 units twice daily), insulin glargine (40 units once daily), linagliptin (5 mg once daily), and dapagliflozin (10 mg once daily).

On examination, the patient appeared critically ill with signs of septic shock, including tachycardia and hypotension. Physical examination revealed a large, fluctuant, necrotic abscess extending from the perineum to the mons pubis and gluteal region, consistent with Fournier gangrene.

The patient's HbA1c was 7.73%, with an estimated average glucose of 175.15 mg/dL, indicating suboptimal glucose control. Laboratory Risk Indicator for Necrotizing Fasciitis (LRINEC) score was calculated to assess the risk of necrotizing soft tissue infection (6). Based on laboratory values presented in Table-1, including CRP (320 mg/dL), WBC (35.49 x10^9/L), hemoglobin (7.5 g/dL), sodium (129 mmol/L), creatinine (5.14 mg/dL), and glucose (175.15 mg/dL), the patient's LRINEC score was determined to be 12, placing the patient in the high-risk category, indicating a probability of necrotizing fasciitis greater than 75%.

Upon admission, the patient was started on intravenous antibiotics, including imipenem-cilastatin (500 mg/500 mg twice daily) and teicoplanin (600 mg loading dose followed by 600 mg every 72 hours), based on infectious disease consultation. Emergent surgical debridement was performed under spinal anesthesia. The surgical site was prepared with sterile drapes and a sterile urinary catheter was inserted. A large abscess was identified from the mons pubis, extending into the gluteal region, including a fistula between the gluteal and perineal regions, as shown in Figure-1.

Wound cultures obtained during surgery revealed growth of *Escherichia coli*, which was sensitive to several antibiotics, including imipenem-cilastatin, piperacillin-tazobactam, imipenem, amikacin and ciprofloxacin.

After debridement, vacuum-assisted closure (VAC) therapy was applied to the wound. The patient required intensive care management, including total parenteral nutrition (TPN) and insulin therapy adjustments.

Table 1: Laboratory test of first day of admission

Labs	Value	Reference
Hemoglobin A1c	7.73 %	4.8-5.9 %
Urea	129.1 mg/dL	15-45 mg/dL
Blood urea nitrogen	60.33 mg/dL	7-21 mg/dL
Creatinine	5.14 mg/dL	0.5-0.9 mg/dL
E - Glomerular filtration rate	8,57	
White blood count	35.49 x 10^9/L	4,49-12,68 x 10^9/L
Neutrophils	33.86 x 10^9/L	2.1-8.89 x 10^9/L
Procalcitonin	2.43 ng/mL	0-0.5 ng/mL
C-reactive protein	320.46 mg/dL	0-5 mg/dL
Alanine Aminotransferase	12.9 U/L	0-33 U/L
Aspartate Aminotransferase	18.7 U/L	8-43 U/L

In the following days, the patient's condition improved, with a decline in CRP (from 320 mg/dL to 37 mg/dL) and WBC (from 35.49 to 15.49 x109/L). On the 28th of October, wound cultures showed no growth. The patient was discharged on day 24 with a serum creatinin level of 3.68 mg/dL, CRP of 6 mg/dL and WBC of 7.26 x109/L.

Discussion

Fournier gangrene is a severe, rapidly progressing infection that often results in significant morbidity and mortality if not diagnosed and treated promptly. The treatment of Fournier gangrene involves a combination of surgical and medical approaches, each playing a critical role in halting the progression of the disease and supporting recovery. The primary treatment involves the surgical removal of all necrotic and infected tissue. Early and aggressive debridement is vital to prevent the infection from spreading further into healthy tissues. In addition to surgery, medical management is a cornerstone of Fournier gangrene treatment. Broad-spectrum intravenous antibiotics are used to target the polymicrobial nature of the infection, which typically involves both aerobic and anaerobic bacteria. Supportive care is equally important and includes stabilizing the patient through fluid resuscitation, managing sepsis, and controlling any underlying conditions such as diabetes or immunosuppression (1, 7, 8).

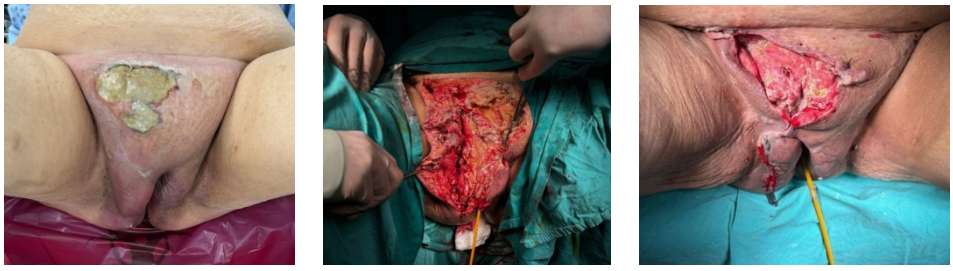

Figure 1. Preoperative image showing the abscess and surrounding tissues, intraoperative image during surgical debridement and postoperative image demonstrating partial closure of the wound

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The use of SGLT-2 inhibitors, such as dapagliflozin, has been associated with an increased risk of genitourinary infections, including Fournier gangrene, due to glucosuria, which may predispose patients to infections. While no direct causal relationship between dapagliflozin and Fournier gangrene can be established, the potential association warrants further investigation.

In comparison to other reported cases of FG, as shown in Table-2, our patient falls within a typical age range. In our case, the patient is 59 years old, while in the literature, most patients with FG tend to be in the 50-70 age range (2, 4, 5, 9, 10). However, there are instances of FG occurring in both younger and older populations, indicating that while age is a factor, it is not the sole determinant for the development of this life-threatening condition (1, 7, 8, 11).

Regarding gender, our patient is female, but the majority of FG cases in the literature involve male patients. In our case, this gender difference aligns with the literature, where men are predominantly affected, likely due to anatomical and hormonal differences that may predispose them to more frequent infections in the genital and perineal regions (1, 2, 4, 5, 7, 11). However, FG in females, although less common, is still recognized, particularly in the presence of comorbid conditions such as diabetes and obesity (8, 9, 10).

Our patient's chronic conditions—DM, HT, CKD, and CAD—are seen in a significant portion of FG cases. In fact, diabetes is one of the most common comorbidities in patients with FG, which is reflected in our case. Several other case reports highlight diabetes as a key risk factor for FG, as poor glycemic control impairs immune function and promotes infection (2, 5, 7, 9).

On admission, our patient's lab results included a significantly elevated white blood cell (WBC) count and C-reactive protein (CRP), which are typical markers of infection and inflammation. The patient's HbA1c was 7.73%, which indicates suboptimal control of her diabetes. In comparison, other FG patients in the literature often present with high WBC counts and CRP levels, and many have suboptimal or poorly controlled diabetes, similar to our case (5, 7, 8, 10, 11). However, the serum creatinine levels in our patient (5.14 mg/dL) were notably higher than those reported in many other studies, reflecting the severity of her chronic kidney disease, which complicates her treatment and recovery.

In terms of management, our patient received broad-spectrum intravenous antibiotics, including imipenem-cilastatin and teicoplanin, which is consistent with the literature. Antibiotic therapy for FG is typically guided by culture results, as polymicrobial infections are common. In our patient, *Escherichia coli* was identified as the causative organism, which was sensitive to a range of antibiotics, including imipenem-cilastatin, amikacin, and ciprofloxacin. However, in cases in the literature *Streptococcus* species were more prevalent (5, 8, 10, 11).

Surgical management, including urgent debridement, is critical in FG, and our patient underwent emergent debridement followed by VAC therapy. The use of VAC therapy is increasingly common in FG cases as it promotes wound healing and reduces infection.

The patient's clinical course following surgery was favorable, with a decline in CRP and WBC counts, and she was discharged after 24 days with significant improvement in her kidney function and resolution of the infection.

In conclusion, our case of Fournier's gangrene highlights several similarities and differences compared to those described in the literature. Like other reported cases, our patient's age, comorbidities, and clinical presentation are consistent with the typical risk factors and symptoms of FG. Early diagnosis, aggressive surgical debridement, and appropriate antibiotic therapy remain the cornerstones of successful management, as demonstrated in our patient and supported by the existing literature.

Conclusion

This case underscores the potential risks associated with the use of SGLT-2 inhibitors, particularly dapagliflozin, in patients with diabetes mellitus. Although these medications offer significant benefits in terms of glycemic control and cardiovascular protection, clinicians must be vigilant in monitoring for rare but serious side effects, including Fournier gangrene. Early detection, appropriate surgical intervention, and antibiotic therapy are crucial for improving outcomes in such cases.

The case report has written in an anonymous characteristic, thus secret and detailed data about the patient has removed.

Table 2: Summary of case reports of Fournier's gangrene associated with SGLT2 inhibitor treatment

Authors	Age	Gender	Smoking	Comorbidities	BMI (kg/ m^2)	Medication (SGLT-2)	WBC (x10^9/L)	Disharge day	CRP (mg/ dL)	HbA 1c (%)	SCr (mg/ dL)	WoundCultureResults	Antibiotics
Jahir et al., 2022	58	Female	n.a.	T2DM, HT, HL	48.3	Empagliflozin	26.6	n.a.	27.0	7.3	1.38	Streptococcusviridans Corynebacterium	Vancomycin, meropenem, clindamycin
Khokhar et al., 2022	55	Male	n.a.	HIV, T2DM, HL, HT, Obstructive sleep apnea	n.a.	Empagliflozin	13	3	n.a.	8.2	0.9	Streptococcusanginosus Staphylococcus epidermidis	Vancomycin, piperacillin-tazobactam, clindamycin (empirically) De-escalated to ampicillin-sulbactam based on culture results
Ellegardand Prytz, 2020	52	Female	Yes	T2DM,HT, asthma, hepatitisB	42	Dapagiflozin	19	18	227	n.a.	n.a.	Combination of aerobic andanaerobicpathogens	Piperacillin tazobactamswitched to meropenem, clindamycin
Elbeddini et al., 2020	72	Male	No	T2DM	n.a.	Canagliflozin	17.8	30	n.a.	7.5	n.a.	Bacteroides ovatus (fragilisgroup),Prevotella denticola,Actinomyces species	Meropenem, vancomycin, clindamycin (emprically) stepped down to oral sulfamethoxazole-trimethoprim, ciprofloxacin, metronidazole
Kumaretal., 2017	41	Male	Yes	T2DM	38	Empagliflozi n	18.3	15	283.1	11.2	n.a.	Streptococcusanginosus Mixedanaerobesand gram negative bacilli	Amoxicillin, gentamicinand vancomycin
Vargoetal., 2021	64	Male	n.a.	AF, CAD, AVR	n.a.	Dapagliflozin	n.a.	9	n.a.	n.a.	n.a.	n.a.	n.a.
Suciuetal., 2024	51	Male	Yes	CAD, AF, HF	n.a.	n.a.	40.2	n.a.	57.78	n.a.	3.29	Klebsiella pneumoniae	n.a.
Naganoetal., 2019	34	Male	Yes	T2DM	28	Empagliflozi n	21.7	41	41	6.5	0.78	Methicillin-resistant Staphylococcusaureus	Meropenem, clindamycinswitched to vancomycin
Elbeddiniet al., 2021	71	Female	n.a.	T2DM, HT, HL	n.a.	Dapagliflozin	33.2	14	n.a.	11.7	2.36	Streptococcusanginosus	Piperacillin- tazobactam, vancomycin, clindamycin (empirically)switched to piperacillin- tazobactam and clindamycin
Thiscase	59	Female	No	T2DM, HT, CKD, CAD	36	Dapagliflozin	35.49	24	320	7.73	5.14	Escherichiacoli	Imipenem-cilastatin, teicoplanin

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Case Report

Journal of Emergency Medicine Case Reports

Recurrence of Germ Cell Testicular Tumor Presenting with Choriocarcinoma Syndrome and Acute Pancreatitis

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Abstract

Choriocarcinoma syndrome (CS), characterized by pulmonary hemorrhage, widespread metastases, and markedly elevated beta-human chorionic gonadotropin levels, poses significant challenges in both diagnosis and treatment. In this case report, we present a rare case of recurrent mixed germ cell testicular tumor presenting to the emergency department with acute respiratory distress syndrome (ARDS) secondary to CS and pancreatitis. CS is a rare but highly fatal syndrome, often arising as a complication of advanced-stage germ cell tumors. Since these patients primarily present to emergency departments, it is crucial for emergency physicians to consider CS in the differential diagnosis when young male patients with a history of testicular tumors present with acute respiratory distress, hemoptysis, abdominal pain, or symptoms resembling pancreatitis.

Keywords: Choriocarcinoma syndrome, Emergency department, Germ cell testicular tumors, Pancreatitis

Introduction

Germ cell testicular tumors (GCTs) are among the most common malignant neoplasms in males aged 15-35 years and are classified into seminomatous and non-seminomatous subtypes. Non-seminomatous germ cell tumors (NSGCTs) include yolk sac tumors, choriocarcinoma, embryonal carcinoma, teratoma, or mixed tumors comprising combinations of these subtypes. Mixed germ cell tumors, accounting for approximately 30-50% of NSGCTs, are of particular importance due to their high malignant potential and propensity for metastasis to distant organs (1,2).

Tumors containing choriocarcinoma components are prone to necrosis and hemorrhage due to rapid proliferation and vascularization, potentially leading to life-threatening complications. Choriocarcinoma syndrome (CS), characterized by pulmonary hemorrhage, widespread metastases, and markedly elevated beta-human chorionic gonadotropin (β-HCG) levels, poses significant challenges in both diagnosis and treatment (3,4). Additionally, mature pancreatic tissue present within teratomas may exhibit endocrine or exocrine function, manifesting as clinical and radiological findings such as pancreatitis or hypoglycemia (5).

In this case report, we present a rare case of recurrent mixed germ cell testicular tumor presenting to the emergency department (ED) with acute respiratory distress syndrome (ARDS) secondary to CS and pancreatitis.

Case Report

A 31-year-old male presented to the ED with complaints of sudden-onset abdominal pain and shortness of breath. His symptoms had begun approximately one hour prior, without associated nausea, vomiting, cough, or sputum production. Initial vital signs revealed an oxygen saturation of 55%, blood pressure of 190/120 mmHg, a pulse rate of 120 bpm,body temperatureof 37.5°C, and heart rhythm wasregular.

Physical examination showed tenderness in the upper abdominal quadrants, diffuse crackles, and bronchospasm on lung auscultation.Neurological examination revealed no altered mental status or focal deficits.Electrocardiogram (ECG) findings demonstrated sinus tachycardia with non-specific ST-T wave changes.

The patient's medical history revealed that he had undergone orchiectomy for a testicular tumor three years earlier, followed by three cycles of chemotherapy. Post-

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treatment Positron Emission Tomography-Computed Tomography (PET-CT) showed no residual disease, and therapy was discontinued. The patient reported being symptom-free for the past two years and had not attended follow-up appointments. The pathology report indicated that the excised 17 × 13 × 12 cm orchiectomy specimen consisted of a mixed germ cell tumor comprising 40% choriocarcinoma, 40% teratoma, and 20% yolk sac tumor.

Before performing laboratory and imaging studies, alternative diagnoses such as cardiac emergencies, acute asthma exacerbation, acute pulmonary edema, aortic dissection and thyrotoxicosis were considered due to the patient's clinical presentation.

Laboratory tests performed in the ED revealed elevated amylase (358 U/L) and lipase (733 U/L) levels, marked leukocytosis (WBC 23.5 × 10^3/μL), and a significantly increased β-HCG (3179.77 IU/L). Thyroid function tests were within normal limits; therefore, thyrotoxicosis was ruled out.Additional laboratory findings are summarized in Table 1.

Table 1: Laboratory results of the patient

Biochemistry Tests	Complete Blood Count	Blood Gas Analysis
Glucose: 54 mg/dL	WBC: 23.5 x10^3/μL	pH: 7.515
Urea: 30 mg/dL	RBC: 4.3 x10^3/μL	PCO2: 21.0 mmHg
Creatinine:0.92mg/dL	HGB: 13.2 g/dL	PO2: 45.5 mmHg
Total Bilirubin: 5.8 mg/dL	HCT: 39.0%	$HCO3^-$ (actual): 16.6 mmol/L
Direct Bilirubin: 0.8 mg/dL	MCV: 90.4 fL	BE: -4.2 mmol/L
ALT: 66 IU/L	MCH: 30.6 pg	SO2: 80.1%
AST: 132 IU/L	PLT: 108.0 x10^3/μL	Na^+: 136.2 mmol/L
GGT: 93 U/L		K^+: 3.84 mmol/L
LDH: 992 IU/L		Ca^{2+}: 1.02 mmol/L
Amylase: 358 U/L		Cl^-: 113 mmol/L
Lipase: 733 U/L		Hb: 13.2 g/dL
CRP: 192 mg/L		$HCO3^-$ (standard): 20.6mmol/L
β-HCG: 3179.77 IU/L		

ALT: Alanine Aminotransferase, AST: Aspartate Aminotransferase, GGT: Gamma-Glutamyl Transferase, LDH: Lactate Dehydrogenase, CRP: C-Reactive Protein, β-HCG: Beta-Human Chorionic Gonadotropin, WBC: White Blood Cells, RBC: Red Blood Cells, HGB: Hemoglobin, HCT: Hematocrit, MCV: Mean Corpuscular Volume, MCH: Mean Corpuscular Hemoglobin, PLT: Platelets, PCO2: Partial Pressure of Carbon Dioxide, PO2: Partial Pressure of Oxygen, HCO_3^-: Bicarbonate, BE: Base Excess, SO_2: Oxygen Saturation, Na^+: Sodium, K^+: Potassium, Ca^{2+}: Ionized Calcium, Cl^-: Chloride, Hb: Hemoglobin

Contrast-enhanced thoracic and abdominal computed tomography revealed the following findings:

- Multiple hypodense nodular lesions in liver segments, some with peripheral contrast enhancement and others non-enhancing,
- An enlarged pancreas with heterogeneous parenchyma and poorly defined hypodense nodular areas,
- 1 cm nodular lesion in the upper pole of the spleen,
- Widespread hypodense nodules in both kidneys,
- Multiple air cysts, nodules, and diffuse ground-glass opacities in both lungs.

The imaging findings were suggestive of widespread metastatic disease and CS (Figure 1).

Given the patient's rapidly worsening hypoxia, altered mental status, and hemodynamic instability, he was emergently intubated in the ED and placed on mechanical ventilation. The decision for intubation was based on profound respiratory failure, a high risk of airway compromise due to hemoptysis, and deteriorating hemodynamic parameters. Following stabilization, the patient was admitted to the intensive care unit (ICU) with diagnoses of acute pancreatitis and ARDS. Despite aggressive supportive care, including mechanical ventilation, vasopressor support, and broad-spectrum antibiotics, the patient succumbed on the third day of ICU stay.

Discussion

This case presents a fatal complication of CS, characterized by pancreatitis and ARDS, resulting from recurrent mixed germ cell testicular tumor and widespread metastases in a young male. This syndrome, characterized by elevated β-HCG levels, widespread metastatic involvement, and rapid progression, stands out as a significant clinical entity that requires a multidisciplinary approach in both diagnosis and treatment.

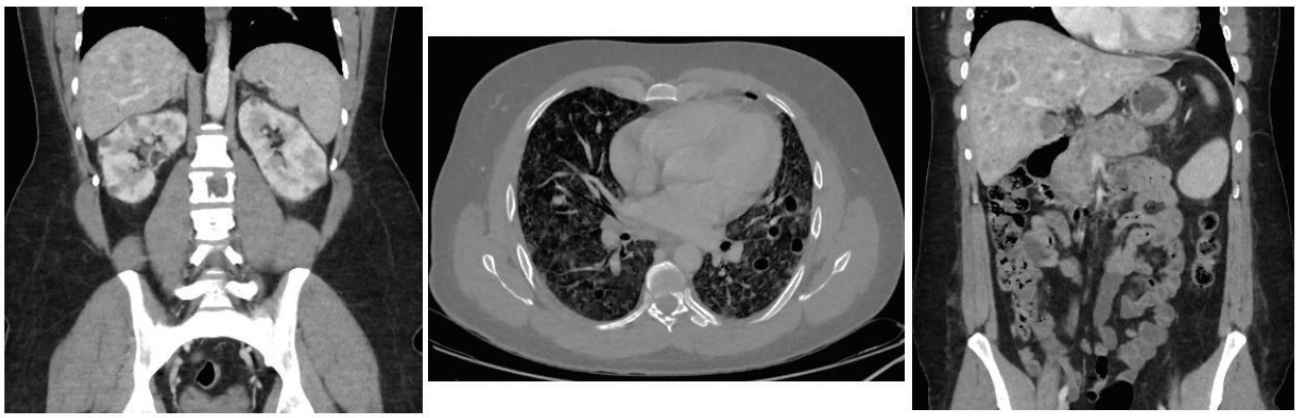

Figure 1. Thorax and abdomen CT of the patient

GCTs are common in young males, but the rarity of CS as a complication makes it challenging for many clinicians to recognize this clinical presentation. Although the exact pathogenesis of CS remains unclear, direct invasion of small vessels by tumor cells and the high vascularization in these areas are proposed as the primary mechanisms leading to hemorrhages at metastatic sites. Notably, diffuse alveolar hemorrhage, which occurs in lung metastases, is both a typical complication of CS and one of the most common causes of death (6-9). This case exemplifies how the development of ARDS due to lung involvement determined the prognosis and how no response was achieved with treatment.

The presence of pancreatitis in this patient is an uncommon but recognized finding in mixed germ cell tumors, and it aligns with certain teratoma cases reported in the literature. Teratomas are known to exhibit exocrine or endocrine activity due to the mature pancreatic tissue they contain (5,9-11). This case suggests that pancreatitis may have developed as a result of both the systemic effects of CS and the teratoma component of the tumor. Furthermore, the coexistence of multisystem complications such as pancreatitis and ARDS has posed additional challenges in clinical management.

In the treatment of CS, close hemodynamic monitoring and prompt intervention in the intensive care unit are essential. However, there is still no consensus on the optimal chemotherapy regimen and timing based on current data. While early recognition of CS may allow for the use of low-dose chemotherapy or induction therapies that could reduce mortality, there is insufficient prospective data to support this (3,4,8,9). In our case, the patient had received chemotherapy two years after orchiectomy, achieved remission, and showed no metastatic involvement at that time. However, due to the absence of any symptoms in the following two years, the patient did not attend follow-up visits. The sudden onset of dyspnea and abdominal pain initially led to other differential diagnoses, but through a detailed history, review of previous tests, imaging findings, and a literature search, the diagnosis was confirmed. Nevertheless, because CS is an aggressive and often fatal complication, the patient passed away on the third day despite supportive therapy.

In conclusion, CS is a rare but highly fatal syndrome, often arising as a complication of advanced-stage germ cell tumors. This case emphasizes the importance of careful monitoring of tumor recurrence and the need to anticipate systemic complications that may develop in metastatic disease. Since these patients primarily present to EDs, it is crucial for emergency physicians to consider CS in the differential diagnosis when young male patients with a history of testicular tumors present with acute respiratory distress, hemoptysis, abdominal pain, or symptoms resembling pancreatitis.

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Case Report

Journal of Emergency Medicine Case Reports

A Case of Salmonella Bacteremia in a New Diagnosed HIV-Positive Adult Patient

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Abstract

Salmonella species are gram-negative bacteria that cause foodborne infections with significant morbidity and mortality worldwide. Salmonella species are known pathogens associated with bacteremia, especially in immunocompromised patients. Salmonellosis is an AIDS-defining disease when it occurs in HIV-positive patients. In this case report, HIV-positive adult patient diagnosed with Salmonella bacteremia with prominent respiratory symptoms but no gastrointestinal symptoms is presented. It is concluded that Salmonella bacteremia is a diagnosis that should not be overlooked among the manifestations of HIV infection.

Keywords: AIDS, bacteremia, HIV, salmonella

Introduction

Salmonella species are Gram-negative bacteria classified in Enterobacteriaceae associated with human and animal infections. The most common initial symptoms of salmonellosis are fever, nausea, vomiting, headache, abdominal pain, diarrhea, chills, and arthralgias. Non-typhoid *Salmonella* (NTS) infections occur with various clinical syndromes, including gastroenteritis, bacteremia, endovascular infection, and focal infections (1-3). Although salmonellosis is generally a self-limiting disease in immunocompetent individuals, HIV-positive patients are at great risk of developing bloodstream infection, such that recurrent salmonella septicemia. So that, recurrent *Salmonella* septicemia has been recognized as an acquired immunodeficiency syndrome (AIDS)-defining illness (3). In developed countries, secondary bacteremia develops in approximately 5% of patients with non-typhoidal salmonellosis, and this bacteremia usually occurs in immunosuppressed HIV patients, patients with malignancy, chronic kidney or liver disease, diabetes, etc. (2). This case report describes a *Salmonella* bacteremia occurring in an HIV-infected adult patient.

Case Report

In this case, is presented a *Salmonella* bacteremia occurring in a 23-year-old newly diagnosed HIV-positive adult male patient. The patient had presented to the emergency department with a five-day history of cough, sputum, and intermittent hemoptysis. In addition to these complaints, fever and nocturnal sweats also accompanied. The patient had nausea but no vomiting. The day before coming to the outpatient clinic, the patient had semi-solid, non-bloody mucoid defecation twice daily. The patient was hospitalized at the infectious diseases clinic due to preliminary diagnoses of bacterial pneumonia, pneumocystis pneumonia (PCP) and pulmonary tuberculosis. The patient was diagnosed with HIV approximately three months before hospitalization and had not previously received any antiretroviral treatment. The patient received fifteen days of outpatient treatment for pneumonia a year ago. He lost approximately 10 kg in ten days at that time. The patient has been smoking for twelve to thirteen years but does not drink alcohol or addictive substances.

On admission, the patient was conscious, presented with a Glasgow Coma Score (GCS) of 15/15, cooperative,

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oriented, hemodynamically and respiratory stable, body temperature of 36.6 °C, pulse rate of 74 per min, blood pressure of 103/70 mmHg, oropharynx hyperemic. There was widespread candidal plaque in the oral mucosa and generalized lymphadenopathy with the nodes 1.1-1.7 cm in diameter in the submandibular and axillary zones. There was tenderness in the right upper quadrant of the abdomen but defense and rebound were negative. Lung breath sounds were normal. No amoeba, parasites, leukocytes, or erythrocytes were observed in stool microscopy but stool was positive for occult blood. *Salmonella* spp was positive in the blood culture taken from the patient during the fever period on the first day of hospitalization. There was no bacterial growth in the sputum culture with negative PCP. They found that anergic purified protein derivative (PPD), sputum acid-fast bacilli for Mycobacteria and I*nterferon Gamma Release Assay* (IGRA) tests were negative. Pre-treatment and post-treatment laboratory data are given in Table-1.

On the patient's thoracic tomography, consolidation areas containing infectious air bronchogram were observed on the posterior upper lobe of the right lung and the mediobasal and posterobasal segments of the left lung (Figures-1 and 2). Pneumonic consolidation zones were consistent with tuberculosis, but laboratory data did not support tuberculosis. No any significant finding was observed in the abdominal ultrasonography. Echocardiography did not reveal any vegetation or thrombus.

Piperacillin-tazobactam 4x4.5 gr treatment was initially started empirically due to respiratory symptoms and consolidations in favor of pneumonia on the thoracic tomography. In the follow-up of the patient, cough, sputum, hemoptysis, and diarrhea symptoms regressed, and the fever continued to be undulating. Piperacillin-tazobactam therapy was stopped on the 11th day in the patient who had bacteremia, and a 7-day tigecycline (100 mg loading dose, then 50 mg twice daily) treatment was applied. On the 11th day of the patient's hospitalization,

Table 1: Pre and post-treatment laboratory data

Parameters	Pre-treatment values	Post-treatment values (18th day)
WBC	3400/mm^3	2400/mm^3
PLT	48,000/mm^3	130.000/mm^3
Hb	8.9 g/dL	9.2 g/dL
AST	80 IU/L	23 IU/L
ALT	46 IU/L	19 IU/L
CRP	385 mg/L	8.4 mg/L
PCT	10.39 ng/mL	0.03 ng/mL
CD4$^+$ T cell	7 cells/µL	-
HIV-RNA	3743194 IU/mL	-

WBC: White blood cell; **PLT:** Platelet; **Hb:** Hemoglobin; **AST:** Aspartate aminotransferase; **ALT:** Alanine aminotransferase; **CRP:** C-reactive protein; **PCT:** Procalcitonin; **HIV-RNA:** Human Immunodeficiency Virus – Ribonucleic acid.

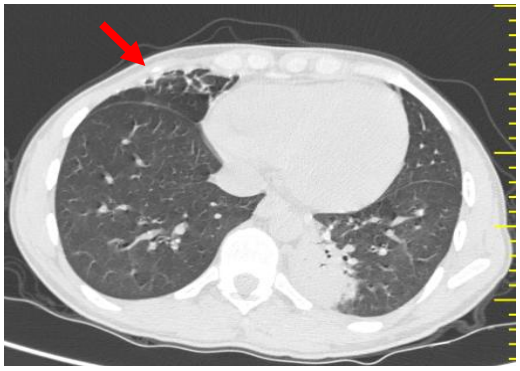

Figure 1. Consolidation area on the anterior upper lobe of the right lung on the thoracic tomography

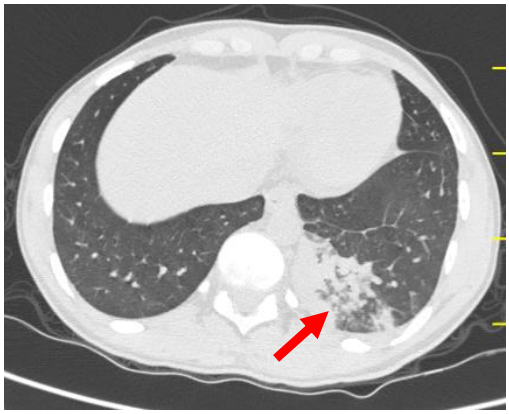

Figure 2. Consolidation area on the the mediobasal and posterobasal segments of the left lung on the thoracic tomography

both antiretroviral therapy with bictegravir/emtricitabine/tenofovir alafenamide (B/F/TAF) 50 mg/200 mg/25 mg once daily and trimethoprim/sulfamethoxazole 160/800 mg once daily as primary prophylaxis was started when pulmonary tuberculosis was excluded. Seven days after the treatment, no bacterial growth was observed in the control blood culture. The patient responded positively to the therapy applied. The patient improved clinically and was discharged on the 18th day of the hospitalization because the fever was limited and he had no active complaints. In the 4th month of antiretroviral therapy, HIV-RNA was negative, and CD4 count was 237 cells per µL. In the 10th month of antiretroviral therapy, the CD4 count was found to be 423 cells per µL, and trimethoprim/sulfamethoxazole prophylaxis was discontinued.

Discussion

Here, we report a rare case from our country in which both Salmonella bacteremia and HIV-infection coexist in an adult patient. Salmonellosis is an infection usually characterized by fever, abdominal pain, diarrhea, nausea, and vomiting. Symptoms of illness usually appear 6-72 hours after the agent is ingested, and this period can last up to 7 days (3). *Salmonella* infections with self-limiting enterocolitis

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generally do not require antimicrobial therapy, but when *Salmonella* bacteria enter the systemic circulation, all tissues and organs are susceptible, causing various focal *Salmonella* infections (4). It is also recommended to investigate whether the patient has a genetic or acquired immune deficiency or an endovascular focus of infection when *Salmonella* bacteremia is diagnosed in an adult patient (5). Therefore, it is reported that *Salmonella* infection and especially bacteremia should be considered as a diagnosis of AIDS in a patient at risk for HIV infection (3,5). We reported a *Salmonella* bacteremia in an HIV-positive adult patient, which is rarely seen in our country.

On the other hand, patients with invasive Salmonella disease often present with focal infection in the lower respiratory tract (2). Respiratory system symptoms such as cough, sputum, and hemoptysis were prominently observed in our HIV-positive patient with *Salmonella* bacteremia but no obvious gastrointestinal symptoms other than diarrhea. A *Salmonella* bacteremia was previously reported in an HIV-positive adult patient in Türkiye, which was also caused by *Salmonella arizonae* (6). Although the cited case reported that the HIV-positive patient also had diabetes, bronchiectasis, and tuberculous lymphadenitis, the patient's respiratory and gastrointestinal symptoms were unclear.

In a retrospective analysis of patients with non-typhoidal *Salmonella* bacteremia in a study conducted in Malaysia, an extraintestinal focus of infection was noted in 30.9% of patients, the majority of which were pulmonary and soft tissue infections. In the mentioned study, 65.5% of the patients had serious underlying clinical immunosuppressive conditions, the most common being malignancy and HIV (7). Similarly, our patient had underlying HIV infection, and *Salmonella* bacteremia was associated with pneumonia. Again, CD4 T-lymphocyte counts are reported as less than 200 cells per μL in the majority of adult patients with invasive non-typhoidal salmonella disease in association with HIV (2,3). Our patient's CD4 T-lymphocyte count was found to be less than the mentioned value (Table-1).

It is reported that treatment of salmonellosis with antibiotics does not reduce the duration of uncomplicated *Salmonella* gastroenteritis but rather significantly prolongs the period of fecal excretion of bacteria and increases the risk of resistance to antibiotics. Therefore, salmonellosis should be treated with antibiotics only in the presence of bacteremia, enteric fever, focal infection or abscess (8). Tang et al. (9) showed that tigecycline had significant in vitro and in vivo antimicrobial activities against extracellular and intracellular *Salmonella* in a murine peritonitis model. However, the emergence of *Salmonella* antibiotic resistance is also a significant public health problem worldwide (1,3). Furthermore, tigecycline was also found to be effective against ceftriaxone-resistant *Salmonella* spp. (10). Since one of our patient's preliminary diagnoses was tuberculosis, we avoided the use of quinolones in order not to encourage tuberculosis resistance. Tigecycline therapy was applied for the treatment of *Salmonella* bacteremia, and our patient responded favorably to this treatment.

Conclusion

In this case report, it was concluded that *Salmonella* bacteremia, which has relatively few gastrointestinal symptoms, is a diagnosis that should not be overlooked among the signs of HIV infection and that careful treatment with effective antibiotic therapy is necessary to a good prognosis.

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Case Report

Journal of Emergency Medicine Case Reports

Retained Foreign Body not Detected by Imaging Methods

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Abstract

In this case presentation, we aimed to present a case of a retained foreign body in the gluteal region of a male child that was overlooked in imaging methods. An 11-year-old male child was brought to the emergency department by his family after reporting that a splinter had entered his left inner hip area while sliding on a slanted board they had placed to play. A lesion was detected on the left inner hip area, where the patient indicated pain, potentially corresponding to the point of entry. However, no foreign object or hardness was felt upon examination. A superficial ultrasound (US) examination was performed, and no pathology was found in the lesion site or the surrounding area. As the patient's severe pain persisted, a pelvic computed tomography (CT) scan was conducted. The pelvic CT scan revealed air densities in the region of the skin lesion, but no other pathology was identified. Later, due to the continuation of the patient's complaints, the area was done repeat physical examination along the air densities starting from the skin lesion. During this examination, a hardness was detected under the skin on the upper outer side of the right hip, at the end of the tract originating from the skin lesion. An incision was made at the site of the palpable hardness. Upon finding a foreign object, the child was made consultation to the pediatric surgery department. The foreign body, measuring approximately 35x0.8 cm, was then removed in a procedure performed by the pediatric surgery team. In cases of retained foreign body, although imaging methods such as US and CT are important for diagnosis, it should be remembered that in rare cases where imaging methods are not helpful, a thorough history and physical examination remain the most effective approach.

Keywords: Computed tomography, retained foreign body, ultrasound

Introduction

Retained foreign body accounts for 7% to 15% of emergency department visits, and it has been found that 38% of these foreign bodies are overlooked during the initial evaluation (1). In the United States, 37% of malpractice lawsuits related to emergency departments have been linked to foreign bodies (2). Materials such as wood, acrylic, and some plastics have densities similar to the surrounding soft tissues, making them difficult to visualize, and it has been reported that only 15% of wooden foreign bodies are detected in plain radiographs (3).

In this case presentation, we aimed to present a case of a retained foreign body in the gluteal region of an 11-year-old male child that was overlooked in imaging methods.

Case Report

An 11-year-old male child was brought to the emergency department by his family after reporting that a splinter had entered his left inner hip area while sliding on a slanted board they had placed to play. The patient's general condition was good, with clear consciousness, cooperation, and orientation. His vital signs were as follows: blood pressure 130/70 mmHg, pulse 76/min, respiratory rate 18/min, body temperature 36.5°C, and SaO2 99%. A lesion potentially corresponding to the point of entry was identified at the site of pain, as indicated by the patient, in the left inner hip region (Figure-1). However, no foreign body or hardness was felt upon examination. A superficial ultrasound (US) examination showed no pathology at the lesion site or in the surrounding area. As the patient's severe pain persisted, a pelvic computed tomography (CT) scan was performed. The pelvic CT scan revealed air densities in the region of the skin lesion (Figure-2), but no other pathology was found. Later, as the patient's complaints continued, the area was re-examined along the tract of air densities starting from the skin lesion. During this examination, a palpable hardness was found under the skin on the upper outer side of the right hip. A superficial US examination was requested at the site of the palpable hardness, but no pathology was identified again. Despite the lack of pathology detected in imaging methods, the palpable hardness and the patient's severe pain prompted a small incision to be made at the site of the hardness (Figure-3). Upon finding a foreign body, the child was consulted to the pediatric surgery department. A foreign

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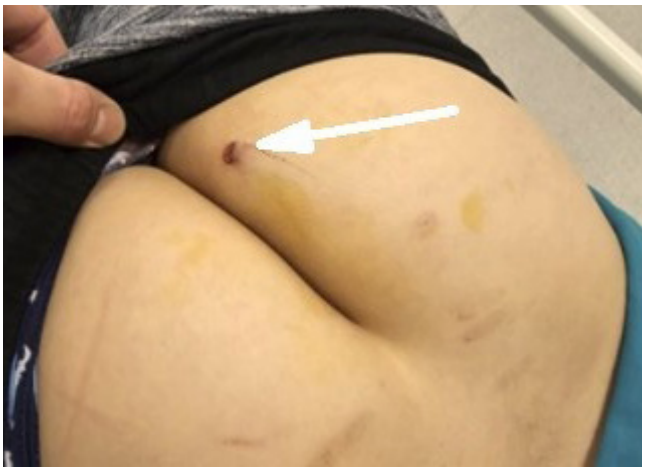

Figure 1. In the initial examination, the area where the patient reported pain, which was considered as the possible entry site, was identified.

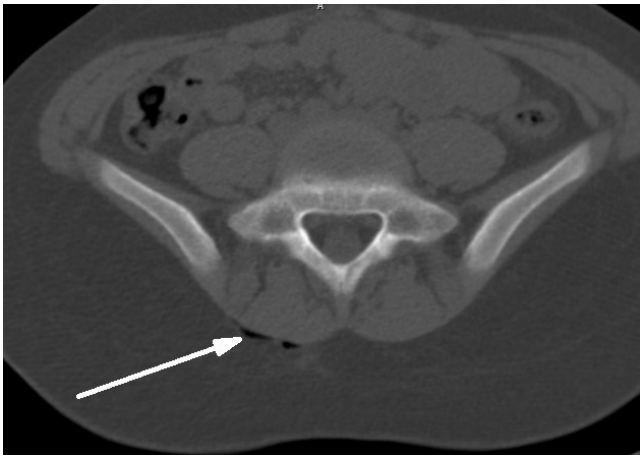

Figure 2. The pelvic CT scan revealed air densities in the region of the skin lesion.

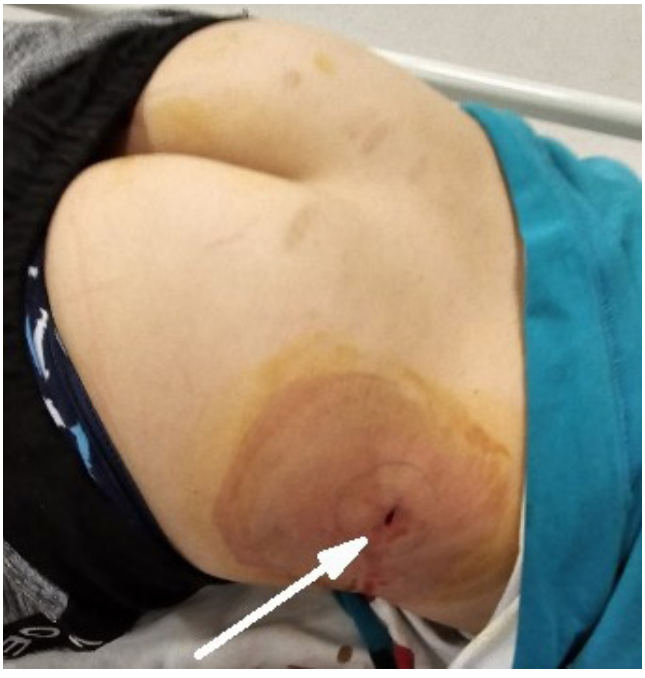

Figure 3. The incision made over the area with hardness during the examination.

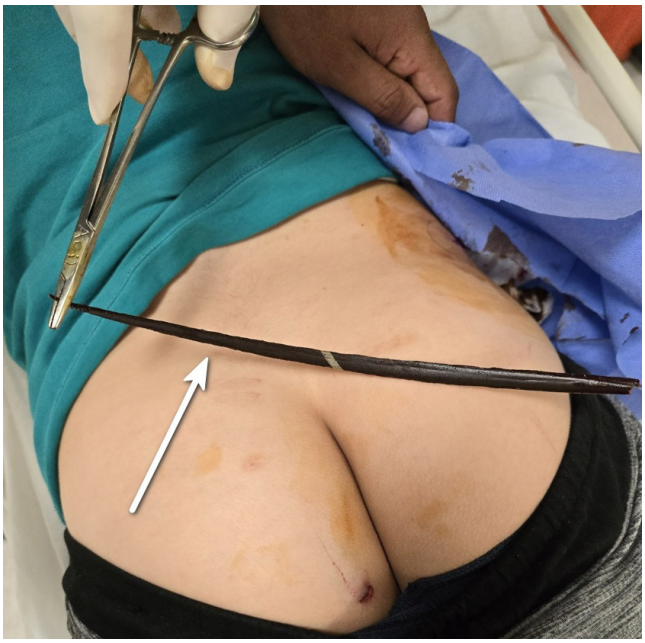

Figure 4. Removed foreign body

body measuring approximately 35x0.8 cm was removed during the procedure performed by the pediatric surgery team (Figure-4). The patient was followed and subsequently discharged.

Discussion

In patients presenting with suspicion of a foreign body, a detailed history and physical examination are crucial during the initial assessment. Metals, glass, and wood are the most commonly encountered foreign bodies (4). Metal objects are easily detected in plain radiographs. However, glass and wood are difficult to visualize on radiographs, and glass alone accounts for 50% of foreign bodies that are overlooked despite physical examination and radiography (5). In our case, the foreign body was a piece of wood, which was not detected through imaging methods at the initial stage but was diagnosed later based on suspicion following a careful physical examination.

In cases of injury within the first 24 hours, the entry site can usually be easily seen, and intervention is more straightforward, making the ideal time for diagnosis and treatment within the first 24 hours (6). Delayed treatment can lead to complications such as infection, delayed wound healing, and loss of function (7). Therefore, early diagnosis is essential; failure to make a diagnosis may lead to malpractice claims and compensation lawsuits (5). In our case, the patient presented to the hospital 2 hours after exposure to the foreign body, and the intervention was performed.

Bedside US is an easily accessible, radiation-free, inexpensive, and safe imaging method commonly used in emergency department practice. Since most wooden

foreign body insertions result from low-energy trauma, they are typically superficial. US is one of the best imaging techniques for diagnosing superficially located foreign bodies (8,9). However, in our case, the foreign body could not be detected using US.

For detecting deeper foreign bodiesCT may be used. CT has been shown to be the best imaging method for plastic, glass, and stone foreign bodies (10). In our case, following the failure to identify the foreign body on USG, a CT scan was performed based on the patient's history and physical examination findings. However, the initial interpretation of the CT scan was normal, with the exception of air densities. Afterward, a foreign body was detected during incision at the site of the palpable hardness, and the foreign body was removed.

Conclusion

In cases of retained foreign body, although imaging methods such as US and CT are important for diagnosis, it should be remembered that in rare cases where imaging methods are not helpful, a thorough history and physical examination remain the most effective approach.

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Case Report

Journal of Emergency Medicine Case Reports

A Rare Complication due to Methanol Poisoning: MINOCA

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Abstract

Myocardial infarction with non-obstructive coronary arteries (MINOCA) is a relatively novel clinical puzzle characterized by limited research. In this case, we aimed to present the co-occurrence of methanol poisoning and MINOCA syndrome. This case demonstrated the importance of monitoring cardiac function with electrocardiography and cardiac biomarkers in patients within a high anion gap metabolic acidosis environment. This case report was showing co-occurrences of MINOCA syndrome with methanol poisoning. This was the first case of myocardial infarction in non-obstructive coronary arteries associated with methanol poisoning accompanied by high anion gap metabolic acidosis in the literature.

Keywords: Emergency, metabolic acidosis, MINOCA, poisoning

Introduction

Myocardial infarction with non-obstructive coronary arteries (MINOCA) is a relatively novel clinical puzzle, accounting for approximately 10% of all acute myocardial infarction cases, characterized by limited research (1,2). In patients presenting with chest pain and acute myocardial injury detected by high-sensitivity troponin (hs-Tn) testing, the diagnosis is established when there is no stenosis of 50% or more in the coronary arteries on angiography (1). MINOCA constitutes a heterogeneous group of conditions that can develop due to various etiologies, including epicardial vascular causes such as coronary plaque rupture, spasm, and spontaneous dissection, as well as microvascular causes such as coronary thromboembolism and microvascular dysfunction, or non-ischemic events such as myocarditis and myopathies (3). Due to the differing pathophysiology, the management of patients with MINOCA differs significantly from that of other acute myocardial infarction (AMI) patients (4). In this case, we aimed to present the co-occurrence of methanol poisoning and MINOCA syndrome. We did not encounter any reported cases of myocardial infarction in non-obstructive coronary arteries associated with methanol poisoning accompanied by high anion gap metabolic acidosis in the literature we reviewed.

Case Report

A 41-year-old male patient was brought to our hospital's emergency department due to the sudden onset of altered consciousness. According to information obtained from the patient's relatives, a history of hypertension, hyperlipidemia and chronic alcohol consumption was reported. It was learned that on the day of admission, the patient consumed approximately one liter of homemade raki. Upon initial evaluation in the emergency department, the patient's overall condition was poor, with a Glasgow Coma Scale score of 6. His body temperature was 36.4°C, heart rate was 119 beats per minute, oxygen saturation (SpO_2) in room air was 85%, blood pressure was 70/40 mm-Hg, and blood glucose level was 100 mg/dL (Table-1). Electrocardiography (ECG) showed sinus tachycardia with T wave inversions in chest leads (Figure 1.A). Bedside echocardiography revealed an ejection fraction of 60%, no segmental wall motion abnormalities, and no evidence of pericardial effusion. The patient was intubated and connected to a mechanical ventilator.

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Methanol intoxication was considered due to the patient's history, clinical findings, and severe metabolic acidosis. Methanol level cannot be measured in our hospital. Paracetamol and salicylate levels were measured as toxicological parameters and the result was negative. Treatment for methanol poisoning was initiated. Fluid boluses and the dual inotropes were initiated due to hypotension. Intravenous sodium bicarbonate therapy was administered to correct acidosis. 50 mg of folic acid was started and the patient was urgently admitted to hemodialysis. The hourly monitoring of blood gas parameters for the patient who underwent continuous hemodialysis for 9 hours, along with admission and follow-up biochemical parameters, is presented in Table 1. After hemodialysis, the blood gas returned to normal. Throughout this period, the patient underwent ECG monitoring. The ECG findings associated with hyperkalemia resolved by the 3rd hour of hemodialysis in the patient whose hyperkalemia was corrected with hemodialysis (Figure 1.B). During the patient's latest follow-up, inferior ST-segment elevation myocardial infarction was detected on the ECG (Figure 1.C). The patient underwent emergency coronary angiography, which revealed normal coronary arteries.(Figure 2) Subsequently, the patient was admitted to the intensive care unit. During his hospitalization, he needed hemodialysis twice and plasmapheresis three times. The patient, who clinically improved during follow-up, was discharged after 47 days. The patient did not require hemolysis in subsequent follow-up.

Discussion

The MINOCA syndrome was first described 80 years ago. The initial studies conducted by De Wood et al. included demonstration of occluded coronary arteries in 90% of ST-elevation myocardial infarction (STEMI) patients. While this groundbreaking study emphasized the importance of coronary obstruction in the pathogenesis of AMI, it also confirmed the absence of obstruction in approximately 10% of patients (5). With the publication of a position paper on MINOCA by the ESC in 2016, its significance has been better understood and has paved the way for many subsequent studies. Particularly due to its occurrence in younger patients and the differences in etiology, prognosis, and treatment protocols, clinicians should pay special attention to it (3). In its etiopathogenesis, coronary artery spasm and coronary plaque rupture are the two most commonly known causes (3). Approximately 40% of MINOCA cases arise from plaque rupture. Plaque rupture leads to less than 50% stenosis of the coronary artery lumen. Coronary artery spasm is another major factor implicated in its etiology. This condition is thought to occur as a result of abnormal responses of vascular smooth muscles to endogenous or exogenous stimuli (3). When we look at other causes, myocarditis is also one of the most common non-coronary reasons for MINOCA. It is estimated to account for approximately one-third of cases. It primarily stems from Coxsackie virus, adenovirus, influenza

Table 1: Monitoring of the patient's blood gas, complete blood count, and biochemical tests

Arterial Blood Gas Parameters		pH	HCO_3	pCO_2 (mmHg)	Base excess	Potassium(mmol/L)	Lactate (mg/dL)
0th hour		6.81	7.2	46.6	-26.9	6.3	21
1st hour		6.91	8.9	49.8	-22.7	5.9	20
2nd hour		7.05	10.3	34.5	-20.9	5.9	16
3rd hour		7.14	14.4	45.0	-13.5	3.9	20
4th hour		7.25	17.2	41.7	-8.5	3.2	27
5th hour		7.29	21.7	51	-1.8	3.2	12
6th hour		7.34	21.3	40.8	-3.7	2.8	10
7th hour		7.33	24.2	51.8	+1.4	2.7	7
8th hour		7.37	23.9	43.7	+0.5	3.1	10
9th hour		7.41	23.3	35.8	-1.6	3.7	12
Biochemical Parameters	**0th hour**	**9th hour**	**1st day**	**2nd day**	**3rd day**	**7th day**	**15th day**
Hs Troponin-I (ng/L)	25	73	-	233500	56500	-	315
Urea (mg/dL)	125	19	32	124	188	116	142
Creatinine (mg/dL)	4.75	0.79	1.1	4.16	5.74	1.62	4.33
Aspartate aminotransferase (U/L)	122	167	433	393	105	47	178
Alanine aminotransferase (U/L)	84	69	85	98	62	32	490
White Blood Cell (10^3/µl)	17.6	4.6	3.8	16.2	24.3	35.2	19.2
Hemoglobin (g/dL)	12.8	12.9	11.4	12.8	10.5	10.2	6.7
hematocrit (%)	46.5	37.1	32.4	37.8	30.5	31.7	21.4
Platelet counts (10^3/µl)	89	50	38	55	80	247	177
INR	1.19	0.94	1.58	1.0	1.08	1.18	1.35
APTT (s)	53.3	30.7		33.2	27.3	29.6	37.5
Ethanol (Promil)	0.074						

HCO_3: Bicarbonate, pCO_2 : partial pressure of carbon dioxide, mmHg: millimeters of mercury,dl: deciliter, g: gram, L: liter, mEq: milliequivalent, mg: milligram, mL: milliliter, mm: millimeter, mmol: millimoles, s: second, U:unit, ng:nanogram

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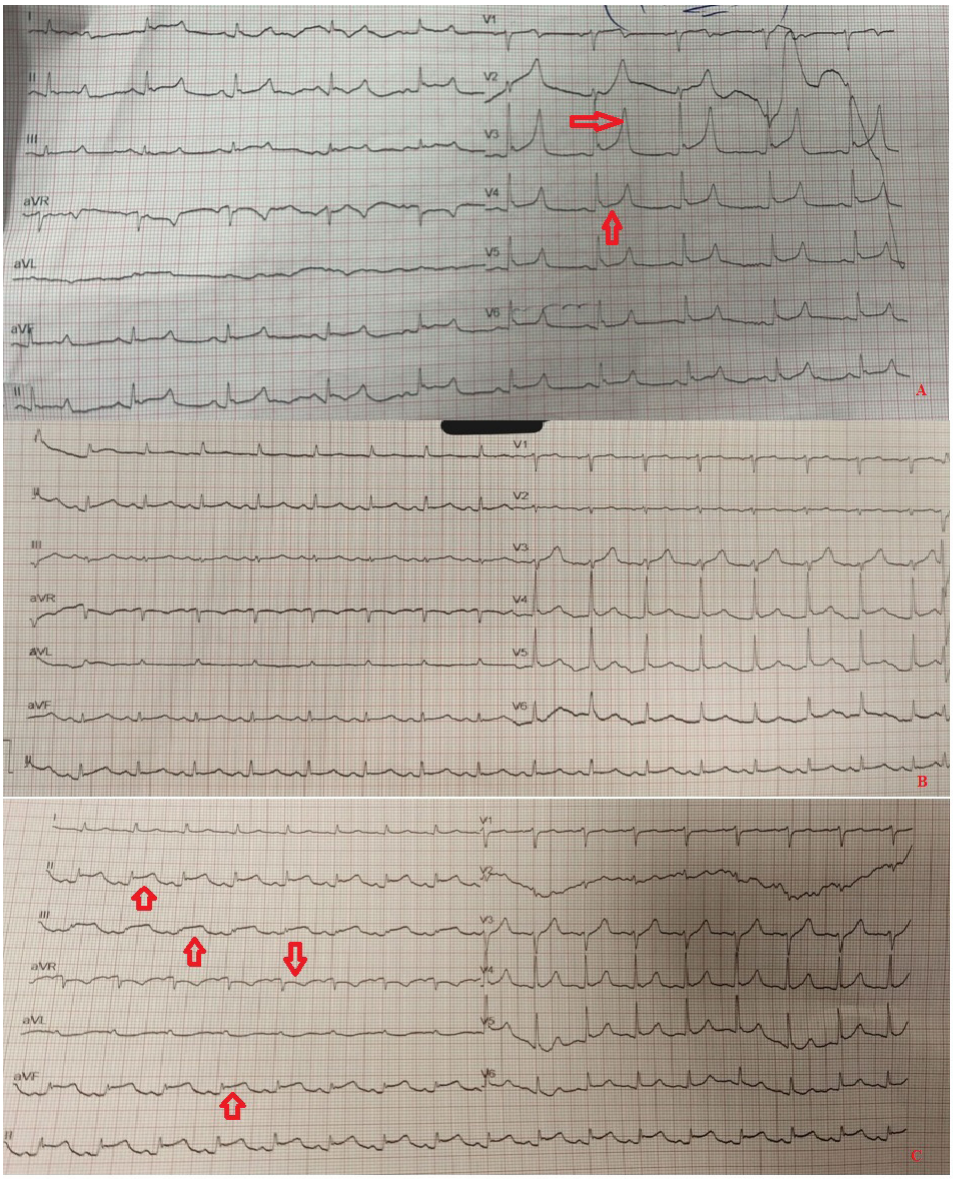

Figure 1. The initial electrocardiogram of the patient highlights T-wave inversions in the chest leads, as indicated by the red arrow **(A)**. This was accompanied by hyperkalemia. It is observed that the T-wave inversions on the electrocardiogram returned to normal after the potassium level returned to normal **(B)**. In the subsequent electrocardiogram, ST segment elevation observed in the inferior leads, indicated by red arrows, is visible **(C)**.

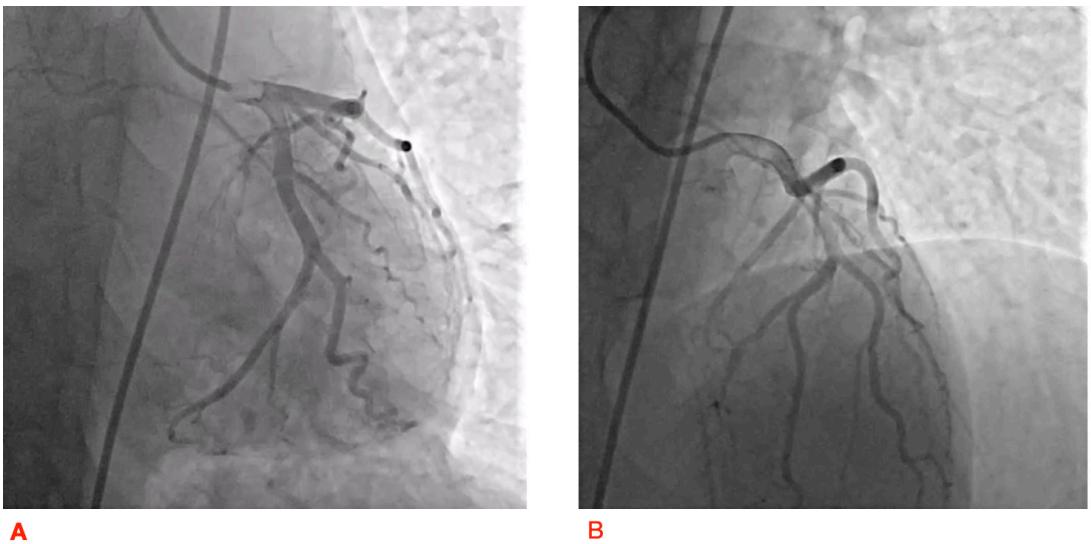

Figure 1. The patient's coronary angiography findings. It is observed the normal right coronary artery and its branches (A). It is observed the normal left coronary artery and its branches (B).

virus, and Epstein-Barr virus (3). When the literature is reviewed, it is observed that MINOCA syndrome accompanied by methanol poisoning has not been reported.

Although methanol level was required for the diagnosis of methanol poisoning, this test was not available in our hospital, as in most centers. Diagnosis is made with clinical suspicion, history of illicit alcohol use, characteristic examination findings (neurological, ophthalmological, etc.) and the presence of metabolic acidosis with a wide anion gap. (6,7) There are very limited datas in the literature regarding the effects of methanol poisoning on the cardiovascular system.(6)

Although the direct cardiovascular effects of methanol or its metabolites are unknown, The hemodynamic instability and high anion gap metabolic acidosis disrupted in methanol poisoning can lead to both impaired coronary artery perfusion and suppression of myocardial tissue function. Some studies have found ECG changes in cases of methanol poisoning.(6-8) Although the observed ST segment elevation in our case provided metabolic and hemodynamic stability, it indicates that coronary and myocardial involvement persists. This case demonstrates the importance of monitoring not only cardiac function but also ECG and cardiac biomarkers in patients within a high anion gap metabolic acidosis environment.

Conclusion

Cardiac involvement, which is less common in patients with methanol poisoning than gastrointestinal and neuro-ophthalmological findings, should definitely be kept in mind. Therefore, routine cardiac monitoring is important in suspected patients.

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Case Report

Journal of Emergency Medicine Case Reports

Acute Maras Powder Intoxication-Case Report

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Abstract

Maras Powder, a smokeless tobacco known as Aztec tobacco globally, is made by mixing dried Nicotiana Rustica leaves with tree ashes. It results in absorption of nicotine that is approximately 10 times higher than that from regular cigarettes. Due to its high nicotine content, it can cause severe toxicity and requires prompt treatment in cases of acute intoxication. This case report describes a 48-year-old male who developed confusion and syncope from Maras Powder use. The symptoms improved within hours, and the patient was fully recovered after 24 hours. Even though there are some cases associated with Maras powder use, this is, to our knowledge, the only documented case of acute symptomatic Maras Powder intoxication in adults. It should be suspected in regions where it is commonly used, with careful attention to patient history.

Keywords: Aztec Tobacco, Maras powder, Maras powder intoxication, nicotine intoxication, smokeless tobacco

Introduction

Smokeless tobacco has become more prevalent worldwide and is known by different names in various cultures (1). A type of smokeless tobacco known worldwide as *Aztec tobacco* and in Turkey as "Maras Powder" (Figure-1 and 2) is generally made by mixing the dried leaves of the *Nicotiana Rustica* plant with the ashes of trees such as oak, although its production methods may vary (2). About a teaspoon of the powder is used either alone or wrapped in cigarette paper and placed on the upper or lower labial mucosa, where it is held for 5 to 10 minutes. The frequency of use varies depending on the level of addiction and the individual's physiology (3). In regular cigarettes, the amount of nicotine absorbed per cigarette ranges from approximately 0.05 to 2.5 mg, whereas the amount of nicotine absorbed from Maras Powder ranges from 7 to 9 mg. This indicates that it delivers nearly 10 times the amount of nicotine compared to cigarettes (4).

Use of Maras Powder has been observed to be more common among married men with low educational and income levels. Additionally, in a study by Akbay and colleagues its use found to be more prevalent among individuals over the age of 46 who have previously smoked. These data suggest that younger users may turn to Maras Powder over time as an alternative to cigarettes to enhance satisfaction (5). It is also known that Maras Powder is preferred as a means of quitting smoking (6). Due to its high nicotine content, Maras Powder is a substance that can cause severe toxicity and must be promptly recognized and treated in cases of acute intoxication (7). In our case report, we aim to describe the clinical presentation of confusion and syncope resulting from Maras Powder use.

Figure 1. Nicotiana Rustica (Wikipedia contributors. Nicotiana rustica (Aztec tobacco, wild tobacco) [Internet]. [cited 2025 Feb 2].

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Figure 2. Maras Powder [Internet]. [cited 2025 Feb 2]

Case Report

A 48-year-old male patient was brought to the emergency department with complaints of syncope and confusion. Apart from a history of coronary angiography three years ago, he had no other medical history and was taking a beta-blocker and acetylsalicylic acid (100 mg). Upon admission, his general condition was moderate, he was confused, and his Glasgow Coma Scale (GCS) score was assessed as 12. During the intervention, he exhibited agitation and purposeless movements.

On physical examination, there was no nuchal rigidity, and Kernig and Brudzinski tests were negative. His body temperature was 36.6°C, heart rate was 78 bpm, respiratory rate was 15 breaths per minute, and blood pressure was 105/60 mmHg. His electrocardiogram (ECG) showed a sinus rhythm. A brain computed tomography (CT) scan revealed no signs of acute hemorrhage and diffusion MRI showed no findings suggestive of acute ischemic stroke.

Laboratory tests showed a pH of 7.24, PCO_2 of 45 mmHg, HCO_3 of 19.5 mmol/L, and lactate of 7.70 mmol/L. Aspartate aminotransferase (AST), alanine aminotransferase (ALT), creatinine, blood urea nitrogen (BUN), and troponin-I levels were within normal limits. His glucose level was 100mg/dl. No findings suggestive of anemia were detected. He and his relatives didn't report any psychiatric history, substance or alcohol use.

However, a history obtained from the patient's relatives revealed that he had been using Maras Powder regularly for the past two weeks to quit smoking. The patient was started on intravenous 0.9% NaCl (1000 mL), 5 mg midazolam, and nasal oxygen at 2 L/min. He was monitored in the emergency department and continued receiving supportive treatment.

Three hours after admission, arterial blood gas analysis showed pH: 7.37, PCO_2: 42 mmHg, HCO_3: 23 mmol/L, and lactate: 0.78 mmol/L. As his blood gas values returned to normal, his confusion improved, and his GCS score increased to 15. After 24 hours of observation, he was discharged from the emergency department.

Discussion

Nicotine intoxication has become more common in recent years, particularly with the increasing use of e-cigarettes, which often include oral nicotine pouches. It exhibits a bimodal distribution, with accidental ingestion being more frequent in children under the age of 10, while intentional use for suicide is more common in adults (8). Similarly, in our country, cases of Maras Powder intoxication among pediatric patients presenting to the emergency department, particularly in Kahramanmaraş and its surrounding areas, have been reported as cases of accidental ingestion (7). In our case, however, intoxication resulted from the unintentional overuse of Maras Powder due to a lack of awareness regarding its high nicotine content.

Acute nicotine intoxication presents with a biphasic pattern due to its ability to both stimulate and inhibit cholinergic receptors. Initially, it may manifest with symptoms such as excessive salivation, nausea, vomiting, diarrhea, and sweating. Additionally, vasoconstriction can

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lead to pallor and increased blood pressure. Tachycardia and, in some cases, cardiac arrhythmias (such as atrial fibrillation) may also occur (9). After a certain period, nicotine-induced desensitization of acetylcholine receptors can result in confusion, somnolence, muscle weakness, and, in severe toxicity, respiratory depression and cardiac arrest (10). The literature also reports delirium associated with oral nicotine gum use and chest pain due to nicotine-induced coronary vasospasm (11). In cases of Maras Powder intoxication observed in the pediatric population in our country, symptoms such as vomiting, somnolence, metabolic acidosis, and convulsions have been reported (7). In our patient, the presence of hypotension and confusion suggested that these symptoms were primarily due to the acetylcholine-related effects of Maras Powder.

Nicotine's half-life varies depending on factors such as gender and genetic influences. In a study by Benowitz and colleagues itranged from approximately 90–150 minutes in non-smokers and 100–200 minutes in smokers (12). Consequently, symptoms tend to resolve quickly, with most patients achieving full recovery within 12 hours (9). In our case, symptom improvement and normalization of blood gas levels were observed within 3 hours, and the patient was discharged in a fully recovered state at the 24-hour mark.

Cotinine, a metabolite of nicotine, is considered the most sensitive and specific biomarker for assessing nicotine exposure. It can be measured in blood, saliva, or urine and has a longer half-life than nicotine (13). However, studies in the literature have shown that cotinine levels do not always correlate with clinical presentation. This discrepancy may be due to the varied causes of nicotine intoxication or liver damage in severe cases affecting nicotine metabolism (10). Additionally, cotinine levels can increase with chronic exposure, meaning that elevated levels do not necessarily indicate acute intoxication. Therefore, routine cotinine measurement in acute poisonings remains debatable (9). There is no specific antidote for Maras Powder/nicotine toxicity. Treatment is primarily symptomatic and supportive. The first priority is to secure the airway and provide respiratory support. Atropine is used to manage parasympathetic symptoms such as excessive salivation, wheezing, and bradycardia. In cases of severe poisoning, endotracheal intubation may be required for airway protection and ventilation support. Seizures should be treated with benzodiazepines. Hypotension is initially managed with fluid boluses and 0.9% NaCl infusion; if unresponsive to volume resuscitation, a vasopressor such as norepinephrine should be administered. Cardiac arrhythmias should be treated according to standard advanced cardiac life support (ACLS) protocols. Nicotine elimination is accelerated in acidic urine, but due to the risks outweighing the benefits, this method is not recommended (9,14). Oral rinsing with water may be advised upon initial presentation. In pediatric patients or cases involving suicidal ingestion of Maras Powder, gastric lavage and activated charcoal administration within the first hours may be beneficial (15). However, in adult intoxication cases, nicotine absorption occurs primarily through the oral mucosa, making these interventions therapeutically ineffective. In our case, due to the patient's agitation upon arrival, intravenous administration of 5 mg midazolam was required.

Conclusion

This case report is, to our knowledge, the only documented case of a smokeless tobacco product or Maras Powder intoxication in the adult population in the literature. Given the broad clinical spectrum of Maras Powder-related intoxication, it should be considered with a high index of suspicion in regions where this product or similar productsare commonly used, and patient history should be carefully assessed. Further research is needed to determine the exact amount of Maras Powder consumption that leads to intoxication, its effects on nicotine and metabolite levels, and the rate at which these changes occur. Additionally, studies should investigate the potential impact of other components in Maras Powder, such as wood ash, on nicotine metabolism.

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Case Report

Journal of Emergency Medicine Case Reports

Single Dose Metformin-Induced Severe Metabolic Acidosis

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Abstract

Metformin, a member of the biguanide family, is one of the most widely used drugs in the treatment of diabetes worldwide. It was first commercialized in 1957, and its use has steadily increased since then. Metformin is recommended as the first pharmacological agent in the treatment of type 2 diabetes. 29-year-old female patient presented to our emergency department due to nausea and vomiting. In her history, she denied diarrhea, urinary symptoms suggestive of urinary tract infection, ingestion of any unusual food, use of any other medications or substances, and reported spending the entire day at home. Upon presentation, the patient was in good general condition, awake, alert, and cooperative. Vital signs were as follows: arterial blood pressure 110/70 mmHg, pulse rate 78 beats/min, respiratory rate 19 breaths/min, oxygen saturation 98%, and body temperature 36.5°C. Physical examination did not reveal any pathological findings. Recently, metformin has been used for weight loss as well as for therapeutic purposes. It is seen that even a single dose of metformin can cause serious side effects. People who use metformin both within and outside the indication should be careful.

Keywords: Lactic acidosis, MALA, metformin, single dose

Introduction

Metformin, a member of the biguanide family, is one of the most widely used drugs in the treatment of diabetes worldwide (1). It was first commercialized in 1957, and its use has steadily increased since then. Metformin is recommended as the first pharmacological agent in the treatment of type 2 diabetes. There is even literature suggesting its use in patients with prediabetes (2). Additionally, it is used to reduce insulin resistance in gestational diabetes and pregnant women with type 2 diabetes (3). However, the American Diabetes Association does not recommend its use as first-line therapy due to its feto-placental transfer (4). Metformin acts through various mechanisms aimed at reducing serum glucose, including increasing insulin sensitivity, antagonizing gluconeogenesis, and enhancing intracellular glucose uptake. However, it also has several significant side effects, including gastrointestinal symptoms (nausea, vomiting, and diarrhea), increased lactate production, decreased lactate clearance, and the potential to induce acidosis (5). Metformin-associated lactic acidosis (MALA) emerges as a cause of high mortality and morbidity (6). MALA occurs in the presence of conditions such as kidney injury, a history of liver or heart failure, or in cases of shock, acute or chronic medical comorbidities (7). Hyperlactatemia arises through several mechanisms. Metformin levels above the therapeutic dose lead to the inhibition of complex 1 in the mitochondrial respiratory chain, resulting in impaired oxidative phosphorylation. Metformin also inhibits pyruvate carboxylase, leading to reduced pyruvate metabolism and increased conversion of pyruvate to lactate. Impaired oxidative phosphorylation and pyruvate utilization result in decreased ATP production. This situation leads to increased AMP levels, which affect the inhibition of fructose-1,6-bisphosphatase, a rate-limiting enzyme in gluconeogenesis. Impaired gluconeogenesis results in decreased hepatic clearance of lactate. Additionally, metformin inhibits glucose-6-phosphatase, disrupting glycogenolysis. Decreased gluconeogenesis and glycogenolysis, reliance on glycolysis, and increased glucose utilization can trigger hypoglycemia observed in some MALA cases (8-10).

Our literature review revealed numerous cases of metabolic acidosis due to metformin overdose or secondary causes such as renal dysfunction and sepsis. However, no case of profound metabolic acidosis due to a single dose of metformin was found in the literature. In this case, we

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aim to highlight the potential for profound lactic acidosis in patients using a single dose of metformin and contribute to the literature. Necessary consents were obtained from the patient for this case report, and they were adequately informed about the process.

Case Report

29-year-old female patient presented to our emergency department due to nausea and vomiting. In her history, she denied diarrhea, urinary symptoms suggestive of urinary tract infection, ingestion of any unusual food, use of any other medications or substances, and reported spending the entire day at home. Upon presentation, the patient was in good general condition, awake, alert, and cooperative. Vital signs were as follows: arterial blood pressure 110/70 mmHg, pulse rate 78 beats/min, respiratory rate 19 breaths/min, oxygen saturation 98%, and body temperature 36.5°C. Physical examination did not reveal any pathological findings.

The patient had no chronic illnesses or surgical history other than cesarean section. She did not take any regular medications. Symptom-directed treatment was initiated after physical examination (intravenous infusion of 500 ml saline containing 40 mg pantoprazole and 4 mg ondansetron), and blood tests were requested. Before the test results were fully available, the patient expressed a desire to be discharged due to clinical improvement. After receiving appropriate information, the patient left the hospital. Laboratory results of the patient's application are given in Table-1. Venous blood gas analysis performed upon arrival showed pH 7.45, PCO_2 27 mmHg, HCO_3 18.4 mmol/L, and lactate level 4.9 mmol/L.

Later the same day (approximately 8 hours later), the patient was brought back to the emergency department via emergency medical services with decreased level of consciousness. Paramedics reported administering 50 mg intravenous dextrose due to a blood sugar level of 30 mg/dL. The patient was evaluated while dextrose infusion continued. On repeat physical examination, the Glasgow Coma Scale was 8, indicating decreased orientation and cooperation. Apart from this, no pathological findings were observed on examination. Vital signs at arrival were arterial blood pressure 90/50 mmHg, pulse rate 98 beats/min, respiratory rate 25 breaths/min, oxygen saturation 99%, and body temperature 36.5°C. Electrocardiography revealed normal sinus rhythm.

Following dextrose therapy, the patient's blood sugar levels rose, consciousness improved, and orientation and cooperation began to normalize, reaching a Glasgow Coma Scale of 15 after some time. Venous blood gas analysis performed upon arrival showed pH 6.96, PCO_2 22 mmHg, HCO_3 4.7 mmol/L, lactate level 15.6 mmol/L, and blood sugar level 33 mg/dL. Thirty minutes after regaining consciousness, repeat blood gas analysis showed worsening metabolic acidosis and increased lactate levels. Based on the patient's history and investigations, toxicological evaluation was initiated.

According to the patient's history, she had taken a single 1000 mg metformin tablet before breakfast that morning with the intention of losing weight. Apart from this, there was no history of alcohol or herbal medicine use, suicidal attempts, or ingestion of any other substances. During follow-up, venous blood gas analysis results were obtained (Table-2). Hourly urine output was normal, and renal function tests were within normal limits. Urine analysis was negative for ketones and showed no signs of infection. Serum ethanol and paracetamol levels were negative. Brain computed tomography did not reveal any pathology explaining the patient's altered mental status. Apart from metformin use, no other etiological cause was identified, and metformin-induced severe metabolic acidosis was considered primary in the etiology of deep metabolic acidosis.

Due to ongoing metabolic acidosis and lactic acidosis during follow-up, the patient received a 50 mEq sodium bicarbonate (Na^+ HCO_3) bolus and maintenance therapy at 20 mEq/hour. Additionally, she received hourly 500 cc saline infusion and 100 cc/hour 5% dextrose infusion. Preparation for hemodialysis began due to the possibility of persistent severe metabolic acidosis. However, as the acidosis began to decrease with medical treatment, hemodialysis was deferred upon normalization of blood gas parameters at the 8th hour of admission.

The patient remained stable during the 12-hour observation period in the emergency department, with stable vital signs and normalization of blood gas parameters. Subsequently, she was admitted to the internal medicine service for further monitoring and investigations. On the 2nd day of hospitalization, no additional etiological cause was found, and the patient was discharged in good condition.

Table 1: Laboratory results of the patient at the time of first admission

WBC	HBG	PLT	Glukoz	Üre	Cr	ALT	AST	Na^+
5,54	11,4	263	80	24	1,0	15	34	140
K^+	**Cl^-**	**Amilaz**	**Lipaz**	**GGT**	**CRP**	**Aptt**	**INR**	**HsTrop**
5,0	111	51	29	12	0,39	18	1,09	Negatif

WBC: Leukocyte ($x10^3/mm^3$); **HGB:** Hemoglobin (mg/dL); **PLT:** Platelet ($x10^3/mm^3$); **Glucose, Urea** (mg/dL); **Cr:** Creatinine (mg/dL); **ALT:** Alanine transaminase (IU/L); **AST:** Aspartate transaminase (IU/L); **Na^+**: Sodium (mEq/L); **K^+**: Potassium (mEq/L); **Cl^-**: Chlorine (mEq/L); **Amylase, Lipase** (IU/L); **GGT:** Gamma Glutamyl transferase (IU/L); **CRP:** C-reactive protein (mg/dL); **INR:** International Normalized Rate; **HsTrop:** High sensitivity troponin

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Table 2: Venous blood gas follow-up results of the patient

Parametre	1. Admission	2. Admission	2. Admission 30th min	2. Admission 60th min	2. Admission 2nd h	2. Admission 4th h	2. Admission 8th h	2. Admission 12th h
pH	7.45	6.95	6.75	6.84	6.97	7.11	7.42	7.45
PCO_2 (mmHg)	27	22.2	21	30.9	35	30	28.7	27
HCO_3 (mmol/L)	18.4	4.7	4.1	5.1	7.9	14	18.2	19.3
Laktate(mmol/L)	4.8	15.8	20.8	22.3	19.9	8.2	4.9	3.3
Na^+ (mmol/L)	141	146	143	142	143	140	138	139
K^+ (mmol/L)	4.1	4.1	4.0	4.5	5.1	4.2	3.5	3.6
Cl^-(mmol/L)	108	116	111	103	102	104	101	100
Glukoz (mg/dL)	80	33	115	272	279	185	176	124

PCO_2: Partial Carbon Dioxide; **HCO_3**: Bicarbonate; **Na^+**: Sodium (mEq/L); **K^+**: Potassium (mEq/L); **Cl^-**: Chlorine (mEq/L); **min**: Minute; **h**: Hour

Discussion

MALA is a rare complication of metformin use. In the literature, it is often associated with high single doses or therapeutic doses in patients with renal or hepatic dysfunction (11,12). Lactic acidosis in patients taking metformin can occur in three different clinical scenarios: metformin-induced, metformin-associated, and unrelated to metformin (13). In cases of metformin-induced lactic acidosis, symptoms typically manifest within 6-12 hours after ingestion, and hemodialysis is considered the gold standard treatment in cases involving renal insufficiency or overdose (14).

In our case, the patient took a single therapeutic dose of metformin without any renal impairment, thus treatment did not require hemodialysis. However, hemodialysis would have been considered if there had been no improvement in severe metabolic acidosis. Considering metformin's half-life of 18 hours, the patient's recovery period aligns with this timeframe (15). Mild compensated metabolic lactic acidosis was observed in our patient during the initial presentation. History of alcohol and substance use was negative, and no chronic illnesses or regular medication use were reported. During the second presentation, the presence of severe metabolic acidosis, elevated lactate levels, and hypoglycemia supports the clinical association with metformin. Therefore, clinicians should consider that even a single therapeutic dose can lead to severe metabolic and lactic acidosis in cases presenting with severe metabolic acidosis (16).

This case presentation could serve as an important guide indicating that even a single dose of metformin can induce severe metabolic acidosis in patients. However, a significant limitation of our study is the inability to measure metformin levels in the patient.

Conclusion

Recently, metformin has been used for weight loss as well as for treatment purposes. It is seen that metformin is not a very innocent drug and even a single dose of metformin can cause serious side effects. People who use metformin both on and off-label should be careful. It should not be forgotten that life-threatening side effects may occur.

Patient consent form-Ethics

The case report has written in an anonymous characteristic, thus secret and detailed data about the patient has removed.

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Case Report

Journal of Emergency Medicine Case Reports

Hair-Thread Tourniquet Syndrome: A Case Report

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Abstract

Hair-thread tourniquet syndrome (HTTS) is a rare condition that occurs when hair strands wrap around body appendages, potentially leading to serious complications such as tissue loss. In this case report, we aim to present an HTTS case identified in a 2-year-old male child who presented to the emergency department with penile pain and dysuria. A two-year-old male patient presented to the emergency department with complaints of penile pain, redness, and burning sensation during urination that had persisted for 24 hours. Physical examination revealed edema, erythema, and circular constriction on the penile shaft. Detailed examination showed a hair strand wrapped around the penis shaft. The hair was removed by cutting under local anesthesia. Doppler ultrasonography confirmed normal circulation. The patient was discharged after four hours of observation as his complaints subsided. In infants with unexplained restlessness, one should be cautious about HTTS, and it should not be forgotten that areas where this syndrome may occur should be carefully examined.

Keywords: Hair-thread tourniquet syndrome, pediatric emergency, penis

Introduction

Hair-thread tourniquet syndrome (HTTS) is a rare, preventable condition that occurs when a body appendage is tightly wrapped with a hair strand or thread-like material, resulting in circulatory disturbance. It typically affects the genital area, fingers, and toes. Hair tourniquet syndrome, which occurs when hair or hair-like material becomes entangled around the coronal sulcus of the penis, is characterized by progressive circulatory disturbance. This condition can lead to serious complications, including glans necrosis. Other risk factors contributing to the development of the syndrome include circumcision, wearing tight clothing, poor hygiene, and low socioeconomic status. Early diagnosis and treatment of HTTS is vital because delayed diagnosis can result in serious complications such as ischemia, tissue necrosis, and even auto-amputation. However, complete recovery is possible with early diagnosis and appropriate treatment (1-3). In this case report, we aimed to present a rare case of hair tourniquet syndrome in a child who presented with penile pain, redness, and dysuria complaints.

Case Report

A 2-year-old male patient was brought to the emergency department by his mother with complaints of penile pain, redness, and burning sensation during urination. According to his history, the complaints started approximately 24 hours ago, gradually intensified, and his restlessness increased in the last few hours. The patient had no significant medical history and no recent history of infection or trauma.

On physical examination; the patient was in good general condition, conscious, oriented, and cooperative. His vital signs were measured as follows; body temperature: 36.8°C, pulse: 98/min, respiratory rate: 22/min, blood pressure: 95/60 mmHg, oxygen saturation: 99%. Significant edema, erythema, and circular constriction were detected on the penile shaft. Detailed examination revealed the presence of a single hair strand completely encircling the penile shaft (Figure-1). The patient was diagnosed with hair-thread tourniquet syndrome. Under local anesthesia, the hair strand was carefully cut and removed using a scalpel and clamp. Post-procedure evaluation showed no signs of deep cuts or necrosis in the penile tissue. Doppler ultrasonography performed to assess circulation showed normal blood flow.

The patient was kept under observation in the emergency department for 4 hours after the procedure. During this time, significant improvement in pain and restlessness was observed. The patient, who developed no problems during the follow-up period and had normal urinary output, was discharged with necessary recommendations given to the family.

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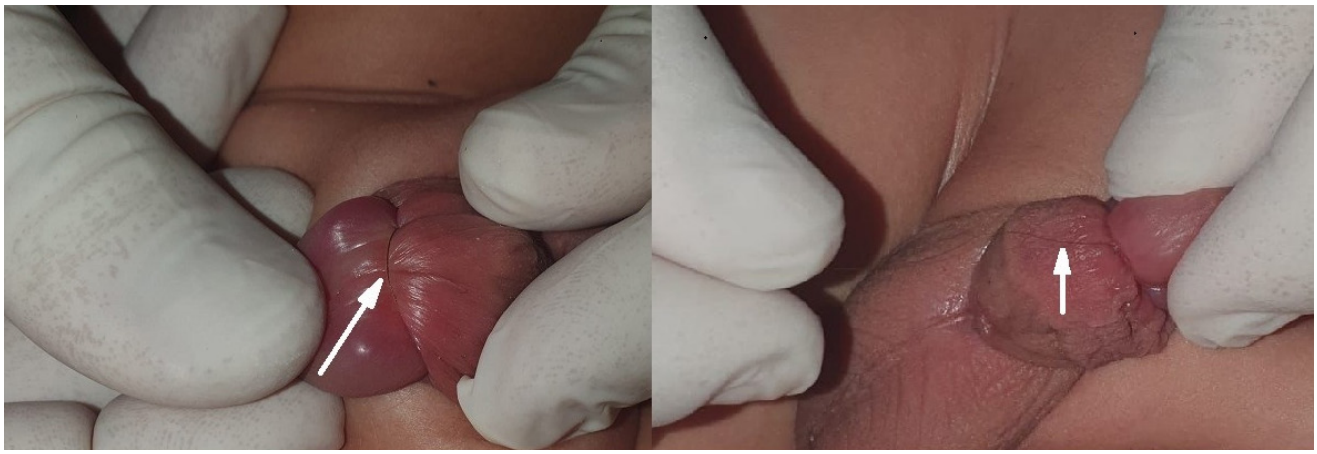

Figure 1. A single strand of hair completely surrounding the shaft of the penis

Discussion

Hair-thread tourniquet syndrome (HTTS) is an important clinical condition that particularly occurs in the newborn and infant period and can lead to serious complications if not diagnosed early. (4)

This syndrome is particularly seen in newborns and young children, and the affected anatomical regions vary according to age. In approximately 44.2% of cases reported in the literature, the penis is affected, in 40.2% the toes, and in 8.6% the fingers. When analyzed by age groups, fingers are generally affected up to 1.5 years of age, penis between 4 months and 6 years, and the genital area between 7-13 years (3). In our case, consistent with these data in the literature, penile hair tourniquet syndrome was observed in a 2-year-old male child.

Risk factors for the syndrome include telogen effluvium in postpartum mothers, poor hygiene conditions, low socioeconomic status, and tight clothing (5). Knowledge of these risk factors and informing families about them is important in taking preventive measures. Careful examination of extremities and the genital area, especially in the newborn period, is vital for early diagnosis. In our case, the family was of foreign nationality, had low income, and poor hygiene conditions.

Treatment approaches vary depending on the affected area and degree of tissue damage. Non-surgical methods include depilatory creams, mineral oil application, and hair/thread dissolving solutions, while advanced cases may require longitudinal incision or surgical removal of the tourniquet material (4, 6). Although the prognosis is generally good with early diagnosis and appropriate treatment, serious complications such as ischemia, tissue necrosis, auto-amputation, and secondary infections can be seen in delayed cases (5). In our case, successful treatment was achieved without any complications due to early diagnosis and appropriate intervention.

HTTS shows that it is not sufficiently recognized by physicians due to its rare occurrence, and therefore delays in diagnosis and treatment may occur. (1) Especially in pediatric patients presenting with unexplained genital pain and restlessness, keeping this syndrome in mind and performing careful physical examination is critically important. Early diagnosis and treatment in HTTS is crucial to prevent complications. If cases are not appropriately treated in a timely manner, serious clinical conditions such as finger, penis, or clitoris amputation may develop (2).

Conclusion

In infants with unexplained restlessness, one should be cautious about HTTS, and it should not be forgotten that areas where this syndrome may occur should be carefully examined.

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Case Report

Journal of Emergency Medicine Case Reports

Bilateral Occipital Condyle Fracture with Clivus Fracture: Case Report

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Abstract

Occipital condyle and clivus fractures caused by trauma are rare injuries that are difficult to diagnose. Only a few publications have reported cases involving both fractures. With the widespread use of high-resolution computed tomography in recent years, the diagnosis of these fractures has become more feasible; however, their clinical presentation and treatment approach remain uncertain for clinicians. The concurrent occurrence of occipital condyle and clivus fractures has recently been incorporated in to the Anderson and Montesano classification system for occipital condyle fractures. In this paper, we aim to discuss the clinical presentation and treatment strategies of a 39-year-old female patient who was brought to our hospital with bilateral occipital condyle and transverse clivus fractures following a pedestrian traffic accident, in light of the existing literature.

Keywords: Clivus, fracture, occipital condyle

Introduction

Traumatic injuries of the skull base are rare but have high mortality rates. Occipital condyle fractures have been recognized as complex and poorly understood lesions by clinicians since they were first described by Bell in 1817. Fractures of the occipital condyle and clivus are uncommon and difficult to diagnose. They may be overlooked in routine X-rays and computed tomography (CT) scans (1).

Clinically, due to the numerous neural and vascular structures located at the craniocervical junction, these fractures may present with various symptoms and are associated with high mortality rates (1,2). Diagnosis is generally established in patients with brain stem findings, rhinorrhea, orthrough post-mortem examinations. However, the coexistence of these lesions has been reported in only a fewcases (3-9). In patients with high-velocity head and cervical trauma, careful evaluation of this region facilitates the diagnosis of occipital condyle and clivus fractures (2). With the recent widespread use of high-resolution and 3D CT imaging, the number of reported cases has increased (9).

In this paper, we present a rare case of bilateral occipital condyle fracture complicated by an inferior transverse clivus fracture, which has recently been added to the classification of occipital condyle fractures. Including our case, a total of eight concomitant clivus and occipital condyle fractures have been reported in the literature to date (8).These types of fractures necessitate intricate clinical decision-making and are challenging to diagnose. This study aims to discuss the clinical presentation and treatment approaches of this rare fracture combination in light of the existing literature.

Case Report

A 39-year-old female patient, who sustained injuries due to a high-energy pedestrian traffic accident, was brought to the emergency department with assisted ventilation using an ambu bag and secured with a philadelphia cervical collar. Upon initial examination, the patient was unconscious (GCS: 3) and was immediately intubated. A blunt scalp laceration approximately 5 cm in diameter was noted on the left side of the vertex, and the patient's vital signs were stabilized.

High-resolution CT revealed an acute subdural hematoma in the left parietal region and extensive subarachnoid hemorrhage (Figure-1). Following high-resolution CT imaging and 3D reconstruction, fractures extending to both occipital condyles and involving the clivus were identified (Figures-2, 3, 4, and 5). The patient also had fractures of the left hand and ankle, for which orthopedic specialists applied a splint.

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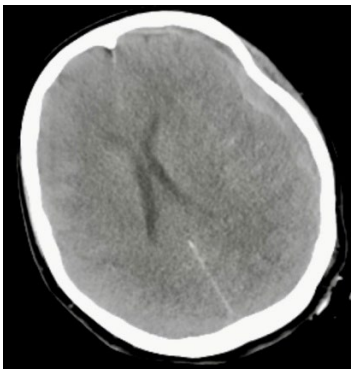

Figure 1. Axial brain CT showing a subdural hematoma located in the left parietal region

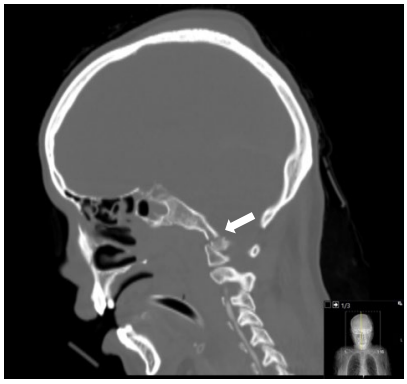

Figure 2. Sagittal brain CT demonstrating a transverse fracture of the clivus (white arrow)

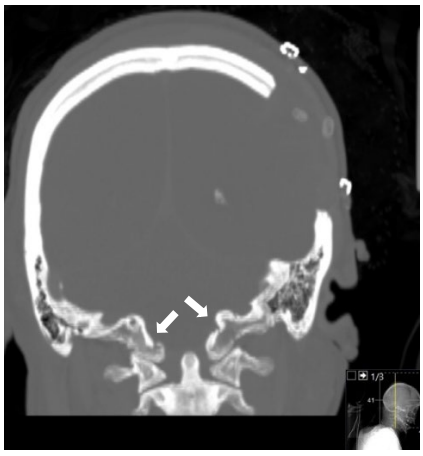

Figure 3. Coronal CT scan showing a postoperative left parietal craniectomy defect and fractures of both occipital condyles (white arrows)

Due to the acute subdural hematoma, the patient underwent emergency surgery, and the hematoma was evacuated via left parietal craniectomy (Figure-3). Postoperatively, the patient was transferred to the intensive care unit with a rigid cervical collar and placed on mechanical ventilation. Cerebral anti-edema therapy was initiated. At the 65th hour of hospitalization, the patient developed cardiac arrhythmias, followed by hypotension and circulatory arrest, ultimately leading to death.

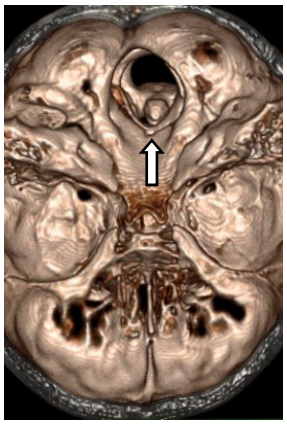

Figure 4. High-resolution 3D reconstructed CT image showing a fracture line extending from the clivus to both occipital condyles when viewed from the internal surface of the cranium (white arrow)

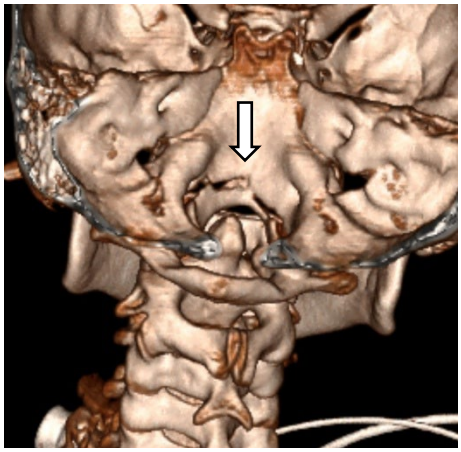

Figure 5. High-resolution 3D reconstructed CT image showing occipital condyle and clivus fractures when viewed from a posterior perspective inside the cranium (white arrow)

Discussion

The simultaneous occurrence of bilateral occipital condyle and clivus fracturesis rare (1). Occipital condyle fractures are difficult to diagnose. High-resolution reconstructed CT imaging is considered the gold standard for diagnosis (9,10). The shape of the fracture, its extent, the degree of displacement, and its impact on treatment planning are crucial factors. The routine use of multi-slice CT in recent years has led to an increase in reported cases of occipital condyle fractures. MRI is of limited value in evaluating the functional integrity of the upper cervical ligamentous complex, as it is hindered by surrounding bone edema and hematoma (11). Although flexion-extension cervical radiographs can indicate occipitocervical instability, this method carries significant risks.

To date, seven cases of concomitant clivus and occipital condyle fractures have been reported in the literature; here, we present the eighth case (3-8).

Occipital condyle fractures are generally classified according to the Anderson and Montesano system,

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specifically under type III (12). However, the underlying mechanism of combined occipital condyle and clivus fractures remains unclear (3,11). In 2022, Carriço et al. proposed a four thtype of occipital condyle fracture, suggesting that these injuries result from axial loading and compression mechanisms. Based on the patient's accident video, clinical evaluation, and scene reconstruction, we believe that a blunt force impact on the left side of the vertex caused axial loading, leading to fractures of both the clivus and occipital condyles. Carriço et al. classified occipital condyle fractures associated with clivus avulsion fractures as Type IV A, while fractures involving occipital condyle fractures with comminuted fractures of the lower clivus were designated as Type IV B. Both Type IV injuries are considered highly unstable (1). Based on this classification, we categorized our patient's injury as Type IV B.

Since occipital condyle fractures are infrequently diagnosed, no widely accepted treatment guidelines exist (9,11). Treatment generally involves either external immobilization with devices such as cervical collars or surgical stabilization. Patients with bilateral occipital condyle fractures and concomitant clivus fractures have a higher risk of craniocervical instability, which increases mortality risk. This suggests that more rigid fixation techniques may be necessary (3). Carriço et al. advocated surgical fixation for both Type IV A and IV B injuries (1). However, among the previously reported seven cases, three were treated surgically and three were managed with conservative methods. One case was diagnosed post-mortem (3-8). At the craniovertebral junction, stabilization is supported not only by the bony structures but also by the occipital condyle joint capsules, alar ligaments, and the tectorial membrane. When ligamentous integrity is preserved, external immobilization is generally considered sufficient to ensure stability. In the three cases managed conservatively, treatment involved halo or cervical collar immobilization for upto 14-16 weeks. Fluoroscopy-guided traction testing is recommended to determine the appropriate timing for terminating conservative treatment (8,9). No standardized surgical approach exists for operative cases. Surgical options range from occipital-C1-C2 fusion to C1 laminectomy without fusion, which has been reported as a successful treatment approach (11). In our case, a rigid cervical orthosis was applied; however, the patient unfortunately passed away before the assessment of craniovertebral junction integrity could be completed.

Occipital condyle and clivus fractures carry significant morbidity and mortality risks. The mortality rate for occipital condyle fractures has been reported as 16.1%, while that for clivus fractures reaches upto 80% (3,13). These injuries can lead to fatal outcomes due to brain stem trauma, cranial nevre deficits, and vertebrobasilar vascular injuries (9). Imamura et al. reported a case of a 25-year-old patient who sustained trauma in a motor vehicle accident. Post-mortem examination revealed bilateral occipital condyle fractures extending into the lower clivus. The authors attributed the patient's death to associated medullaoblongata and basilar artery injuries (14). Similarly, we believe that our patient's fatal outcome resulted from brain stem edema secondary to vertebrobasilar injury.

Conclusion

In patients who sustain high-energy axial trauma, the concurrent presence of occipital condyle and clivus fractures should be considered. High-resolution computed tomography and 3D reconstruction imaging are recommended for definitive diagnosis. Early diagnosis and appropriate treatment planning for such fractures, which may lead to craniocervical instability, can be life-saving. The choice between conservative and surgical treatment should be determined based on the patient's overall condition and the stability of thelesion.

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Case Report

Journal of Emergency Medicine Case Reports

An Unexpected Result of Increased Cosmetic Anxiety: Botulinum Toxin Side Effect

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Abstract

Botulinum toxin inhibits acetylcholine release at the neuromuscular junction, thereby reducing muscle contraction. Although it was initially used therapeutically to treat strabismus, its clinical role has expanded rapidly over the years and its range of use has expanded to include the treatment of various musculoskeletal, neurological disorders and dermatologic and cosmetic areas. A previously healthy 34-year-old woman presented to the emergency department with complaints of diplopia, slurred speech and generalized muscle weakness that started about 4 hours ago. The patient's medical history is negative for any surgical interventions. It was learned that the patient had her 4th botox procedure 6 days ago for wrinkles on the glabella, nasal tip and around the eyes. Following a 12-day inpatient stay, the patient was discharged to outpatient care as their symptoms improved. Treatment of side effects of botulinum toxin primarily involves administration of botulinum antidote, close neurologic and respiratory monitoring and supportive care. This case report presents a unique instance of systemic manifestations following facial botulinum toxin administration, diverging from the predominantly reported localized adverse effects in existing literature, thereby highlighting a potentially underrecognized spectrum of clinical presentations.

Keywords: Botulinum, deglutition disorders, diplopia

Introduction

The bacterium Clostridium botulinum produces botulinum neurotoxin (BoNT), the world's most powerful poison. BoNT blocks acetylcholine release at nerve terminals. This interrupts the communication from the nerve to the muscle fiber and reduces muscle contraction. Since its Food and Drug Administration (FDA) approval in 1989, BoNT has been used in many different areas beyond the conditions for which it was initially approved, such as facial movement disorders and strabismus (1). The most frequently reported adverse effects are local effects such as erythema, edema, bruising and are self-limiting. Serious side effects such as muscle weakness and allergic reactions may also be observed but are very rare. The incidence of complications increases in a dose-dependent manner. Complications are approximately 33 times more likely to occur in therapeutic cases compared to cosmetic cases (2). Reasons for side effects include the use of elevated doses of toxin than recommended or the use of unlicensed products (3). Treatment of botulinum toxin side effects primarily includes administration of botulinum antidote, close neurologic and respiratory monitoring and supportive care. Individuals suspected of having botulism should be hospitalized in the Intensive Care Unit (ICU) for close monitoring of their vital signs and rapid assessment of their need for antitoxin treatment. The most important step of treatment is antitoxin administration and the earlier it is administered, the better the clinical response (2). As an anticholinesterase agent, pyridostigmine prolongs the action of acetylcholine by inhibiting acetylcholinesterase at synaptic sites, thus improving muscle strength in conditions characterized by impaired neuromuscular transmission (4). A retrospective review of patients experiencing significant adverse events, including dysphagia and dyspnea, after head and neck botulinum toxin injections demonstrated that pyridostigmine was well-tolerated and resulted in notable symptom improvement, indicating its potential in modulating more widespread effects (5). The purpose of this case report is to emphasize side effects of botulinum toxin, the use of which is increasing especially in the cosmetic field today, and its systemic effects and treatment process besides its local effects in the application area.

Case Report

A previously healthy 34-year-old woman presented to the emergency department with complaints of diplopia, slurred speech and generalized muscle weakness that started about 4 hours ago. The patient's medical history is negative for any surgical interventions. During the medical history taking,

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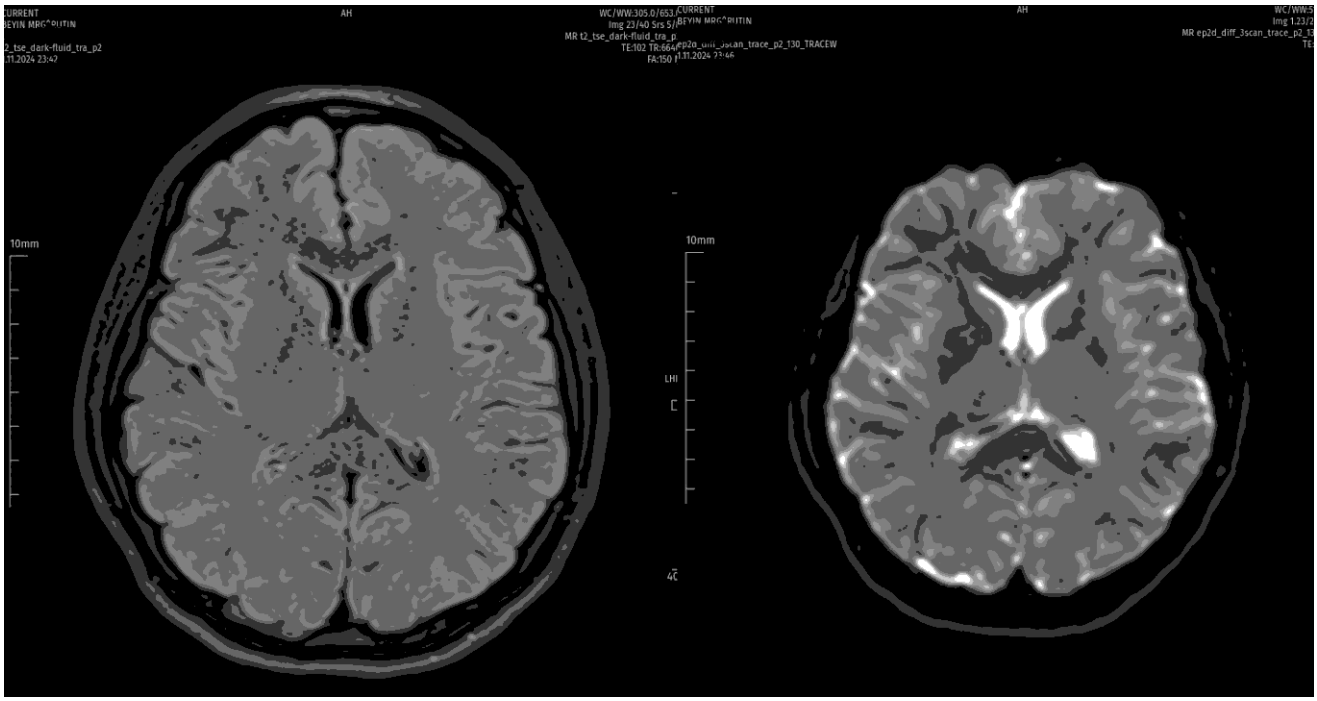

Figure 1. No pathological findings in patient's MRI

it was learned that she underwent botox procedure 6 days ago for wrinkles on glabella, tip of the nose and around the eyes. It was stated that this was the patient's fourth botox procedure. The patient's blood pressure was 117/76 mmHg, pulse rate: 96/min, saturation: 99%, temperature: 36.6 oC. No pathologic findings were detected on electrocardiogram (ECG). To comprehensively evaluate the patient's atypical presentation of systemic neurological symptoms following botulinum toxin administration, and to rigorously exclude alternative etiologies encompassing autoimmune neuromuscular disorders such as Myasthenia Gravis and inflammatory polyradiculopathies like Guillain-Barré syndrome, as well as acute cerebrovascular events including ischemic or hemorrhagic stroke, a thorough neuroimaging workup, including computed tomography (CT) and/or magnetic resonance imaging (MRI) (Figure-1) of the central nervous system, was promptly undertaken. Cranial imaging and blood tests were ordered to exclude a differential diagnosis; no pathology was found in these tests. The patient was consulted to a neurologist with a prediagnosis of BoNT side effect. Botulinum antidote was administered. Symptomatic supportive treatment was started. The patient was transferred to the ICU. 4*60 mg pyridostigmine was added to the intensive care unit treatment. On the second day of intensive care unit follow-up, dysphagia developed with progression in speech disorder. The patient's nutritional support continued with nasogastric tube. The intensive care period continued for 5 days and swallowing function improved. After the swallowing function improved, ward follow-up was initiated. After the patient's dysphagia and diplopia were completely resolved and his speech disorder improved to a great extent, he was discharged after 7 days of ward follow-up and was discharged to outpatient care.

Discussion

The occurrence of pronounced systemic bulbar symptoms, specifically progressive dysphagia and diplopia, after routine facial botulinum toxin administration, as presented in this previously healthy 34-year-old woman, represents a rare observation in contrast to the prevailing literature focused on localized effects. The subsequent positive response to pyridostigmine therapy suggests a crucial role for cholinergic modulation in managing these less common systemic manifestations.

While local adverse events following cosmetic botulinum toxin administrations are commonly reported, systemic adverse events are considered rare. However, a recent systematic review (6) indicates the potential for systemic symptoms such as fatigue and muscle weakness. In rare instances, distant spread of the toxin has been associated with severe systemic effects, including dysphagia (7). This case report presents a novel instance of an unusual systemic reaction within the existing literature.

BoNT is a neurotoxic protein; it causes flaccid paralysis by inhibiting the release of acetylcholine neurotransmitter from axon terminals. Botulinum inhibits the exocytosis of acetylcholine (ACH) in the cholinergic nerve endings of motor nerves, preventing it from binding to the membrane

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where the neurotransmitter can be released in the vesicle where acetylcholine is stored. BoNT exerts this effect through endopeptidase activity against SNARE (Soluble N-ethylmaleimide-sensitive factor Attachment protein REceptor) proteins (the protein required for ACH vesicle binding to the presynaptic membrane). The presence of acetylcholine stores in the synaptic cleft acts as a buffer that delays the effects of neurotoxins and therefore toxicity may take 24-48 hours to occur. In our case, toxicity symptoms appeared on the 6th day. The paralytic effect of neurotoxin continues until the axonal regeneration process required for the repair of damage to the neuromuscular junction is completed, and this period is generally estimated to be between 2-6 months (8, 9). In the case we presented, recovery was observed in a period of approximately 2 weeks from the onset of toxicity findings. This was probably because we started antidote treatment early after the toxicity findings were observed. Although it was initially used therapeutically to treat strabismus, its clinical role has expanded rapidly over the years and its range of use has expanded to include the treatment of various head, neck, gastrointestinal, urogenital, musculoskeletal, neurologic disorders and dermatologic and cosmetic areas (2). In the present case, it was detected that BoNT was used for cosmetic purposes rather than therapeutic purposes. According to the literature, our case is an example of the less expected side effects of Botox in cosmetic use compared to its therapeutic use. Botulinum toxin administration has demonstrated a favorable safety profile and high patient satisfaction rates. Pain, edema, erythema, ecchymosis and short-term hyperesthesia may be observed after botulinum toxin injection. These are general local side effects that may also occur after other drug injections (2). The most common complication of botulinum toxin administered to the glabella region is ptosis of the upper eyelid (blepharoptosis). The most prevalent side effects of BoNT injections in the lateral periorbital region are diplopia, transient strabismus, ectropion, lagopthalmus and xerophthalmia (2). The same side effects were observed in our case in accordance with the literature. Hoarseness, dysphagia and neck weakness may occur after BoNT injection. Dysphagia, xerostomia, neck weakness and dysarthria may occur as a result of high doses of botulinum toxin or its administration into deeper muscles. In a study, it was reported that dysphagia and odynophagia occurred in a significant proportion of patients with cervical dystonia who received BoNT injection (10). These side effects are more common in elderly patients. The reason is that higher doses of toxin are required in elderly patients and the toxin diffuses easily into the deeper muscles of the neck because of the decreased soft tissue in the neck. These complications take at least 3-4 weeks to heal and are very rare (10). In our case, these side effects were observed in a young patient and healed in a time compatible with the literature data.

Conclusion

This case report presents a unique instance of systemic manifestations following facial botulinum toxin administration, diverging from the predominantly reported localized adverse effects in existing literature, thereby highlighting a potentially underrecognized spectrum of clinical presentations.

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Declaration of Helsinki

This study was conducted in accordance with the principles outlined in the World Medical Association's (WMA) Declaration of Helsinki - Ethical Principles for Medical Research Involving Human Subjects.

Written informed consent to publish this case report (including all clinical information and any related images) was obtained from the patient.