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Contents

Case Report

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A Rare Cause of Small Bowel Obstruction: Congenital Peritoneal Encapsulation

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Abstract

Congenital peritoneal encapsulation (CPE) is a rare condition in which part or all of the small intestine is surrounded by an accessory peritoneal layer congenitally. Although it rarely causes small bowel obstruction, it is usually asymptomatic and the diagnosis is mostly made incidentally during surgery or autopsy. In this presentation, it is aimed to remind CPE, which is a rare disease, with a case report. A 41-year-old male patient presented to the emergency department with diffuse and cramping pain lasting for approximately 8 hours. He had nausea and vomiting. No gas or faeces output for 72 hours. Hemodynamically stable patient had normal hemogram and biochemical parameters and afebrile. After surgery, the patient was discharged uneventfully. CPE should be considered in small bowel obstructions of unexplained etiology. Laparoscopic evaluation is effective in diagnosing CPE.

Keywords: Congenital peritoneal encapsulation, small bowel, emergency, surgery

Introduction

Congenital peritoneal encapsulation (CPE) is a rare condition in which part or all of the small intestine is surrounded by an accessory peritoneal layer congenitally (1). Although it rarely causes small bowel obstruction, it is usually asymptomatic and the diagnosis is mostly made incidentally during surgery or autopsy (2). Abdominal cocoon (AC) and sclerosing encapsulated peritonitis (SEP) are other entities that cause peritoneal encirclement in the small intestine. While CPE is congenital, AC and SEP are acquired diseases (3,4). In this presentation, it is aimed to remind CPE, which is a rare disease, with a case report.

Case Report

A 41-year-old male patient presented to the emergency department with diffuse and cramping pain lasting for approximately 8 hours. He had nausea and vomiting. No gas or faeces output for 72 hours. Hemodynamically stable patient had normal hemogram and biochemical parameters and afebrile. On physical examination, there was abdominal distension. There was tenderness in the right abdominal quadrants on palpation. On auscultation, bowel sounds were locally hyperactive. He had no history of chronic disease or previous surgery. Abdominal pain and ileus attacks not exceeding 24 hours have occurred once or twice a year in the last three years, but they have resolved spontaneously or with conservative treatment. Air-fluid levels in the small intestines were seen in the abdominal X-ray. Abdominal ultrasonography (USG) revealed marked dilated small bowel loops and a small amount of free fluid between these loops. Abdominal computed tomography (CT) showed dilated abdominal small intestines and findings consistent with obstruction (figure 1). The patient was hospitalized with the diagnosis of ileus. Decompression was performed with a nasogastric tube. It was decided to perform diagnostic laparoscopic surgery for the patient who did not respond to 24-hour observation and medical treatment. A signed consent form was obtained from the patient for all procedures to be performed. Laparoscopic examination revealed a thin membrane covering the small intestine from the terminal ileum to the middle of the jejunal segment on the right side of the abdomen (figure 2). It was separated from the abdominal wall laparoscopically. Due to the presence of extensive bands between the small bowel loops, open surgery was performed. All adhesions were separated, the small intestines were released from the pressure of the accessory peritoneum and placed in the abdomen. The surgery took about 100 minutes and was completed without any problems. The patient was discharged without complications on the 6th postoperative day.

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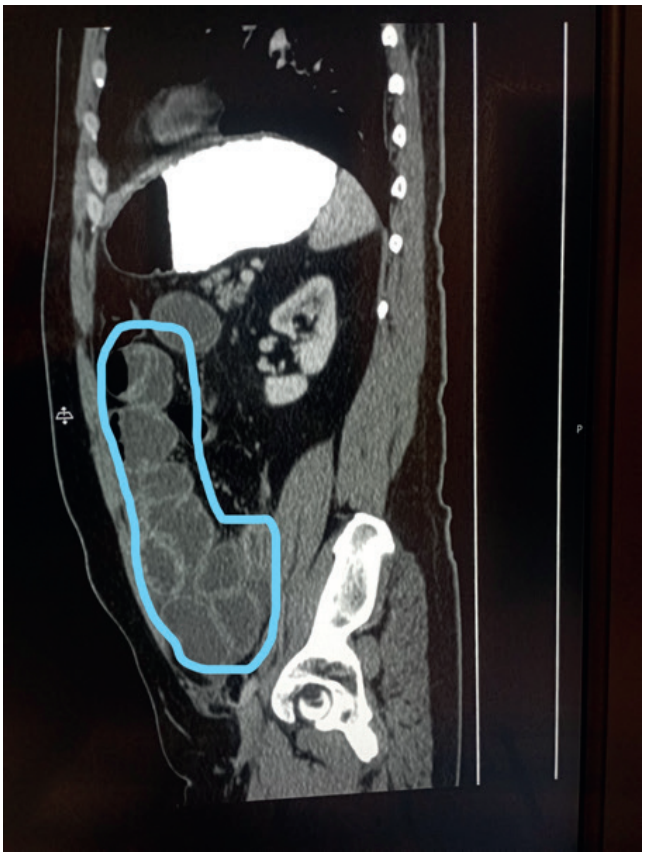

Figure 1. Abdominal CT shows dilated small bowel loops (blue stripe)

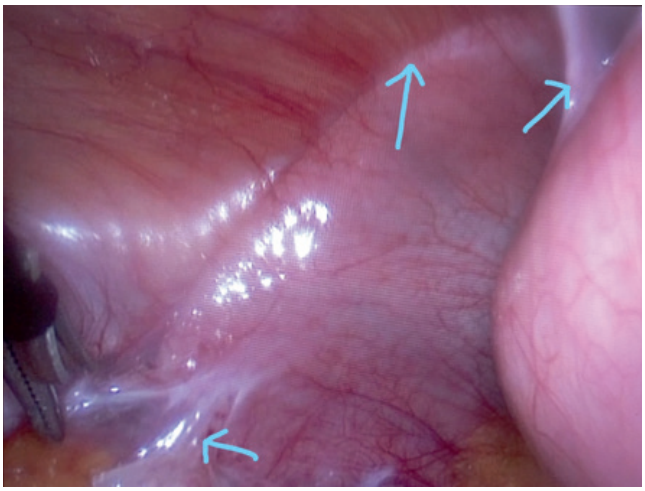

Figure 2. Peritoneal membrane surrounding the small intestine in diagnostic laparoscopy

Discussion

CPE was first described by Cleland in 1868 (5). In the 12th week of pregnancy, due to the abnormal rotation of the small intestines, it leads to the formation of accessory membranes that completely or partially surround the ans. During this rotation, the yolk sac, which should remain on the umbilical pedicle, migrates with the small intestines. The accessory peritoneum extends from the ascending and descending colon laterally, superiorly to the transverse colon, inferiorly to the pelvic parietal peritoneum, and encapsulates the small intestine (6,7).

AC and SEP are two other entities that surround the small intestine, causing capsule formation. However, these are acquired diseases, unlike CPE. The SEP capsule is thicker and fibrous. The most common cause is chronic peritoneal dialysis. It causes SEP in recurrent peritonitis, ventriculoperitoneal and peritoneo-venous shunts, sarcoidosis, intra-abdominal tuberculosis, Mediterranean fever, systemic lupus erythematosus and fibrogenic foreign materials. Emergency operations due to SEP have a higher mortality rate than CPE. AC, on the other hand, is another disease whose cause is unknown and progresses with encapsulation. Etiologically, recurrent gynecological infections and retrograde menstruation are held responsible (8,9). In our case, none of these etiological factors were present and the diagnosis was compatible with CPE.

Since CPE is a congenital disease, malformations such as situs inversus and congenital epigastric hernia can be seen together (10). No additional congenital malformation was observed in our patient.

Dave et al. In this study, 45 cases shared in the literature were examined. The mean age was 40.8 years, and it was found to be more common in males (11). However, the true incidence is difficult to determine. These data are compatible with our case.

Preoperative diagnosis is difficult. There are no diagnostic laboratory or imaging parameters. For this reason, it is necessary to suspect first of all for the diagnosis. CT may show small clumping formed by the peritoneal membrane. Dilated small intestines form a spiral sign (1,12). Its treatment is surgical excision of the peritoneal membrane. Adhesions must be separated at all junction points between the small bowel loops. The intestinal loops in the capsule are released and placed in the abdomen. If there is a necrotic or perforated segment, it is resected. Histological examination reveals normal peritoneal tissue and fibrovascular tissue covered with mesothelium without inflammation (13). In our patient, after making the definitive diagnosis with diagnostic laparoscopy, all areas that could be separated laparoscopically were separated, and open surgery was performed to excise the deeply located bands. Thus, possible morbidities were prevented.

The prognosis for CPE is very good. No recurrence was reported after surgery. The longest follow-up period in the literature is 7 years without complications (6,11).

Conclusion

CPE is a rare clinical entity that occurs as a result of a congenital malformation of accessory peritoneal tissue surrounding the small intestine. CPE should be considered in small bowel obstructions of unexplained etiology.

Laparoscopic evaluation is effective in diagnosing CPE, but in cases where the long small bowel segment is affected, as in our case, we think that it would be appropriate to switch to open surgery to prevent morbidity, as well as the necessity of separating all bands.

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Case Report

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Acute Onset Isolated Abducens Paralysis After Exposure to Arc Welding

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Abstract

Exposure to welding is associated with many diseases, especially some of the eye injuries. We present a 49-year-old male patient applied to us with the complaints of double vision and blurred vision for a week. The symptoms started after looking welding machine light for a few minutes without protective glasses. After excluding another cause, the diagnosis of idiopathic abducens paralysis was made. As a result of exposure to metals and ultraviolet radiation, conjunctival disorders, presbyopia, refractive errors, corneal damage, and retinal pathologies can be seen in patients. It is also known that UV is a stress factor that will initiate reactions that lead to apoptosis of neurons. We think that exposure to UV rays may have played a role in the possible etiological process in our case. The patient was followed up with conservative treatment and all symptoms resolve within a month.

Keywords: Abducens paralysis, welding, double vision

Introduction

Welding is a process that is widely practiced in many industrial areas, the main purpose of which is to join two metals. It has known that exposure to welding is associated with many diseases, especially some of the eye injuries (1-3).

Abducens nerve is an isolated motor nerve which is innervates the lateral rectus muscle which abducting the ipsilateral eye (4, 5). Its nucleus is in the pons and is located lateral to the medial longitudinal fascicle and at the base of the fourth ventricle. It leaves the brain stem via the pontomedullary junction and follows a long subarachnoid course. It reaches the orbit through the superior orbital fissure via passing through cavernous sinus near carotid artery. During this long course, injury of the abducens nerve can occur anywhere and for any reason (4-6). Therefore, it requires extensive research to find the right cause in abducens paralysis.

In our article, we report a case of a patient has an acute isolated abducens paralysis after looking at a high light source from welding, which is unique in the literature.

Case Report

A 49-year-old male patient applied to us with complaints of double vision and blurred vision for a week. The symptoms started after the patient looked at an intense light source from an electric-powered welding machine for a few minutes without protective glasses. When his symptoms did not go away, he applied to our center. During this period, he did not have any complaints such as headache, vision loss, nausea, vomiting, and cough. His double vision did not improve during the day and was present constantly without showing a diurnal rhythm. The patient had no regular medication and no diagnosed chronic disease. He has a history of subacute thyroiditis and the presence of maternal diabetes in his family history.

At first admission, he was conscious, cooperative, and oriented. The vital signs were normal (Tension arterial 115/70; heart rate: 84, SpO2:94; capillary blood glucose: 98). There was no nuchal rigidity or signs of meningeal irritation. Direct and indirect light reflexes were normal in both eyes. His left eye could not abduct and could not cross the midline

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when looking to the left. He did not feel pain during eye movements. However, he had photophobia, and to prevent this, the patient was voluntarily closing his left eye. He was able to open his eyes when asked. The sclera and cornea were normal in both eyes. Other neurological and systemic examinations, including visual field examination and fundus examination, were normal. Isolated left abducens paralysis was detected in the patient.

Complete blood count (CBC), kidney function tests, liver function tests, serum electrolyte levels, sedimentation, C-reactive protein (CRP), HbA1c, thyroid function tests, anti-thyroglobulin, anti-thyroid peroxidase, and antiacetylcholine receptor antibody tests were normal. Also, his COVID-19 Polymerase chain reaction (PCR) test was negative. No significant findings were detected in thorax computerized tomography (CT), diffusion magnetic resonance imaging (MRI), Brain CT, and Optical coherence tomography (OCT) examinations. Contrast-enhanced brain MRI showed gliotic foci with a tendency to coalesce in periventricular and subcortical white matter (Figure 1), but the lesions seen did not explain the patient's complaint. Orbital MRI with contrast, brain MRI angiography, cervical MRI angiography, visual evoked potential (VEP), and electromyogram (EMG) examinations were normal. Isolated abducens paralysis developed secondary by welding light exposure was considered in the patient.

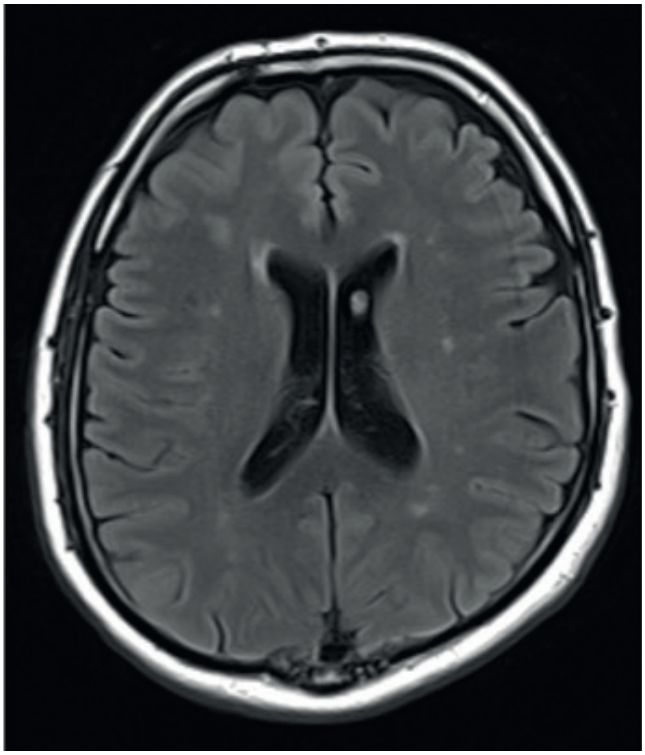

Figure 1. Gliotic foci with a tendency to coalesce in periventricular white matter in MRI

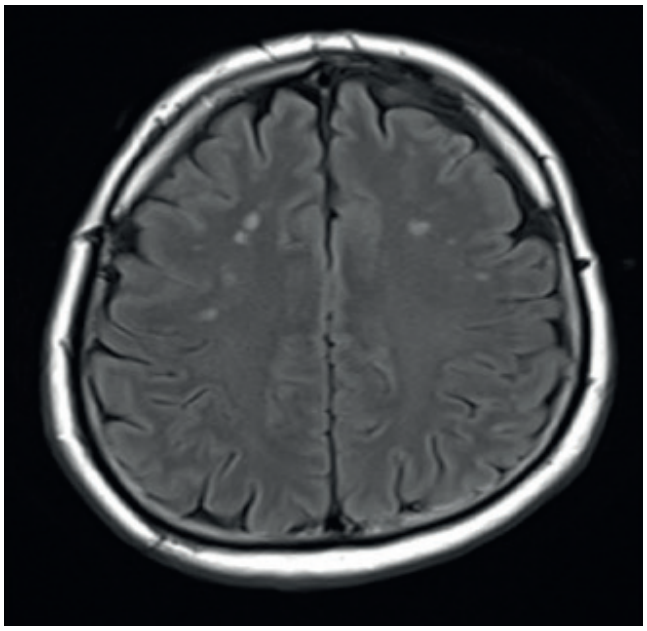

Figure 2. Gliotic foci with a tendency to coalesce in subcortical white matter in MRI

Discussion

Arc welding is recognized as a well-known potential occupational hazard. Pathologies in the eye occur due to UV light burns, flying metal objects, harmful metal gases, and thermal burns (1,2). As a result of exposure to metals and ultraviolet radiation, conjunctival disorders, presbyopia, refractive errors, corneal damage, and retinal pathologies can be seen in patients (3). In addition, it has been reported that the use of appropriate protective glasses during the welding process reduces the harmful effects of infrared, visible, and ultraviolet radiation, providing a kind of mechanical protection for the eye against welding spatter and reducing the intensity of visible light (2,3). The patients present with complaints of tearing and pain in the eyes, blepharospasm, photophobia, and vision loss (2-4). Injuries can include corneal opacity, corneal and conjunctival foreign bodies, traumatic iritis, traumatic mydriasis, or burns (2-4). Diplopia after exposure to welding light is very rare, and isolated abducens paralysis has never been reported in the literature.

Isolated abducens nerve palsy is a common cause of acute horizontal diplopia and is the most common isolated ocular motor cranial neuropathy in adults (5). A study reported its annual incidence as 11.3/100.000 (6). Its etiology includes vascular diseases, infective causes, demyelinating diseases, tumors and other structural lesions, neuromuscular junction disorders, trauma, idiopathic intracranial hypertension, and diabetic mononeuropathy. Also, idiopathic cases were reported. Etiological causes according to anatomical

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Table 1: Etiology of abducens paralysis according to anatomical locations[8]

Anatomical location	Etiology
Brainstem fascicle	Infarction, tumor, demyelination
Subarachnoid space	Meningitis, high or low intracranial pressure
Petrous apex	Petrous bone fractures, Gradenigo's syndrome, clivus tumors
Cavernous sinus	Aneurysm, neoplastic infiltration, idiopathic inflammation (Tolosa-Hunt syndrome), infection, cavernous-carotid fistula
Orbital apex	Infection (mucormycosis, aspergillosis), neoplastic infiltration, idiopathic inflammation

localizations are summarized in Table 1. Imaging and laboratory examinations were performed in our case for the differential diagnosis, and possible other reported causes were excluded.

In a study conducted on young patients, the rate of idiopathic abducens nerve palsy was shown to be 22%, and almost all of them resolved in 6-8 weeks (8). After excluding the above-mentioned causes, the diagnosis of idiopathic abducens paralysis was made. It is generally a benign condition, and patients should undergo a follow-up period of several months to exclude the possibility of recurrence (9). In our case, the patient was followed up with conservative treatment and all symptoms resolved within a month.

A study showed that myelin structure absorbs UV (10). The effects of UV are not limited to skin and eye lesions. Pathologies related to the immune system and neuroendocrine system can be explained according to the degree of absorption and transmission of UV (11). UV entering the eyes has been shown to act directly on the central nervous system, with effects on the skin to a similar degree as exposure (12). It is also known that UV is a stress factor that will initiate reactions that lead to apoptosis of neurons (13). We think that exposure to UV rays may have played a role in the possible etiological process in our case

Conclusion

Since there is no example of abducens damage developed after exposure to arc welding in the literature, this case was found worthy of being presented. It is known that UV exposure can play a role in neuron damage. In this case, we think that the possible etiological cause may be UV exposure.

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Case Report

Journal of Emergency Medicine Case Reports

Platypnea-Orthodeoxia Syndrome Associated with COVID-19 Pneumonia and Prolonged Treatment Due to Tamsulosin Use: A Case Report

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Abstract

Platypnea-orthodeoxia syndrome (POS) is an extremely rare condition. There are no other cases in the literature where tamsulosin has been reported to prolong POS treatment. A 67-year-old male patient was hospitalized due to COVID-19. He was followed up in the ward after being in the intensive care unit for ten days. There was a significant decrease in saturation (SaO2) when sitting and standing compared to lying. He was diagnosed with POS. However, there was no significant improvement in POS with exercises. After stopping the tamsulosin he was using, there was a dramatic improvement. He was discharged on the 72nd day of his hospitalization. Due to COVID-19, POS is more likely to appear than before. It is a condition that clinicians should recognize. The pathophysiology of POS has not been fully elucidated. The case we present suggests that alpha-blockers may also be related to pathophysiology.

Keywords: COVID-19; Platypnoea-orthodeoxia syndrome; SARS-CoV-2.

Introduction

Platypnea-orthodeoxia syndrome (POS) is a clinical entity characterized by positional dyspnea (platypnea) and arterial desaturation (orthodeoxia) that occurs when sitting or standing up and usually resolves when lying down. The decrease in oxygen saturation is considered significant when the PaO2 falls more than 4 mmHg, or the SaO2 falls more than 5% from the supine position to the upright position (1). POS is a rare finding, and its true prevalence is unknown (1).

It was first described in 1949 and was first called "orthostatic cyanosis" (2). The terms "platypnea" and "orthopnea" were used to describe shortness of breath and arterial desaturation that worsened in the upright position and improved in the supine position in 1969 and 1976, respectively (3,4).

We present a case of POS that developed due to COVID-19 and whose treatment we think has been prolonged due to tamsulosin use.

Case Report

A 67-year-old man was admitted to hospital with cough, fatigue and low oxygen saturations. He tested positive for SARS-CoV-2. SaO2 at 80% with FiO2 60%. He was taken to the intensive care unit due to the need for high-flow oxygen. He had previous diagnoses of chronic kidney disease (never on dialysis) and benign prostatic hypertrophy. The drugs he uses constantly are tamsulosin and allopurinol 150 mg. Tamsulosin was discontinued because a urinary catheter was inserted, and allopurinol was continued. Pulse 500 mg/day methylprednisolone was given for three days in the intensive care unit, 250 mg/day for three days, 120 mg/day for three days, and 120 mg/day when he came to the ward. Empirical antibiotic therapy (ceftriaxone and clarithromycin) and antithrombotic prophylaxis with enoxaparin (4,000 IU/day) were administered. He was not intubated while in the intensive care unit. He did not need a non-invasive mechanical ventilator and was followed up with high-flow oxygen. He was taken to the ward after ten days of intensive care hospitalization. SaO2 was 94% in the supine position with nasal O2. Sitting SaO2 was 85%, standing SaO2 was 78%. Breathing exercise was taught by physical therapy with the Triflow ball exercise. He practiced exercises harmoniously and regularly. The urinary catheter was removed, and tamsulosin was started once a day due to difficulty and pain in urination. With nasal oxygen in the supine position, with 4 lt/min, his saturation was 94%, at a 45-degree angle, his shortness of breath started, and although oxygen was provided in the

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sitting position, his saturation decreased to 65% within one minute, and he had severe shortness of breath, hypotensive complaints, and blood pressure was measured 70/40 mm/Hg. When the patient was supine, SaO2 rose above 92%, and his blood pressure rose to 100/70 mm/Hg within two to three minutes. Echocardiography was performed to look for the etiology of a cardiac shunt, atrial myxoma, atrial thrombus with stalk or platypnea, and orthostatic hypotension as he had significant platypnea. The ejection fraction was 60%, and heart cavities were normal, and there was no finding to explain platypnea. Contrast-enhanced thorax computed tomography angiography was not performed due to renal failure. On the 52nd day of the patient's hospitalization, non-contrast thorax computed tomography was performed, and fibrotic areas were observed compared with the first tomography (Figure 1). The patient was clinically evaluated as POS. The angle of the daily bed was increased to make it steeper, and short-term sitting exercises were done. There was no significant improvement after exercises such as intermittent sitting and straightening the bed angle applied for six weeks. Noradrenaline infusion

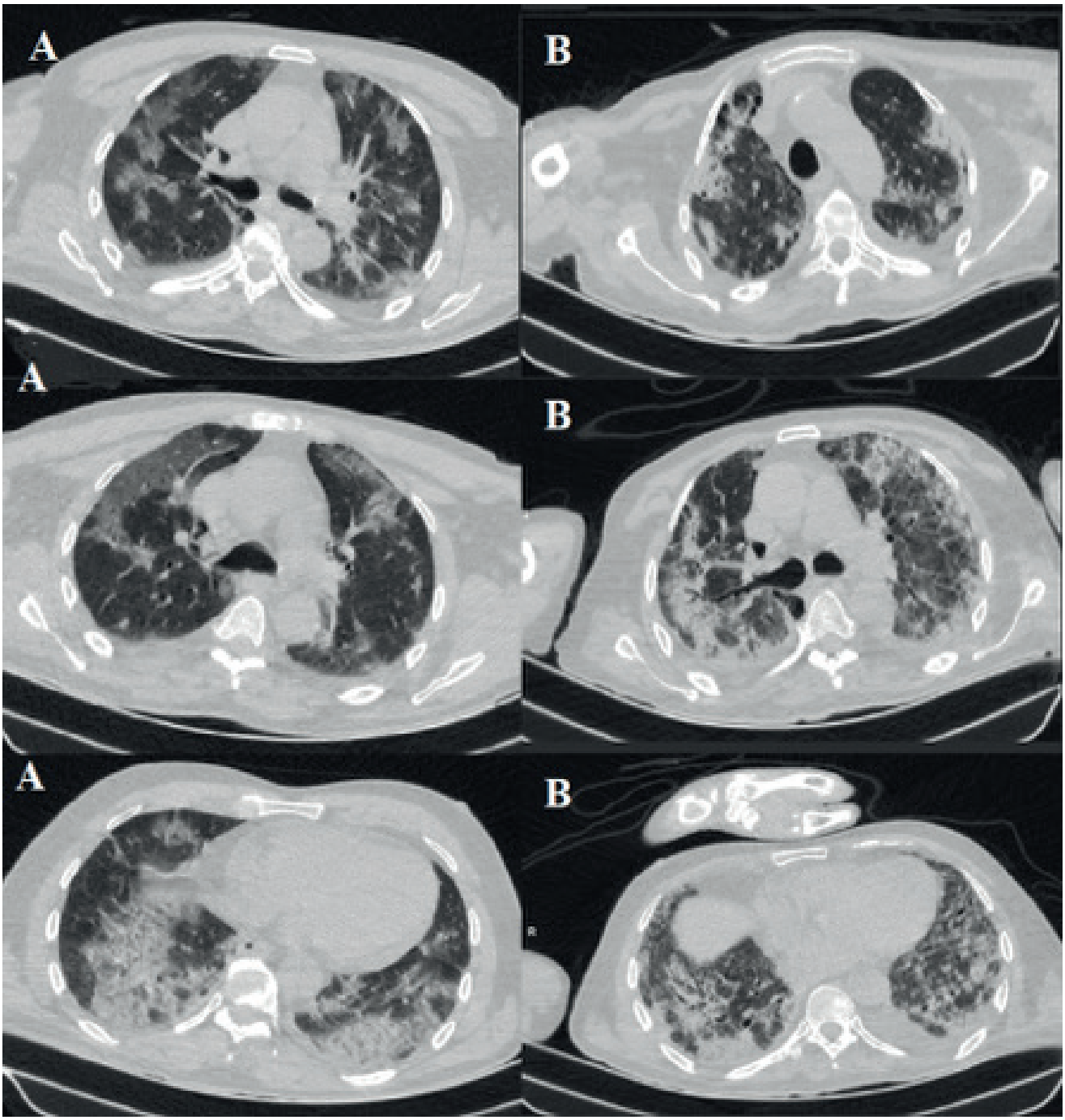

Figure 1. A: Thorax CT taken on the first day of hospitalization, diffuse ground-glass opacities, B: Thorax CT taken on the 52nd day of hospitalization, fibrotic tissues.

at a 0.11 microgram/kg/minute dose was given twice as 12 hours/day. While receiving the infusion, the patient's SaO2 did not decrease below 80% in the sitting position (it decreased to 65% when noradrenaline was not infused), and the patient did not feel shortness of breath clinically as before. When he stood up, he showed hypotensive symptoms for about 30 seconds, and then his blood pressure was around 100/70 mm/Hg. However, even though he did not take a step while standing, his SaO2 fell below 70%. Tamsulosin was discontinued and a urinary catheter was inserted. Approximately 24 hours after the last dose of tamsulosin, the patient was dramatically better. His saturation in sitting position was 93%, and he could sit for minutes. He still had hypotensive symptoms for the first 20-30 seconds when he stood up, but his saturation did not fall below 88%. Walks with 2-3 meters intervals were carried out with support. SaO2 did not fall below 89% in room air without oxygen support in the sitting position. Nasal oxygen support was needed while standing and exerting. He had difficulty walking due to the decrease in muscle strength since he had been lying down for a long time. We discharged the patient with an oxygen concentrator on the 77th day of hospitalization, six days after discontinuing tamsulosin. Information was received by phone three days after discharge, and he was walking 3-4 meters without support. On the 15th day of discharge, he could walk at home without support.

Discussion

A case of POS with orthostatic hypotension, which developed due to COVID-19 and whose treatment we think was prolonged using tamsulosin, was shared. POS is a rare condition, and its true prevalence is unknown.[1] We think that its possible prevalence is higher than known. According to our research in the literature, there are 366 publications in Pubmed when "platypnea orthodeoxia syndrome" is written, and 16 as of 18.12.2021 when "covid and platypnea orthodeoxia syndrome" is written. The feature that distinguishes our case is the prolongation of the treatment due to tamsulosin. We also did not find a case with a previously reported association between tamsulosin and POS. The POS, which did not improve for about two months, showed dramatically positive results about one day after the discontinued tamsulosin. Also, when our case was in a sitting or standing position, SaO2 decreased from 92% to 65%, much more than the cases in the literatüre (5-8).

In the retrospective analysis, we found that the increase in this decrease was more significant after using tamsulosin. It has been reported that the possible pathophysiology of POS may be dilatation and the development of physiological shunt in alveolar capillaries, as in hepatopulmonary syndrome (9). We think that tamsulosin, an alpha-blocker, may have triggered dilatation of alveolar capillaries and delayed treatment. There may also be a different pathophysiological relationship between POS and tamsulosin. It has been stated that physical therapy, albumin, norepinephrine, and indomethacin may benefit treatment. We received a clinically effective response to norepinephrine, and we did not start indomethacin because he had CRF. However, we saw more effective improvement after discontinuing tamsulosin. The pathophysiology of POS is not known exactly (10).

Conclusion

We think that our report of a case worsening with tamsulosin may inspire future research on the relationship between alpha receptors and POS to elucidate the pathophysiology of POS.

Acknowledgement: We would like to thank our patient for allowing sharing as a case and the pandemic service workers for their efforts. Each author has contributed to patient's follow-up, literature search and manuscript writing process.

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Case Report

Journal of Emergency Medicine Case Reports

Stubborn Cough; A Rare Symptom Due to Pacemaker Lead Perforation

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Abstract

Cardiac perforation by pacemaker (PM) lead is a rare but potentially life-threatening complication. The clinical presentation is highly variable. Patients may be asymptomatic or present with chest pain, dyspnea or tamponade. In this case, we report a patient with pacemaker implantation who developed a stubborn cough secondary to lead perforation.

Keywords: Cough, lead perforation, pacemaker

Introduction

Cough is a natural defense system of the respiratory tract. It is a common symptom that can be seen in many pathological conditions in addition to respiratory system diseases. It is not always easy to detect the underlying etiology (1).

With the increase in the elderly population all over the world, the incidence of atrioventricular (AV) block is rising (2). In patients with third degree AV block, PM implantation is recommended regardless of symptom status (3). Depending on the procedure and device type, complication rate after cardiac pacing is approximately 5-15%. This rate is even higher in the elderly (2). Cardiac perforation is a rare complication with a rate of 0.3-0.7% (3). Here we report a patient with pacemaker implantation who developed a stubborn cough secondary to lead perforation.

Case Report

A 91-year-old female patient was admitted to the emergency department with stubborn cough and dyspnea. She had a history of hypertension and VVI-R PM (Vitatron SR MRI SureScanTM SSIR, Maastricht, The Netherlands pacemaker and Medtronic CapSureFix Novus MRITM SureScanTM 5076 – 58 cm, Minneapolis, Minnesota, USA ventricular lead; pacing threshold = 0.9 mV, lead impedance = 510 Ω) implantation 1 month earlier due to high-grade atrioventricular block. Her complaints started approximately after 2 weeks of PM implantation. Physical examination was unremarkable with a pulse rate of 80 bpm, a blood pressure of 125/75 mmHg, an oxygen saturation level of 94%, and a respiratory rate of 26/minute. It was learned that the patient was started on antibiotics by a pulmonologist 1 week earlier, however her cough did not regress. Pneumonia, pneumothorax and pulmonary embolism were excluded in chest computed tomography (CT) imaging. Electrocardiography revealed PM rhythm with a left bundle branch block pattern and intermittent capture failure (arrow) (Figure 1a). On transthoracic echocardiography, the global ejection fraction (EF) was 60% and there was minimal pericardial fluid surrounding the heart. On modified apical 4-chamber view, PM lead protrusion through the right ventricular (RV) apex was observed (arrow) (Figure 1b). The position of the lead was evaluated with chest X-ray (Figure 2a). Although the chest X-ray did not show the lead position accurately, CT showed that the tip of the lead had perforated the RV apically and extended into the pericardium (arrow) (Figure 2b). A complete PM check showed that the pacing threshold of the RV lead had increased to 5 volts/1.0 ms. Initially, open thoracic surgery and epicardial lead placement were planned for the patient, but a conservative approach was decided due to advanced age and high surgical risk of the patient. After 1 week of follow up, the patient was discharged since there was no clinical deterioration or further changes on control echocardiography and CT.

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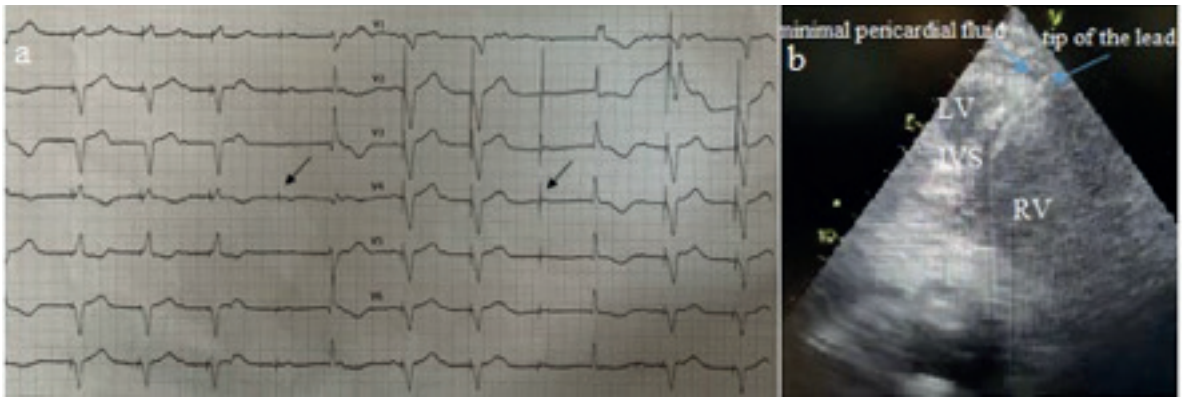

Figure 1. Electrocardiography revealed pacemaker rhythm with a left bundle branch block pattern and intermittent capture failure (arrow) (a), and modified apical 4-chamber view on transthoracic echocardiographic examination showing pacemaker lead protrusion through the right ventricular apex was observed (arrow) and minimal pericardial fluid surrounding the heart (arrow) (b).

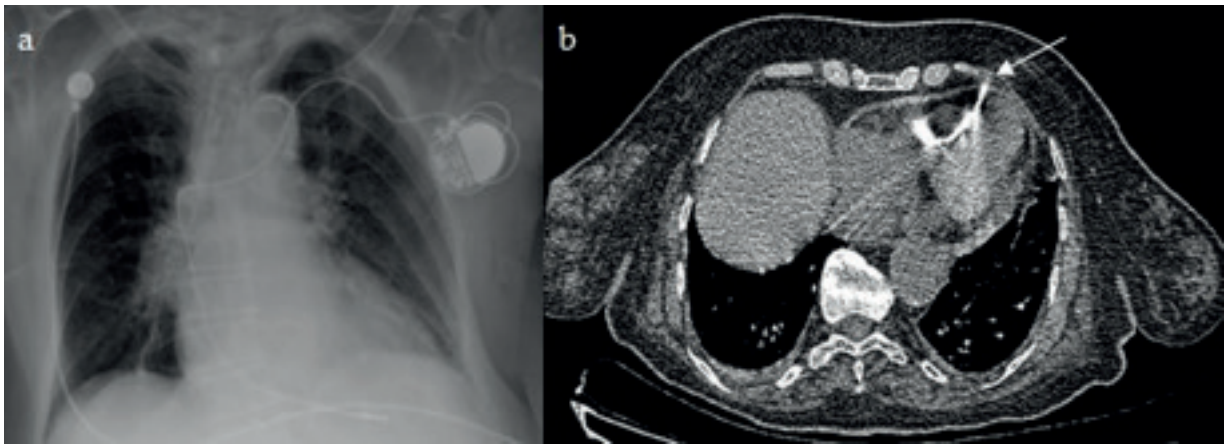

Figure 2. Chest X-ray showing the position of the pacemaker lead (a), and computed tomography showed that the tip of the lead had perforated the right ventricular apically and extended into the pericardium (arrow) (b).

Discussion

In this case, it is demonstrated that permanent PM perforation can cause stubborn cough. Cardiac perforation by PM lead is a rare but potentially life-threatening complication (4). The clinical presentation is highly variable. Patients may be asymptomatic or present with chest pain, dyspnea or tamponade (5,6).

Since cough is a common symptom in respiratory tract diseases, it may not be associated with PM-related complications. In a case report, Steiner S. and friends report a PM lead perforation related chronic cough (7). VVI pacing may be associated with a variety of symptoms called PM syndrome. Cough is an unusual but known symptom of this syndrome (8). Diaphragmatic stimulation is rare but can occur when a misplaced lead stimulates the diaphragm directly or via the phrenic nerve. This can be the first sign of cardiac perforation. The most common complaints of these patients are hiccups, contractions and contraction induced chest pain and shortness of breath (9). Cough usually occurs as a result of stimulation of sensory nerves in the larynx and tracheobronchial tree by various inflammatory, mechanical, thermal or chemical stimuli. These sensory nerves are also located in organs such as pericardium, esophagus, diaphragm, and stomach (10). We thought that the cough and dyspnea that developed in our patient was due to mechanical irritation of the pericardium and diaphragm.

If we evaluate other conditions that may cause cough; since the left ventricular EF was normal on echocardiography and there was no sign of pulmonary congestion or peripheral edema, the complaints were not considered to be due to heart failure. Diseases affecting the respiratory system such as pneumothorax, pneumonia and pulmonary embolism, which are other causes of cough, were ruled out with thorax CT. The use of angiotensin converting enzyme inhibitors, which is also a frequent cause of cough, was not seen in our patient. Considering the mechanisms discussed above and since the patient did not have symptoms prior to PM implantation, we attributed her symptoms to cardiac perforation.

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Conclusion

Although it is rare, PM-related cardiac perforation should be considered in the differential diagnosis of stubborn cough as it can be fatal.

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Case Report

Journal of Emergency Medicine Case Reports

Isolated Hyoid Fracture Associated with Blunt Trauma

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Abstract

Trauma-related hyoid fractures are rare entities. These fractures represent only 0.002% of head and neck region fractures. Victims of hanging and strangling constitute the great majority of cases. Fractures associated with trauma are extremely rare. These fractures are difficult to diagnose and can easily be overlooked during physical examination. However, they are also important traumas since airway safety is endangered in these rare cases. We describe a case of a young male presenting with isolated neck injury associated with hitting an electric cable while riding a motorbike. Tenderness was present in the anterior neck region at physical examination, but movement was not restricted and no respiratory difficulty was determined. Isolated hyoid fracture was detected at tomography of the neck performed in the emergency department. Hyoid bone fractures should not be forgotten in patients with pain and tenderness in the anterior neck region following blunt trauma to the neck.

Keywords: Isolated blunt trauma, cervical computed tomography, hyoid bone fracture

Introduction

Hyoid bone fractures may appear during strangulations, hangings, and intubation. The majority may accompany multiple fractures and polytraumas such traffic accidents and falls from heights (1). However, isolated hyoid fractures are very rare. Airway safety is threatened in traumas specific to the neck region. Loss of time in or uncertainty of diagnosis increase mortality. Physical examination must therefore be carefully evaluated in patients with histories of blunt head and neck trauma (3). Very few cases of hyoid bone fracture caused by blunt neck fracture have been reported to date (1)

We present a case of a young man with an isolated fracture of the hyoid bone associated with blunt neck trauma caused by a traffic accident.

Case Report

A 26-year-old motorcyclist presented to our emergency department with pain in the anterior neck region associated with his neck hitting an electric cable on the road during a nocturnal journey. The patient reported falling from the motorbike and feeling pain in the anterior part of the neck, but no other injury. He also stated that he was wearing protective equipment such as a crash helmet. At presentation to our emergency department his blood pressure was 130/85 mm Hg, heart rate 78/min, respiration rate 11/min, body temperature 36.5° C, and oxygen saturation 97%. Neck movements were not restricted at physical examination, but the left horn region of the cricoid bone in the anterior midline of the neck was tender when palpated. His Glasgow Coma Scale score was 15/15. His pupils were isochoric, and no pathology was encountered at neurological examination. No bone tenderness was present at repeated cervico-spinal examinations. No redness or localized mass/swelling were observed in the anterior neck region. No traces of trauma related to the accident were observed other than minimal abrasions on the inner surface of the arm, the hands, and knees.

No pathological findings were encountered on posterior-anterior and lateral cervical radiographs taken during presentation. There was no fracture line on the lateral cervical radiograph, and soft tissues were normal in appearance. Cervical computed tomography (CT) scanning was performed due to localized tenderness persisting in the

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anterior part of the neck. The CT report described a fracture of the greater left horn of the hyoid bone and bilateral anterior half irregularity and edema at the level of the vocal cords (Figure 1,2). An ear, nose, and throat physician was consulted. Bedside indirect transoral laryngoscopy was performed, and no edema or perforation were observed in the airway. Symmetrical movement was present in the bilateral vocal cords, and minimal edema was observed. Lung x-ray performed at the same time was unremarkable. The patient's COVID-19 PCR test analyzed during presentation to the emergency department was delta variant-positive. Due to the neck trauma and COVID-19 positivity, the patient was admitted to the COVID-19 isolation ward. No COVID-19-related symptoms were present. Following 24-h emergency department follow-up, the patient was discharged with restricted neck movements due to hyoid bone fracture and advice concerning a fluid or soft diet for a few days and subsequent check-up. He was placed under home quarantine due to COVID-19.

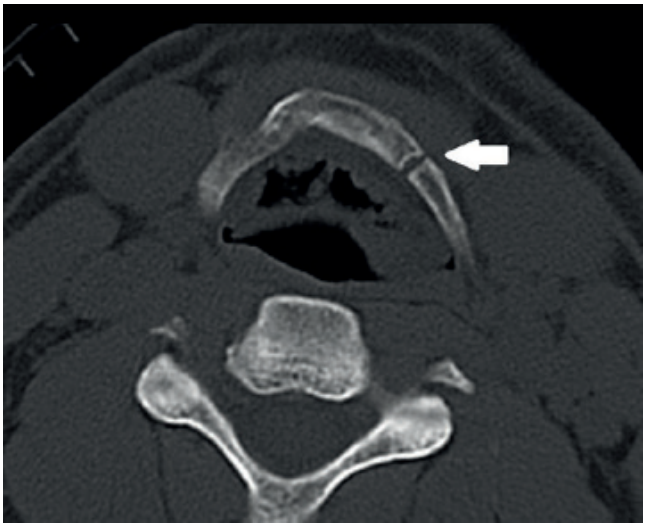

Figure 1. Cervical CT axial section; fracture at the juncture of the hyoid bone left corpus major horn

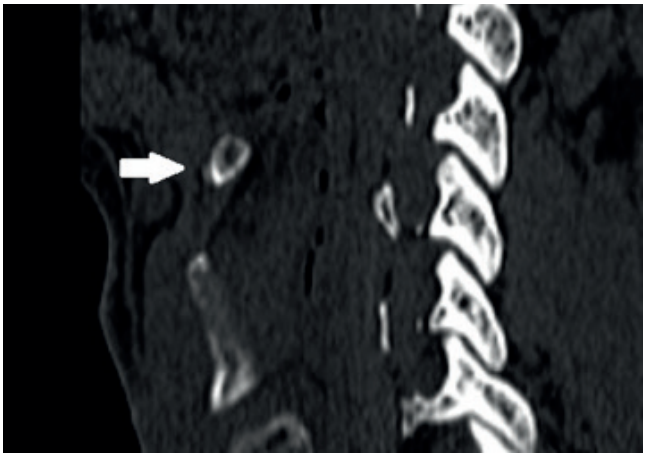

Figure 2. Cervical CT sagittal section; fracture at the juncture of the hyoid bone left corpus major horn

Discussion

Examination of the literature shows that approximately 40 cases of hyoid fracture have been reported in the last century, including in the pediatric age group. The great majority of these cases were hyoid fractures accompanying maxillofacial area polytraumas. Isolated hyoid fractures, however, are extremely unusual. Blunt neck trauma is generally caused by hanging, sport, or attack (3). The hyoid bone consists of a main body, and two greater and two lesser horns. It lies at the level of the third and fourth vertebrae in the anterior part of the neck. The bone is a suspended styloid protrusion immediately below the chin, just above the thyroid cartilage, anterior to the cervical spine, supporting the movement of the tongue. These structures protect the hyoid bone against direct blows and injuries. Since the bone lacks a fixed structure, it is not easily affected by traumas, and fractures are therefore rare. In the present case, localized contact in the neck region, even though he was wearing protective equipment against motor vehicle accidents, exposed the patient to severe force due to the speed of the vehicle.

Pain may be the only symptom in hyoid fractures. However, these fractures should be suspected when pain is accompanied by odynophagia, dysphagia, and dyspnea. Short-term observation and conservative treatment are sufficient in the majority of cases. However, albeit theoretically and rarely, the need for endotracheal intubation, tracheostomy, or laryngeal-pharyngeal perforation has also been reported (3).

Diagnosis relies on suspicion in patients with clinical signs. Pain with movement or palpation, swallowing difficulty, and pain with coughing may be observed. Respiratory difficulty is a sign of severe trauma-related injury and edema. Radiological imaging is used to confirm diagnosis. Fractures are difficult to visualize on cervical radiographs due to the surrounding osseous structures and the superimposition thereof. Evaluation using cervical CT is therefore recommended when a fracture is suspected. CT permits the evaluation of soft tissues, the cervical spine, and vascular structures. Direct laryngoscopy is also recommended to determine potential airway damage in patients with such injuries. Conservative treatment is generally sufficient, and surgical intervention is rarely required.

Fracture in the left greater horn of the hyoid bone and bilateral irregularity and edema at the level of the vocal cords were detected in this case. Direct laryngoscopy revealed symmetrical motility in the vocal cords, and no injury or edema posing a threat to the airway were observed. The patient was discharged with conservative treatment following emergency department follow-up and observation.

Conclusion

Hyoid fractures are rare entities, and insufficient cases have been reported in the literature. Care is required in terms of hyoid fracture in case of traumas threatening the face and neck. Outcomes threating the airway may be encountered due to hyoid fractures, particularly in case of simple-looking blunt neck traumas. Symptoms that may develop following blunt trauma should be carefully evaluated, and the possibility of hypoid fracture should not be forgotten.

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Case Report

Journal of Emergency Medicine Case Reports

Mortal Complication of Kikuchi–Fujimoto Disease; Lower Gastrointestinal Bleeding, Case Presentation and Literature Review

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Abstract

In this report, we inform you that a patient with CFD, known as a benign disease, died due to massive lower GI bleeding. Although KFD is a benign disease, we have seen in this case that it can result in death. A 28-year-old Turkish male patient was brought to the hospital with complaints of high fever, fatigue, sweating and difficulty breathing. A cervical examination revealed that the patient had lymphadenopathy. The patient did not have any cutaneous lesions. Genital examination revealed melena. A normal cranial and abdomen, thorax CT was obtained. The patient was intubated because of respiratory failure and unstable hemodynamics. The patient developed pancytopenia as a result of blood tests. The patient died due to massive lower gastrointestinal bleeding.

Keywords: Kikuchi–fujimoto disease, lower gastrointestinal bleeding, histiocytic necrotizing lymphadenitis

Introduction

Kikuchi–Fujimoto Disease (KFD) or histiocytic necrotizing lymphadenitis is known to be a benign, generally self-limiting condition, which usually affects female patients under the age of 30 years. Most of the cases improve within six months. It is a benign disease mainly characterized by high fever, lymph node swelling, and leukopenia (1,2). It presents most often with cervical lymphadenopathy, which may be painful and can be accompanied by fever and upper respiratory tract symptoms. Unilateral involvement of the posterior cervical group is the most common manifestation (3). Less common manifestations include fever, axillary and mesenteric lymphadenopathy, splenomegaly, parotid gland enlargement, cutaneous rash, arthralgias, myalgias, aseptic meningitis, bone marrow haemophagocytosis, and interstitial lung disease. The cutaneous lesions include erythematous macules, papules, plaques, and nodules (4). Since the first description of the disease by the Japanese pathologists Kikuchi and Fujimoto (1,2) its etiology has remained unknown, although environmental factors, in particular viruses, have been suspected (5). Treatment is symptomatic with nonsteroidal anti-inflammatory drugs, and most symptoms improve within a few months (6). Laboratory findings are usually normal except for inflammatory syndrome or mild cytopenia, sometimes associated with hemophagocytosis (7). With this case, we present the coexistence of KFD and lower GIS bleeding for the first time in the literature. Our case aims to inform that there may be complications with a mortal course in KFD and to inform our physicians about these complications.

Case Report

A 28-year-old male patient was brought to the hospital with complaints of high fever, fatigue, sweating and difficulty breathing. In August 2022, it was found that the patient has been diagnosed with KFD as a result of a bone marrow biopsy. Biopsy is the gold standard diagnostic method for this disease. It was clarified that the patient had no additional disease. His vital signs during admission were as follows: blood pressure 121/71 mmHg, pulse 132/minute, SpO2 81%, and a fever of 38,3. His ECG showed a sinus tachycardia rhythm. His cervical examination revealed lymphadenopathy. The patient did not have cutaneous lesions. His breath sounds were normal bilaterally. Genital examination revealed melena. Laboratory results at our hospital were as follows: white blood cell count, 5,84/µL (33,1% neutrophils); absolute neutrophil count, 1,94/µL; hemoglobin level, 8.1 g/dL; platelet count, 32,000/µL; aspartate aminotransferase, 1210 IU/L; alanine aminotransferase, 1187 IU/L; and γ-glutamyl transferase, 197 IU/L. The patient's radiological imaging (brain, thorax and abdominal CT) was reported as normal. Epstein-Barr virus (EBV), Parvovirus B19, Anti-nuclear antibody (ANA), Brucella, Toxoplasma,

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anticardiolipin IgM tests, as well as negative cytoplasmic and perinuclear anti-neutrophil cytoplasmic antibodies (C-ANCA and PANCA), anti-double-stranded DNA, and HLA-B57 values that have been tested at an external medical center were found to be negative, these tests are used to confirm the diagnosis. The patient was started on immediate replacement therapy (blood replacements, 2 Units of ES, 2 units of TDP). During the treatment, his hemodynamics deteriorated and intubation was performed in the emergency room for airway protection. After his hemodynamics improved, the patient was admitted to the internal medicine ICU from the emergency room. The blood panel that was taken in the ICU and showed hgb value of 5.4 mg/dl. He developed cardiac arrest 10 hours after hospitalization and the patient died despite all interventions.

Discussion

Although KFD usually presents with fever, cervical lymphadenopathy, or involvement of other lymph nodes, it may also present with symptoms related to other systems. These include maculopapular rash due to skin involvement, anemia due to bone marrow involvement, night sweats due to systemic exposure, weight loss, nausea, vomiting, sore throat due to respiratory system involvement and abdominal pain hepatomegaly and splenomegaly due to gastrointestinal system involvement (3).

Kucukardali et al. reported the characteristics of the patients in a study that analyzed 244 cases published by them. Accordingly, in these cases, the most common symptoms were weakness and joint pain, while the most common physical examination findings were lymphadenomegaly and erythematous rashes. Our case was also compatible with this information. In this case report, anemia and lymphopenia were frequently detected as laboratory tests, and this was consistent with our case. While additional diseases were detected in 51% of the patients in this article, there were none in our case. Küçükkardalı et al. reported that 64% of the patients recovered on their own without any treatment. In our case, he was not currently taking any medication (3).

Dumas et al. reported asthenia, weight loss and fever as the most common symptoms in their series of 91 cases. As for laboratory tests, high inflammatory parameters and neutropenia came to the fore. Gastrointestinal bleeding was not reported in either of the two multi-case analyses (6).

The association of KFD disease with bleeding has been reported only rarely in the literature. Bilateral preretinal hemorrhage has also been reported in a recent scientific article (8).

As the rate of diagnosis of KFD due to medical progress increases, the disease is better recognized and more complications are reported. Akhavanrezayat et al. have linked the mechanism of bilateral preretinal hemorrhage to autoimmunity. According to this, the cause of KFD disease is not clearly known and many causes are being investigated, but the most plausible is autoimmune causes. Increased vascular permeability due to direct vascular damage by autoimmune reactions or in response to the release of immune-mediated cytokines can lead to vascular injury and bleeding. Increased fragility of superficial capillaries due to vasculitis is also considered as one of the possible mechanisms (8). In our article, we think that gastrointestinal bleeding is due to increased vascular permeability and auto-immune system involvement. Since gastrointestinal system bleeding would not be expected in a normal young patient, we think that this patient had bleeding due to the effect of Kikuchi syndrome.

Our article reports the association between KFD and gastrointestinal bleeding, which was reported for the first time in the medical literature. Reported complications help us to manage diseases properly. It is much more important to report complications, especially in rare diseases.

Conclusion

In conclusion, the association between KFD and gastrointestinal bleeding, which was reported for the first time in the medical literature, is very important. Physicians need to know the complications that develop especially in rare diseases. It should be kept in mind that CFD should be considered especially in patients with fever, cervical lymphadenopathy and fatigue triad, and bleeding may occur as a complication of KFD.

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Case Report

Journal of Emergency Medicine Case Reports

Atypically Located Hydatid Cyst Cases: Four Unusual Case Reports

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Abstract

Hydatid cysts are frequently located in liver (65-70%) and lungs (25%). The disease is usually asymptomatic when located in the extrahepatic regions. Sometimes, diagnosis can be established by observing germinative membrane, rockwater, or daughter vesicles during the operation.. A 41-year-old female patient applied with the complaint of intermittent left inguinal pain. Echinococcus Indirect Hemagglutination (IHA) test was positive. In the ultrasonographic examination, a hypoechoic area was observed in the posteromedial of the left femoral head.. A 56-year-old female patient, applied with the complaints of intermittent nausea, vomiting, and epigastric pain. She had a history of surgery for liver hydatid cyst 15 years ago and for lung hydatid cyst 10 years ago. IHA test of the patient was positive. The lesion involving septation and accompanied by calcifications around the cyst was reported as a hydatid cyst at dynamic CT for pancreas and upper abdominal MRI. A 22-year-old female patient applied with perianal pain complaint. Drainage was planned for the patient who had a pre-diagnosis of perianal abscess. Tissues similar to the hydatid cyst membrane were excised together with purulent fluid. The patient was diagnosed with hydatid cyst after the pathology result was reported "as compatible with hydatid cyst". A 32-year-old female patient had a non-metabolic adrenal subcapsular cyst (hydatid cyst?) in the right adrenal gland on ultrasonography performed for abdominal pain. In abdominal tomography, a "cystic mass compatible with hydatid cyst located in the adrenal gland in the upper pole of the right kidney" was reported. IHA test of the patient was negative. Hydatid cysts located in the perianal region and muscles have been reported as atypical localizations in the literature, and some of them were diagnosed as a result of preoperative clinical evaluation and radiological imaging. Some were diagnosed as a result of pathological examination after the operation. Surgical treatment is an option in suitable cases, and the definitive diagnosis can be established by pathology. Consequently, it should be considered that cystic lesions detected in the patients living in endemic regions can be extrahepatic hydatid cyst that is localized in different anatomical regions.

Keywords: Atypical, hydatid cyst, echinococcus

Introduction

Hydatid cyst is a zoonotic disease and is an important endemic health problem in South America, the Middle and Far East and the Mediterranean region (1,2). The disease is frequently located in liver (65-70%) and lungs (25%). When the hydatic disease is located in other extrahepatic regions, it is generally asymptomatic and symptoms related to the cyst compression or rupture can be seen (3,4). Advanced imaging techniques together with physical examination may be required for the detection of the organs involved. Sometimes, diagnosis can be established in the patients, who were taken to operation with non-specific findings, by observing germinative membrane, rockwater, or daughter vesicles during the operation. The aim of the present study is to present the hydatid cyst cases detected in the anterior of the left abdominal acetabulum, head of the pancreas, in the perianal region, and on the right adrenal gland in the patients who applied to the Department of General Surgery of Gaziantep University Sahinbey Hospital, together with the treatment approaches and clinical results in view of the literature.

Case Report

Case-1

A 41-year-old female patient, who was living in a rural area and had close contact with animals, applied to our clinic with the complaint of intermittent left inguinal pain lasting for 5 months. The patient did not have any comorbidities and history of drug use. In the physical examination of the patient, who did not have any history of operation other than the femoral surgery 10 years ago, no finding was found except for tenderness in the left inguinal region. In the laboratory examinations, biochemistry and hemogram tests were taken and the results were found normal. Only echinococcus Indirect Hemagglutination (IHA) test was positive. In the

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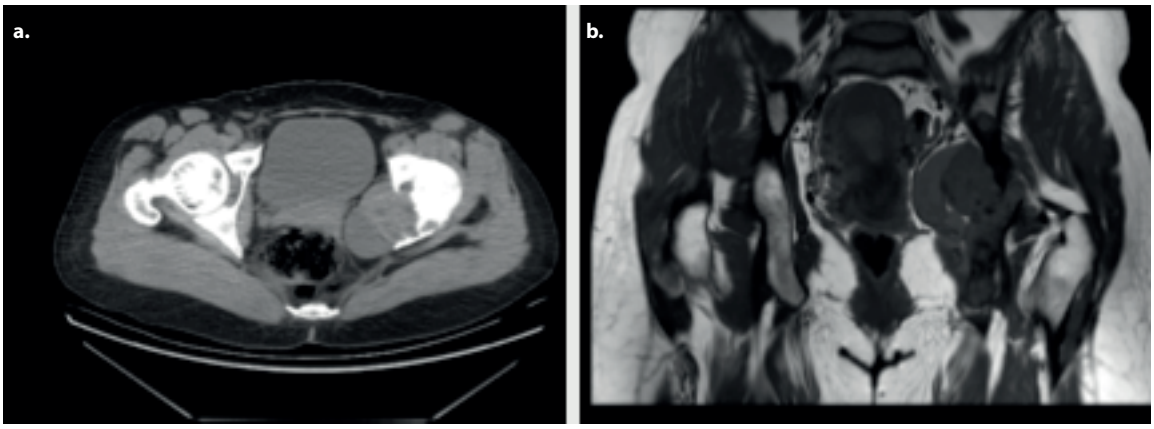

Figure 1a. Cystic lesion with a size of 52x53 mm in the left iliac bone-acetabulum localization. Described expansive cystic lesion continues till the ischium level in the left side.
Figure 1b. No multi-ocular, cystic appearance in the left iliac bone-acetabulum or no significant contrasting in the bone component of the lesion was observed. In the medial side of this lesion, peripherally contrasted multiple areas with the largest one having a size of 34x22x54 mm.

ultrasonographic examination, a hypoechoic area with an approximate size of 6x4 cm was observed in the posteromedial neighbourhood of the left femoral head. In pelvic computed tomography (CT) and magnetic resonance imaging (MRI), a lesion that is compatible with a cyst of approximately 5 cm was observed in acetabulum (Fig. 1a. and Fig 1b).

During the preoperative imaging, no lesion was detected in the patient except for the existing cyst. The patient was taken to operation and cystectomy was performed, albendazole (10 mg/kg/day) treatment started after the operation. Pathology result of the patient was assessed as "compatible with hydatid cyst". The patient had no additional complaints or physical examination findings in the control follow-up. Albendazole treatment was terminated at the end of 3 months and control was recommended every 6 months.

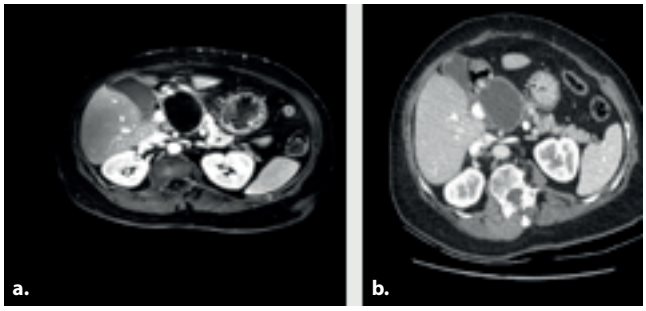

Figure 2a. Upper abdominal MR; a 64x47 mm hyperintense cystic lesion with lobulated contours, starting from the proximal part of the pancreatic body and extending to the superioraperiportal area.
Figure 2b. Abdominal CT; a 68x61 mm cystic lesion at the level of the pancreatic head and body section, which is contrasted peripheral oedematous after IVCM, accompanied by septation and peripheral calcifications (hydatid cyst?, IPMN?)

Case-2

A 56-year-old female patient, applied with the complaints of intermittent nausea, vomiting, and epigastric pain for 1 year. The patient was diagnosed with pancreatic cyst at another centre and was referred to our centre for further examination and treatment due to the possible cystic neoplasms of the pancreas. The patient, who did not have any comorbidity, had a history of surgery for liver hydatid cyst 15 years ago and for lung hydatid cyst 10 years ago. In the physical examination of the patient, no finding except for incision scar caused by the previous operation and epigastric tenderness was found. In the laboratory examinations, biochemistry and hemogram tests were assessed as normal. IHA test of the patient was positive. The patient was assessed together with dynamic CT for pancreas and upper abdominal MRI (Fig. 2). The lesion involving septation and accompanied by calcifications around the cyst was reported as a hydatid cyst.

Albendazole treatment was started for the patient and a surgical treatment was planned. The patient was taken to operation and cystotomy was performed for the cystic lesion at the head part of the pancreas. Germinative membrane and daughter vesicles were observed in the cyst. Pathology result was assessed as "compatible with hydatid cyst".

Case-3

A 22-year-old female patient applied to our outpatient clinic with perianal pain complaint. The patient had severe perianal tenderness that complied with the localization of the lesion. The patient had no comorbidity. Drainage was planned for the patient who had a pre-diagnosis of perianal abscess. In Jack-knife position, swelling and redness were observed at the radius of 9 o'clock position in perianal region. Under local anaesthesia, drainage was performed with a 0.5 cm incision. Tissues similar to the hydatid cyst membrane were excised together with purulent fluid. The content was sent to pathologic examination with the suspicion of hydatid cyst. The patient was diagnosed with hydatid cyst after the pathology result was reported "as compatible with hydatid cyst".

Other organs were normal in the imagings conducted for screening purposes.

No abnormal values were found in the hemogram and biochemistry tests of the patient. Albendazole (10 mg/kg/

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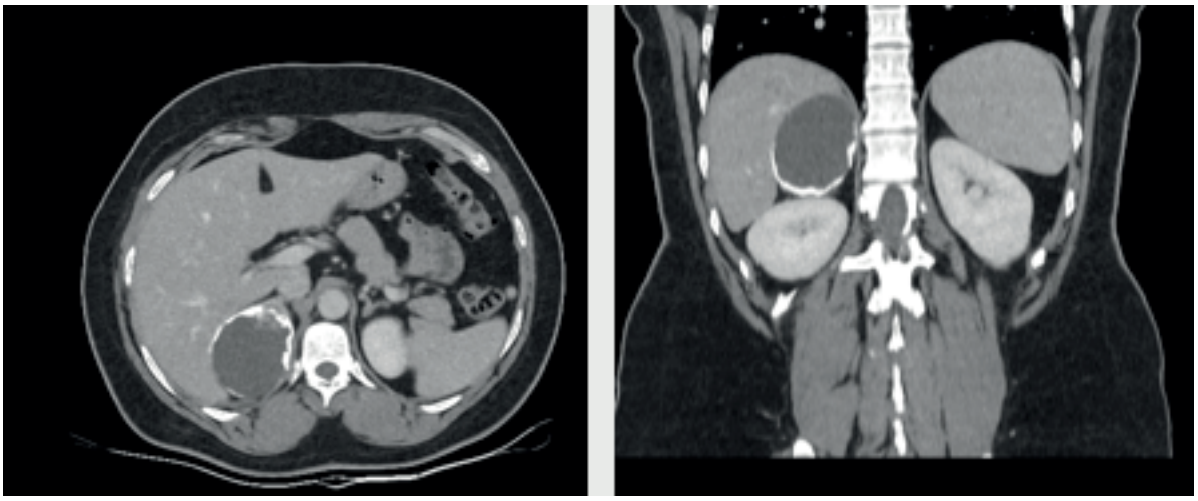

Figure 3a,b. In the abdominal tomography, a 62x65 mm nodular-linear calcified cystic lesion with dense membrane applying compression on the adrenal gland and liver in the upper pole of the right kidney without any significant contrast after IVCM E, had attracted attention. The imaging finding was reported as 'It was primarily thought to be adrenal hydatid cyst '

day) treatment was started. After 3 months, the treatment of the patient was terminated as she had no complaints. There was no need for a second surgery.

Case-4

A 32-year-old female patient was examined by an endocrinologist with the pre-diagnosis of a cystic mass in the right adrenal gland in the examinations performed due to the right upper quadrant pain. Additional examination was performed in terms of differential diagnosis of adrenal masses. The patient was consulted to us with the diagnosis of a non-metabolic adrenal mass. Ultrasonography of the patient was reported as an 80x65 mm subcapsular cystic lesion (hydatid cyst?) in the right lobe posterior of the liver. At the abdominal tomography, a "cystic mass compatible with hydatid cyst located in the adrenal gland in the upper pole of the right kidney" was reported (Fig. 3a and Fig 3b).

IHA test of the patient was negative and her other routine blood examinations were normal. Surgical intervention was not considered for the patient, who was thought to be diagnosed with adrenal hydatid cyst with a calcified membrane, and follow-up decision was taken.

Discussion

Echinococcus eggs infect people with direct contact with water, food, and dogs. From the intestines, the larvae reach the liver through the veins. Sometimes they cause the disease by passing through the liver or reaching the lungs via lymphatics. Then, they can reach other places in the body, less frequently through the blood.

It is known that Turkey is in an endemic region in terms of hydatid cyst (1).

Hydatid cysts located in the perianal region and muscles have been reported as atypical localizations in the literature, and some of them were diagnosed as a result of preoperative clinical evaluation and radiological imaging. Some were diagnosed as a result of pathological examination after the operation (5).

Hydatid cyst lesions that are located close to the pancreas are reported in the literature, and abdominal distension, jaundice, and pain can be seen in the patients. IHA test in these patients may be negative (6). Surgical treatment is an option in suitable cases, and the definitive diagnosis can be established by pathology.

Consequently, it should be considered that cystic lesions detected in the patients living in endemic regions can be extrahepatic hydatid cyst that is localized in different anatomical regions.

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Case Report

Journal of Emergency Medicine Case Reports

Toxic Epidermal Necrolysis as a Result of Hair Dye Allergy

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Abstract

More cases of hair dye allergy and poisoning are being presented to the Emergency Department (ED). There were rare TEN cases due to the hair dye in the literature. An 18-year-old female presented to our ED with the complaint of a common rash in her whole body, edema on her face that closed her eyes, and blisters on her face and neck. Her relatives reported that she dyed her hair 4 days ago 5 times in 24 hours. her initial vital signs were as follows: Blood pressure: 90/55 mmHg, Heart Rate:128 beats /min, Respiratory Rate: 18 beats/min, Oxygen saturation:100%, Body Temparature:40.3 2C. According to her history and physical examination, she was diagnosed with toxic epidermal necrolysis. According to starting the appropriate treatment modalities immediately in ED, she was discharged with full recovery in spite of her high predicted mortality. Although emergency medicine physicians encounter TEN patients infrequently, they must be aware of and initiate the appropriate treatment modalities immediately

Keywords: Hair dye allergy, toxic epidermal necrolysis, emergency department

Introduction

In our developing world, the use of hair dye is increasing throughout all age groups, from children to the elderly. Due to this trend, more cases of hair dye allergy and poisoning are being presented to the Emergency Department (ED). The most popular and preferred hair dyes are permanent hair colors which include P-Phenylenediamine (PPD), hydrogen peroxide (usually a 6% solution form), and Propylene glycol(1). Due to P-henylenediamine's high allergic and toxigenic effects, 1 to 6 % of all dermatitis patients all around the world were reported due to the hair dyes containing PPD (2).

Toxic Epidermal Necrolysis (TEN) is defined as an immune-mediated mucocutaneous disease characterized by the detachment of the epidermis and mucous membrane. It is also named 'immunologic burn' because of the same appearance of skin as a burn. The main causes are drugs (allopurinol, carbamazepine, phenytoin, sulfasalazine, sulfonamides, and non-steroidal anti-inflammatory drug) and infections (3). The severity-of-illness score for TEN (SCORTEN) is used to predict mortality (Table 1) (4). Clinical features including fever, sore throat, and malaise accompany the dermatologic symptoms of blisters and vesiculobullous rash with epidermal sloughing and necrosis. Herein we present a case in that hair dye allergy mutated to TEN.

Case Report

An 18-year-old female presented to our ED with the complaint of a common rash in her whole body, edema on her face that closed her eyes, and blisters on her face and neck. Her relatives reported that she dyed her hair 4 days ago 5 times in 24 hours. This was the third ED presentation over the course of three days. On her first presentation, 3 days ago; she had urticarial plaques on her body and extremities; she was treated with intravenous antihistaminics (pheniramine hydrogen maleate 45.5 mg) and discharged with the oral antihistaminic drugs (cetirizine and clemastine). After 24 hours, she presented to the ED due to a worsening of her symptoms. Because of the recurrent visit in 24 hours, she was consulted with the Dermatology and they rearranged her oral antihistaminic drugs and scheduled a control exam for two days later. On the 3rd admission, her initial vital signs were as follows: Blood pressure: 90/55 mmHg,

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Table 1: Risk factors, scores and mortality prediction of SCORTEN

Risk factors for SCORTEN "Score of Toxic Epidermal Necrosis"	
Age over 40 years	
Heart rate >120 beats per minute	
Presence of cancer or hematologic malignancy	
Epidermal detachment area involving body surface area >10%	
Blood urea nitrogen >28 mg/dL (10 mmol/L)	
Blood glucose >252 mg/dL (14 mmol/L)	
Bicarbonate <20 mEq/L	
Mortality rate in SCORTEN	
Number of risk factors	**Mortality rate (%)**
0–1	3.2
2	12.1
3	35.3
4	58.3
5	90

Heart Rate:128 beats /min, Respiratory Rate: 18 beats/min, Oxygen saturation:100%, Body Temparature:40.3 2C. Her Glasgow Coma Scale score was 15 and she was oriented and cooperated. Her respiratory system examination was normal. Her dermatologic examination revealed a severe, painful erythematous rash all across the surface of her whole body, including her extremities, as well as partially bullous lesions on her face and neck, as well as bilaterally enlarged eyelids that made it difficult for her to open her eyes (Figure 1).

According to her history and physical examination, she was diagnosed with toxic epidermal necrolysis. Her blood tests revealed a BUN of 14.8 mg/dl, blood glucose of 95 mg/dl, and HCO3 of 21.7 mEq/L. The SCORTEN score was calculated as 2 yet the mortality was predicted over 50% because of the involvement of the whole body. Isotonic saline infusion, 45.5 mg pheniramine hydrogen maleat, 250 mg methylprednisolone, and analgesics (paracetamol and fentanyl) were administered immediately and she was consulted with the dermatology, intensive care, and ophthalmology departments. Dermatology and ophthalmology departments recommended adding oral 100 mg of cyclosporin, topical mupirocin, polyvinyl alchol+povidon eye drop, moxifloxacin HCL as an ophthalmic solution, carbomer ophthalmic gel to the medical treatment and hospitalization in the burn intensive care. She was hospitalized in Burn intensive care for 17 days and in the dermatology ward for 22 days and was treated with a combination of methylprednisolone, cyclosporin, and immune globulin. On the fortieth day of the hospital stay, she was discharged with a full recovery of her lesions.

Discussion

TEN is a dermatological emergency characterized by diffuse epidermal necrolysis and the involvement of two or more mucosal surfaces. Additionally, the respiratory, gastrointestinal, genitourinary and renal systems might be affected (5). The most common trigger agents for TEN are drugs especially antibiotics, antiepileptics, allopurinol, and immune checkpoint inhibitors. In the absence of a definitive pathophysiological mechanism, it is assumed that T-cells activation by medications or infection plays a role in the formation of TEN. Our patient did not take any drugs however but she did color her hair five times within 24 hours. There were several case reports about hair dye poisoning in the literature ranging from basic urticaria to anaphylactic shock. According to our knowledge, no TEN cases have been linked to hair dyes. It is important to immediately start the fluid resuscitation, antipyretic, corticosteroid, and cyclosporin with the dermatology consultation. The TEN patients must be monitored in intensive care units and if it is possible in burn intensive care. The TEN patients receive the same supportive care as burn patients. Particular attention should be paid to the respiratory and cardiovascular organ systems.

Conclusion

Although emergency medicine physicians encounter TEN patients infrequently, they must be aware of and initiate the appropriate treatment modalities immediately. In addition, the management of these patients consists of multidisciplinary teams.

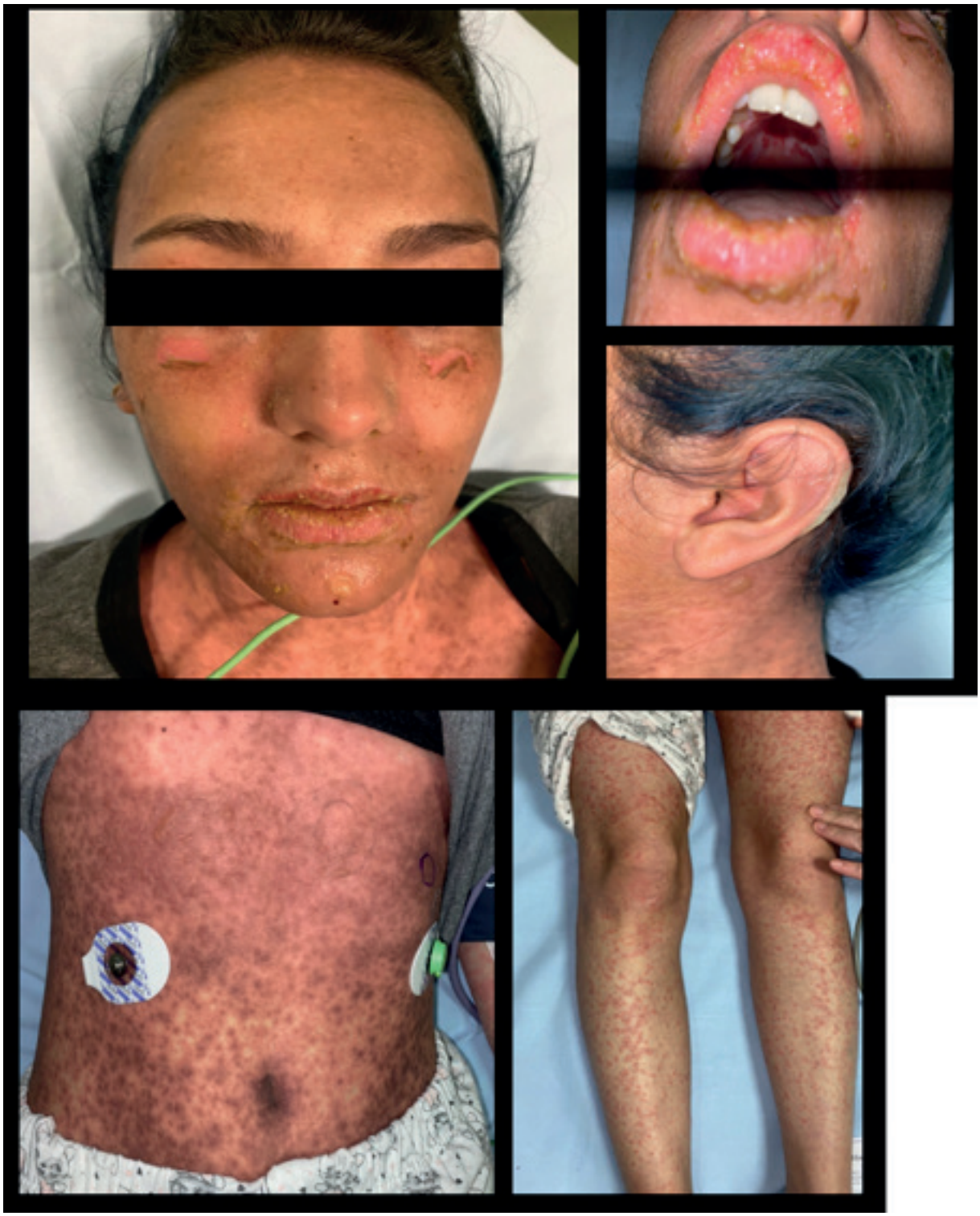

Figure 1. Computed tomography angiography (CTA) of the left hand

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Case Report

Journal of Emergency Medicine Case Reports

An Aortic Abscess Associated with Psoas Abscess: A Case Report

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Abstract

In this article, we report a case in which a psoas abscess opened into the aneurysmatic abdominal aorta, and abscess formation was observed in the aortic lumen containing a stent. A 57-year-old male patient presented to the emergency department with a complaint of abdominal pain. During the abdominal examination of the patient, tenderness and defense were detected in the right lower quadrant. Contrast-enhanced computed tomography (CT) angiography of the abdominal aorta was ordered because the patient had a history of previous stenting and right lower quadrant deficiency. In the right iliopsoas muscle, there was an increase in size suggestive of abscess formation with air densities. The diameter of the ascending aorta was 44 mm. At the level of the descending thoracic aortic bifurcation, an aneurysmatic appearance was noted in a segment of approximately 140 mm extending to the proximities of both main iliac arteries, and USG showed air in the wall in the aneurysmatic section and mural wall thickening with thrombus. In this case, although the abscess eroded the aneurysmatic aortic wall, acute bleeding did not develop due to the presence of a stent. We wanted to share the rare image on computed tomography (massive air-fluid level around the stent in the aortic lumen), especially in our case with the medical literature.

Keywords: Psoas abscess, aortic abcess, aneurysmatic

Introduction

A psoas abscess is an accumulation of inflammation in the iliopsoas muscle [1]. It can occur primarily or secondary. It may present with nonspecific symptoms such as abdominal pain, fever, loss of appetite, weight loss, and limping [1-7]. The most common pathogens detected in the abscess fluid are Staphylococcus aureus (88.4%), Streptococcus (4.9%), and Escherichia coli (2.8%) [6]. The best diagnostic method is computed tomography [1, 7-10]. After the diagnosis of an abscess is confirmed, the best treatment method is drainage of the abscess and then initiation of antibiotic therapy. A psoas abscess may rarely cause aortic rupture. Its prognosis is quite poor [11].

In this article, we report a case in which a psoas abscess opened into the aneurysmatic abdominal aorta, and abscess formation was observed in the aortic lumen containing a stent.

Case Report

A 57-year-old male patient presented to the emergency department with a complaint of abdominal pain. During the abdominal examination of the patient, tenderness and defense were detected in the right lower quadrant. No significant pathology was observed in other system examinations.

It was learned that the patient had a history of prostate malignant neoplasm, coronary artery disease, diabetes mellitus, hypertension, three percutaneous coronary interventions (PCI), one coronary artery bypass grafting (CABG), and one year ago, stenting of the abdominal aorta and both main iliac arteries with Endovascular Aneurysm Repair (EVAR). His medications were Sitagliptin+Metformin 50/1000 mg, Nebivolol hydrochloride 5 mg, Pentoxifylline 400 mg, Bicalutamide 50 mg, Perindopril arginine+Amlodipine 10/5 mg, Acetylsalicylic acid 150 mg, Alfuzosin 10 mg.

The patient's vital signs were as follows: arterial blood pressure 100/65 mm Hg, heart rate 84 beats/min, respiratory rate 16 breaths/min, oxygen saturation 96%, body temperature 37.7 °C. In the blood tests of the patient, hemoglobin: 8.55 g/dl (13-17), (control Hb after 2 hours: 8.95 g/dl), lymphocyte: 0.32 10^3/mm3 (1-5), platelets: 109x103/mm3 (150-500), CRP:8.86 (0-0.5), except for complete blood count, blood glucose level, liver function tests, renal function tests, were within the normal reference ranges, including highly sensitive cardiac troponin-I.

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Contrast-enhanced computed tomography (CT) angiography of the abdominal aorta was ordered because the patient had a history of previous stenting and right lower quadrant deficiency. CT angiography imaging of the abdominal aorta revealed stent images in both main iliac arteries after bifurcation in the abdominal aorta. There was also a 110x96 mm suspicious penetrating area with multiple air images around the external iliac artery on the right. In the right iliopsoas muscle, there was an increase in size suggestive of abscess formation with air densities. The diameter of the ascending aorta was 44 mm. At the level of the descending thoracic aortic bifurcation, an aneurysmatic appearance was noted in a segment of approximately 140 mm extending to the proximities of both main iliac arteries, and USG showed air in the wall in the aneurysmatic section and mural wall thickening with thrombus. The images were compared with the images taken one year ago (Figure 1&2&3).

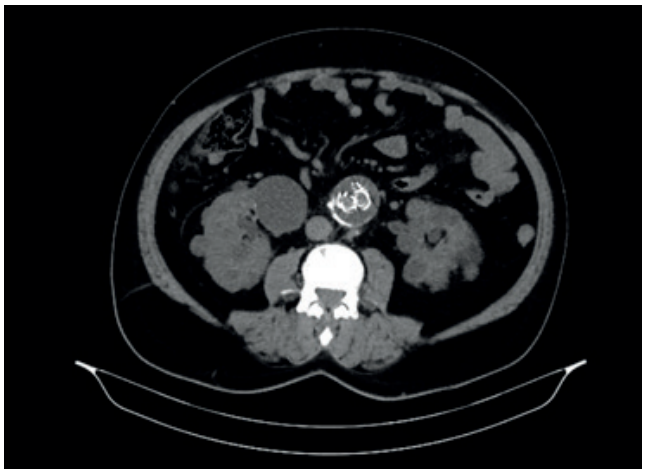

Figure 1. A non-contrast abdominal CT scan one year ago showed an aneurysmatic aorta, mural thrombus in the lumen, and a stent image of the EVAR procedure. No prominent infective findings were detected.

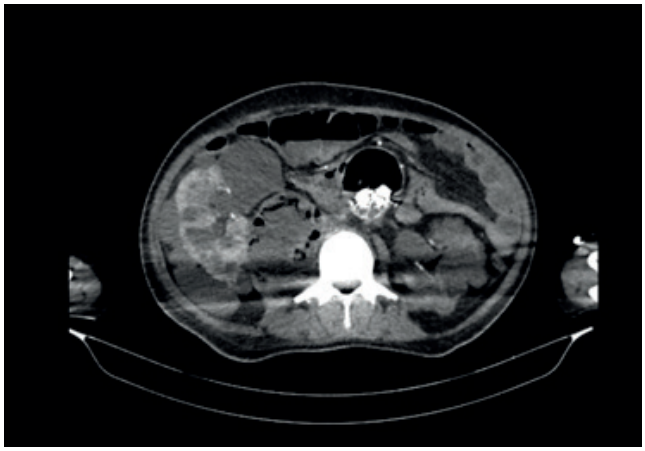

Figure 2. Contrast-enhanced abdominal CT scan performed at the patient's last admission revealed abscess formation in the right psoas and massive air and fluid level in the true lumen of the aorta.

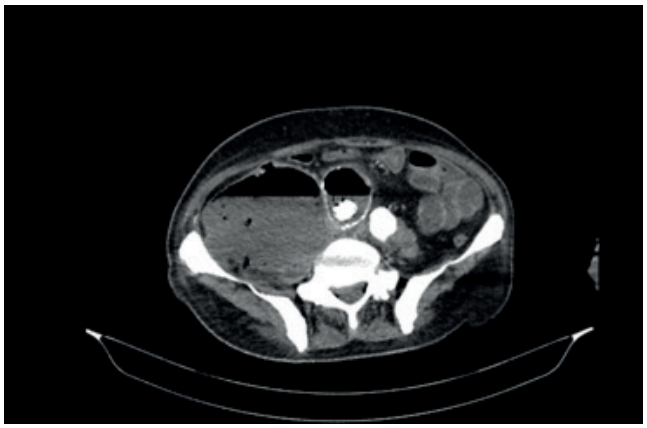

Figure 3. Contrast-enhanced abdominal CT scan performed at the patient's last admission showed massive air and fluid level in the lumen of the aortic true lumen and right iliac artery due to abscess in the right psoas.

Blood and urine cultures were obtained, and Ceftriaxone 2x1 gr and Moxifloxacin 1x400 mg were started empirically. Since the patient had an intra-abdominal abscess, general surgery and infectious diseases were consulted for drainage and antibiotherapy. Cardiovascular surgery was consulted because air image and thrombus were observed in the aneurysmatic section at the level of the abdominal aortic bifurcation. General surgery and cardiovascular surgery did not consider emergency surgery.

The patient was consulted to department of infectious diseases for antibiotherapy. Urine and blood cultures were sent with the recommendation of infectious diseases, and Piperacillin-tazobactam 4x2.25 gr was started empirically. Urine and blood cultures grew Escherichia coli. Due to deterioration in the patient's general condition, he was asked to be re-evaluated by infectious diseases for a change in antibiotherapy. Piperacillin-tazobactam treatment was stopped, and meropenem 2x500 mg, levofloxacin 750 mg loading dose, and maintenance 500 mg every 48 hours were recommended. After three days, the patient underwent percutaneous abscess drainage by interventional radiology. A 14F drainage catheter was applied to the abscess pouch, fistulizing into the aorta from the right lower quadrant lateral section. The next day, the patient's oxygen saturation decreased to 70. There was no response to Noninvasive Mechanical Ventilator. The patient was intubated. He was transferred to the intensive care unit. After one day of intensive care unit follow-up, he died.

Discussion

A psoas (or iliopsoas) abscess is an accumulation of pus in the iliopsoas muscle compartment [1]. It can occur due to contiguous spread from adjacent structures or hematogenous

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spread from a distant site. The incidence is low, but the use of computed tomography has increased the frequency of this diagnosis, with most cases diagnosed postmortem[2].

Iliopsoas abscess (IPA) can be classified as primary or secondary. Primary IPA results from an organism's hematogenous or lymphatic spread from a distant site. Risk factors include diabetes, intravenous drug use, human immunodeficiency virus (HIV) infection, renal failure, and other forms of immunosuppression [1,2]. Secondary IPA occurs when a nearby infectious/inflammatory process extends directly into the iliopsoas [3,4]. Secondary psoas abscess occurs due to the direct spread of infection from a neighboring structure into the psoas muscle. It may be unclear whether the involvement of a neighboring structure is a cause or a consequence of the psoas muscle abscess [5].

In this case, diabetes mellitus, history of malignant prostatic neoplasm, and previous EVAR (2 years ago) were considered personal risk factors for IPA. Symptoms and signs of psoas abscess include abdominal pain, back or flank pain, fever, groin mass, anorexia, limping, and weight loss [1,6,7]. Common pathogens of primary psoas abscess are Staphylococcus aureus (88.4%), Streptococcus (4.9%), and Escherichia coli (2.8%) [6]. Computed tomography (CT) is the most appropriate radiographic modality to evaluate a psoas abscess, but sensitivity may be limited early in the disease [1,7-10]. In most cases, the abscess is apparent; other findings may include a focal hypodense lesion, infiltration of surrounding fat, and a level of gas or air-fluid within the muscle [9,11]. Primary psoas abscess has a better prognosis with a mortality rate of 2.4%. Mortality in untreated cases is 100% [1]. The association of the psoas abscess and the abdominal aortic aneurysm is infrequent. When aortic rupture occurs, the prognosis of aortic infection secondary to psoas abscess is very poor. Immediate abscess drainage following correct diagnosis and arterial reconstruction before aortic rupture is mandatory [11].

In our case, Escherichia coli was grown in a blood culture. It was thought to have progressed to the aorta by direct dissemination. Contrast passage was observed in the stent of the EVAR procedure in the aorta, but the diffuse gas image and air-fluid level were observed in the actual lumen of the aorta surrounding the stent. Our case was mortal despite drainage and antibiotherapy.

Conclusion

The association of psoas abscess and aortic rupture is infrequent. It can be rapidly fatal due to acute hemorrhage after rupture. In this case, although the abscess eroded the aneurysmatic aortic wall, acute bleeding did not develop due to the presence of a stent. We wanted to share the rare image on computed tomography (massive air-fluid level around the stent in the aortic lumen), especially in our case with the medical literature. We should remember that this association, which is very rare even in the world of medical literature, has a high mortality rate in emergency departments, we should remember what we should do when we encounter it.

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