

- Small Bowel Obstruction Caused by Rare Intraluminal Hematoma Secondary to Gastric Bypass: A Case Report Shamil Aliyev, Hakan Seyit, İlke Dolğun
- 2. Arnold Chiari Malformation Type 1 with Cerebellar Tonsil Herniation through Foramen Magnum: A Case Report Rahul Shil, Adelarisa Mitri
- Disproportionate Short Stature in Nail Patella Syndrome: Clinical, Radiological, and Genetic Insights

Yüksel Yaşartekin, Onur Dirican, Abbas Ali Husseini, Ayşe Derya Buluş, Uğur Ufuk Işın, Mehmet Ali Ergün

4. A Case of Pneumopericardium after Pericardiocentesis Conservatively Managed with Multimodality Imaging Methods

Ahmet Seyfeddin Gürbüz, Serhat Kesriklioğlu, Ahmet Taha Şahin

 A Rare Case Requiring Emergency Tracheotomy, Isolated Tracheal Mucormycosis: It is Important to Suspect First

Bilge Tuna, Gökhan Tüzemen, Emine Kıyım Altıntaş

6. An Interesting Cause of Abdominal Pain in the Emergency Room

Naim Hikmet Kalkan, Muhammet Gökhan Turtay, Mustafa Çifçi, Berke Yıldırım 7. Testicular Infarction: A Rare Complication of Epididymo-Orchitis

Kaan Karamık, Ceren Aydın

- 8. Current Approaches in the Treatment and Reconstruction of Frostbite Injuries: A Case Report ihtişam Zafer Cengiz, Fatma Çakmak
- Abdominal Aortic Aneurysm Presenting with Acute Pancreatitis: A Rare Clinical Manifestation Mevlana Gül, Yunus Emre Ek
- 10. Lead Ingestion in a Child: A Case Report Mustafa Ümit Can Dölek, Ahmet Karanfil, Alp Gökçe Altındaş
- 11. Importance of Recombinant Tissue Plasminogen Activator (rt-PA) Thrombolysis in Hyperacute Stroke Patients with Aphasia-A Case Report

Ahmad Luqman Bin Md Pauzi, Juliana Hashim

12. Superior Trunk Block: A Novel Approach for Shoulder Reduction in Pregnancy

Chun Chau Tan

13. Unexpected Ant Bites

Levent Şahin, Barış Kaban

 Severe Pancytopenia Induced by Methotrexate in a Rheumatoid Arthritis Patient

Müge Özgüler, Alper Koç, Hakan Ayyıldız



Owner and Responsible Manager

Başar Cander Bezmialem Vakif University, Department of Emergency Medicine İstanbul, Turkey

Editors in Chief

Doç. Dr. Muhammed EKMEKYAPAR

Editorial Board

Doç. Dr. Levent ŞAHİN Dr. Öğr. Üyesi Ayça ÇALBAY Dr. Öğr. Üyesi Barış KABAN Uzm. Dr. Yasin UĞUR

Printing and Graphics Department



Seyrantepe Mah. İbrahim Karaoğlanoğlu Cd. İspar İş Merkezi, D: No: 105 D:124, 34418 Kâğıthane/İstanbul Telefon: 0553 199 95 59 info@puntodizgi.com www.puntoajans.com

1.	Small Bowel Obstruction Caused by Rare Intraluminal Hematoma Secondary to Gastric Bypass: A Case Report
2.	Arnold Chiari Malformation Type 1 with Cerebellar Tonsil Herniation through Foramen Magnum: A Case Report
3.	Disproportionate Short Stature in Nail Patella Syndrome: Clinical, Radiological, and Genetic Insights
4.	A Case of Pneumopericardium after Pericardiocentesis Conservatively Managed with Multimodality Imaging Methods12 Ahmet Seyfeddin Gürbüz, Serhat Kesriklioğlu, Ahmet Taha Şahin
5.	A Rare Case Requiring Emergency Tracheotomy, Isolated Tracheal Mucormycosis: It is Important to Suspect First
6.	An Interesting Cause of Abdominal Pain in the Emergency Room
7.	Testicular Infarction: A Rare Complication of Epididymo-Orchitis
8.	Current Approaches in the Treatment and Reconstruction of Frostbite Injuries: A Case Report
9.	Abdominal Aortic Aneurysm Presenting with Acute Pancreatitis: A Rare Clinical Manifestation
10.	Lead Ingestion in a Child: A Case Report31 Mustafa Ümit Can Dölek, Ahmet Karanfil, Alp Gökçe Altındaş
11.	Importance of Recombinant Tissue Plasminogen Activator (rt-PA) Thrombolysis in Hyperacute Stroke Patients with Aphasia-A Case Report34 Ahmad Luqman Bin Md Pauzi, Juliana Hashim
12.	Superior Trunk Block: A Novel Approach for Shoulder Reduction in Pregnancy38 Chun Chau Tan
13.	Unexpected Ant Bites
14.	Severe Pancytopenia Induced by Methotrexate in a Rheumatoid Arthritis Patient44 Müge Özgüler, Alper Κος, Hakan Ayyıldız

Journal of Emergency Medicine Case Reports

Small Bowel Obstruction Caused by Rare Intraluminal Hematoma Secondary to Gastric Bypass: A Case Report

⑤ Shamil Aliyev¹, ⑥ Hakan Seyit², ⑥ İlke Dolğun³

¹Department of Interventional Radiology, Istinye Univesity Medicalpark Gaziosmanpasa Hospital, İstanbul, Türkiye

²Department of General Surgery, Istinye Univesity Medicalpark Gaziosmanpasa Hospital, İstanbul, Türkiye

³ Clinic of Anesthesiology and Reanimation, Haseki Training and Research Hospital, İstanbul, Türkiye

Abstract

Mechanical small bowel obstruction after gastric bypass is a common complication and the most common causes of obstruction are kinking or stenosis of the entero-enterostomy, internal herniation, adhesions, external compression on the transverse mesocolon, incarceration of an abdominal wall hernia, or intussusception. One of these complications is intraluminal hematoma, which, although rare, has dangerous consequences that can even lead to mortality. Since it is a life-threatening complication, early diagnosis and emergency intervention are important. A 39-year-old female patient with a history of previous laparoscopic bariatric surgery and current BMI=39.8 was admitted due to weight regain. After the Preoperative preparation and anesthesia consultation, laparoscopic roux-NY distalization operation was performed. During the follow-up, it was observed that the patient's general condition worsened on the postoperative 12th hour. In the IV contrast-enhanced whole abdominal CT scan, spontaneous hyperdense dense content obstructing the lumen was observed, and the radiologist stated that this hyperdense content could be primarily in favor of intraluminal bleeding and hematoma. In this case report, we wanted to emphasize the early effectiveness of radiology and surgery in the diagnosis and treatment processes of a patient with intraluminal hematoma after laparoscopic roux-NY gastric bypass. The most common location of intraluminal hematoma is the jejunojejunal anastomosis area. Symptoms may ocur earlier (at the 12th postoperative hour). Whole-abdominal MDCT remains the main method of diagnosing intestinal obstruction, including small bowel obstruction resulting from intraluminal hematoma.

Keywords: Complication, computed tomography, gastric bypass, intraluminal hematoma

Introduction

Obesity is an important global health problem that especially affects the Western world. At least 300,000 deaths annually in the United States are thought to be related to obesity (1). In obesity surgery, which is increasing in paralel with the increase in obesity, laparoscopic Roux-en-Y gastric bypass (LRYGB) surgery, a restrictive and malabsorptive technique, is among the Standard procedures to achieve consistent and sustainable weightloss. Small bowel obstruction (SBO) after LRYGB is a significant complication that contributes to increased morbidity and mortality, and its incidence ranges from 0.4% to 7.45% (2). While there are many reasons in the etiology of SBO, acute mechanical intestinal obstruction caused by intraluminal hematoma (IH) after LRYGB is much rarer (upto 0.71%) (3). Multidetector computed tomography (MDCT) is the primary technique in the evaluation of symptomatic patients after LRYGB, especially in terms of detecting anastomotic leaks and SBO in the early period after surgery (4,5). MDCT after oral and IV administration of contrast material is invaluable in confirming the presence, location, and associated complications of SBO (1). In this case report, the clinical and radiological management of early SBO occurring after distalization due to weight regain after LRYGB is presented.

Case Report

A 39-year-old female patient with a history of laparoscopic bariatric surgery, the details of which she did not know, at an external center approximately 20 years ago, applies to our hospital for revision surgery. The preoperative demographic findings of the patient, who lost weight to 65 kg in the first postoperative year but gained weight again during the follow-upperiods, were measured as height: 167 cm, weight: 111 kg, and BMI: 39.8.

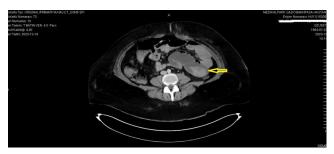
After the Standard procedure of internal medicine department, psychologist, dietician consultations and abdominal ultrasonography, it was understood that she had Roux-NY gastric bypass during gastroscopy performed by us. Distalization was planned for the patient, who was planned for revision surgery due to weight regain.

Preoperative preparation and anesthesia consultation was performed, and then laparoscopic roux-NY distalization operation was performed. The operation was completed without any problems, the patient was woken up and sent to the ward.

In the postoperative follow-up, her vital signs were found to be stable and her drain was 50 cc serohemorrhagic. During the follow-up, it was observed that the patient's general condition worsened on the postoperative 12th hour and that she was agitated, tachypneic, normotensive and normocardic. Since distension was also detected during the abdominal examination, a complete blood count and whole abdominal CT with intravenous (IV) contrast were planned.

In the whole abdominal CT scan with IV contrast, significant dilatation was observed in the remnant stomach, duodenum and jejunal loops proximal to the jejunojejunostomy anastomosis, and it was reported that spontaneous hyperdense dense content obstructing the lumen was observed in the lumen of the jejunal loops in the left upper quadrant (Figure-1). It was stated by the radiologist that the spontaneous hyperdense content mentioned may be primarily in favor of intraluminal bleeding and hematoma, and the dilatation in the remnant stomach and duodenum may have developed secondary to the obstruction caused by the hematoma.

In the exploration after laparotomy performed under emergency conditions with these preliminary diagnoses; The



 $\textbf{Figure 1.} \ \textbf{Itraluminal hematoma and proximal dilatation}$



Figure 2. Evacuation of intraluminal hematoma by milking method

jejunojejunostomy anastomosis was intact, the alimentary leg and common channel jejunum loops were normal, but intraluminal hematoma and dilatation were observed in the biliary leg. The stum padjacent to the anastomosis was opened and intraluminal hematoma drainage was performed using the milking method from this area (Figure-2) and the anastomosis line was checked. The hemorrhagic area on the anastomosis line was primarily sutured. The patient's perioperative general condition improved and the operation was completed with no additional pathology detected.

The patient was followed up as extubated in the postoperative intensive care unit, and two erythrocyte replacements were performed during this two-day hospitalization. The patient's general condition improved and she was discharged on the 7th day after the follow-up and treatment was completed.

Discussion

Mechanical small bowel obstruction after LRYGB is a common complication occurring in 0.4% to 7.45%, and the most common causes of obstruction are kinking or stenosis of the entero-enterostomy, internal herniation, adhesions, external compression on the transverse mesocolon, incarceration of an abdominal wall hernia, or intussusception (6). Intraluminal hematoma causing proximal small bowel obstruction at the anastomotic site after LRYGB remains an unusual event.

In our literature review, it was stated that this rare complication occurs with in the second to fifth day following the gastric bypass procedure (6,7). However, in our patient, it was noticed in the first 12 hours and treated quickly. With this case report, we would like to emphasize once again that close follow-up of patients in the early postoperative period is important in such surgeries.

Although close follow-up is performed, the symptoms of these patients may not present as classic small bowel obstruction symptoms. There may be findings that may suggest obstruction, such as nausea, vomiting, tachycardia, and abdominal pain, or there may be no specific symptoms (6). However, it has been mentioned in the literature that Tachycardia and the feeling of "dooms day is approaching" are among the common symptoms of intraluminal hematoma causing obstruction in the jejunojejunostomy area (8). Although our patient's findings were not specific, the presence of agitation was an indication that something was wrong. An attempt was made to make a diagnosis by performing a CT scan with IV contrast under emergency conditions.

It has been stated in the literature that upper gastrointestinal examination is very valuable in the evaluation of symptomatic patients after Roux-en-Y gastric bypass, especially in terms of detecting anastomotic leaks and SBO within 4 months after surgery (1). MDCT after oral

and IV administration of contrast materialal lows optimal evaluation of postoperative anatomy and complications, including SBO. In our patient, CT was very useful in making the diagnosis, as the clinical symptoms did not lead to a specific diagnosis. The reason why oral contrast was not given to our patient was purely to save time.

After laparotomy, a hemorrhagic area in the jejunojejunostomy anastomosis line and an intraluminal hematoma in the biliary leg were detected in the patient's CT scan, as mentioned. When we look at the literature, it is stated that intraluminal hematoma can most commonly occur in or near the an astomosis area, as in our patient (3,6,7,9).

Obstructions caused by intraluminal hematoma can lead to serious consequences. It must be intervened urgently as it may cause not only intestinal perforation and peritonitis, but also mortality. There are treatment recommendations for both laparoscopic and laparotomy in the literature (3,7,10). However, if clinical suspicion is high, the idea that exploration should be continued to prevent subsequent perforation and peritonitis prevails, even if imaging findings are suspicious (8-11). In parallel with the literature, we performed a laparotomy in our patient, opened the stump adjacent to the anastomosis, and performed intraluminal hematoma drainage from this area using the milking method.

Conclusion

In conclusion, although acute mechanical intestinal obstruction (SBO) caused by intraluminal hematoma after LRYGB is rare, it should be treated immediately due to its life-threatening character. The most common location of IH is the jejunojejunal anastomosis area. Symptoms may ocur earlier. Whole-abdominal MDCT remains the main method of diagnosing intestinal obstruction, including SBO resulting from intraluminal hematoma.

Conflict of Interest

No conflicts of interest between the authors and/or family members of the scientific and medical committee members or members of the potential conflicts of interest, counseling, expertise, working conditions, share holding and similar situations in any firm. Informed consent was obtained from the patient for this case report.

- Sunnapwar A, Sandrasegaran K, Menias CO, Lockhart M, Chintapalli KN, Prasad SR: Taxonomy and imaging spectrum of small bowel obstruction after Roux-en-Y gastric bypass surgery. AJR Am J Roentgenol 2010, 194(1):120-128.
- **2.** Felsher J, Brodsky J, Brody F: Small bowel obstruction after laparoscopic Roux-en-Y gastric bypass. Surgery 2003, 134(3):501-505.
- **3.** Green J, Ikuine T, Hacker S, Urrego H, Tuggle K: Acute small bowel obstruction due to a large intraluminal blood clot after laparoscopic Roux-en-Y gastric bypass. J Surg Case Rep 2016, 2016(8).
- Husain S, Ahmed AR, Johnson J, Boss T, O'Malley W: Smallbowel obstruction after laparoscopic Roux-en-Y gastric bypass: etiology, diagnosis, and management. Arch Surg 2007, 142(10):988-993.
- **5.** Uppot RN: Impact of obesity on radiology. Radiol Clin North Am 2007, 45(2):231-246.
- **6.** Peeters G, Gys T, Lafullarde T: Small bowel obstruction after laparoscopic Roux-en-Y gastric bypass caused by an intraluminal blood clot. Obes Surg 2009, 19(4):521-523.
- Mustafa RR, Khaitan L, Saleh AA, Elghalban H, Kedia A, Abbas M: Blood clot causing small bowel obstruction after Roux en-Y Gastric bypass: a Case Report & Review of Literature. Annals of Laparoscopic and Endoscopic Surgery 2019, 4.
- **8.** Awais O, Raftopoulos I, Luketich JD, Courcoulas A: Acute, complete proximal small bowel obstruction after laparoscopic gastric bypass due to intraluminal blood clot formation. Surg Obes Relat Dis 2005, 1(4):418-422; discussion 422-413.
- Shimizu H, Maia M, Kroh M, Schauer PR, Brethauer SA: Surgical management of early small bowel obstruction after laparoscopic Roux-en-Y gastric bypass. Surg Obes Relat Dis 2013, 9(5):718-724.
- 10. Nelson LG, Gonzalez R, Haines K, Gallagher SF, Murr MM: Spectrum and treatment of small bowel obstruction after Roux-en-Y gastric bypass. Surg Obes Relat Dis 2006, 2(3):377-383, discussion 383.
- **11.** Lewis CE, Jensen C, Tejirian T, Dutson E, Mehran A: Early jejunojejunostomy obstruction after laparoscopic gastric bypass: case series and treatment algorithm. Surg Obes Relat Dis 2009, 5(2):203-207.

Journal of Emergency Medicine Case Reports

Arnold Chiari Malformation Type 1 with Cerebellar Tonsil Herniation through Foramen Magnum: A Case Report

Rahul Shil¹, Adelarisa Mitri²

Department of MSN (Neuroscience), Sapthagiri Institute of Medical Sciences and Research Centre, Sapthagiri NPS University, Bengaluru, India

Abstract

Arnold Chiari malformation is an uncommon disorder that mainly occurs in the posterior fossa. The prevalence is relatively unknown, but it is estimated that around one child will suffer from Arnold Chiari malformation in every 1000 births. In this case study, we presented a case of a 45-year-old female suffering from Arnold Chiari malformation type I. Following her clinical examination, she underwent foramen magnum decompression surgery.

Keywords: Arnold-Chiari malformation, case report, epidural anaesthesia, tonsillar herniation

Introduction

Arnold Chiari malformation is a structural abnormality of the brain's cerebellum, which controls stability (1). In this syndrome, there could be an herniation of the cerebellar tonsil into the spinal canal through the foramen magnum to a complete absence of the cerebellum. Depending upon the severity, the malformation is divided into four types. Which are Arnold Chiari malformations type I, type II, type III, and type IV (2). Previously, scientists held the belief that the disease was extremely rare. But due to the advancement of medical imaging techniques such as CT scans and MRIs, we could see the disease is not rare anymore (1). However, the majority of the type I CM are asymptomatic, and not all the patients can afford to access the imaging techniques. Therefore, it is believed that the worldwide cases are under estimated. Some of the risk factors are increasing age, increasing size of cerebellar tonsils greater than 5 mm, being female, ethnicity, and race. To make a diagnosis, further causes of cerebellar tonsillar ectopy must be ruled out in addition to a physical examination, additional testing, neurological assessment, and neuroimaging procedures including CT and MRI (3). In India, we don't have any specific data on the total number of people who got affected by the ACM type I, as it is usually asymptomatic until adulthood (4). Moreover, the true pathogenesis of the disease

is still unknown, so to avoid further challenges, it is crucial to diagnose the illness as soon as possible. In this study, we report a case of a 45-year-old female who presented with a chief complaint of suboccipital headache and paraesthesia of limbs.

Case Report

A 45-year-old female was admitted to Apollo Hospital in the department of neurosurgery on 28 June 2024 with the complaint of headache and neck pain, where she was provisionally diagnosed with an Arnold Chiari malformation. Later after the MRI, her final diagnosis was an Arnold Chiari malformation with cerebellar tonsil herniation through foramen magnum.

Clinical Signs

On examination, patients complain of headache and neck pain since three months, unable to sit for long durations. However, the patient did not have any signs of tingling sensations or numbness of limbs. CVS: S1 S2 positive, P/A soft and nontender, PAC panel II, HMF: intact, CN: intact. Furthermore, the patient did not have a history of falls or previous episodes of vomiting.

²Department of Psychiatric Nursing, Arya College of Nursing, Guwahati, India

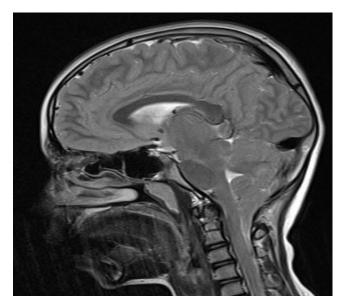


Figure 1. MRI shows tonsillar herniation

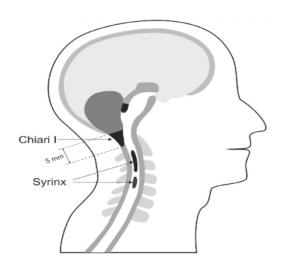


Figure 2. Anatomical positioning of the chiari 1 and syrinx

Investigations

On the arrival of the patient, the relevant investigations were done. Blood grouping and typing test results show ABO group: B, Rh (D) type: positive, Anti-HCV antibodies: non-reactive; HBsAg: non-reactive; retrovirus test: non-reactive. EDTA result shows platelet count: 163 103 / mm3, WBC count: 7.63 103 / mm3, neutrophils: 71.0*%, lymphocytes: 23.9*%, eosinophils: 2.0%, monocytes: 2.6%, basophils: 0.5%, ESR: 15 mm/1st hr, Hb: 11.5* g/dl. LFT function test reveals albumin serum: 3.4* g/dl, globulin serum: 3.7* g/dl, albumin-globulin serum: 1*. MRI cervical spine reveals disc osteophyte complex at C5-C6 disc level intending the ventral thecal sac without spinal canal stenosis or significant nerve compression and low lying cerebellar tonsils. It is noted that the tip of the cerebellar tonsil is approximately 2.9 mm below the level of the foramen magnum.

Therapeutic Intervention

After the confirmation of Arnold Chiari malformation type, I foramen magnum decompression surgery was done, and all the vitals were stable post-operatively. Furthermore, the patient was prescribed Tab peuron-CD3 10' S OD X 3 months, Cap sutril Q10 OD X 6 weeks, Tab pregadoc NT OD X HS X 3 months, Inj ceftriaxone 1gm X 1V X 12 hourly, Inj Pan 40 mg X IV X 24 hourly, Injdexa X 4 mg X IV X 8 hourly, and Inj MVI 100 mg with 100 ml NS X IV X 24 hourly. Furthermore, the patient was also advised to use a cervical collar.

Discussion

Arnold Chiari malformation is commonly diagnosed with the help of MRI testing. However, the fundamental challenge lies in the early radiological diagnosis. Still, the exact cause is unknown, but it was suggested that MTHFD 1 G1958A gene polymorphism is strongly linked with the neural tube defect. Furthermore, few more genes are detected that are associated with the adult Chiari malformation type 1, such as ALDH1A2, CDX1, and FLT1 (5). The herniation is diagnosed if one or both cerebellar tonsils are 5 mm or below the basion-opisthion line as measured on the midline sagittal T1-weighted MRI scan. However, the pathophysiology is far more complex, where there could be different tonsiller descent. Several previous studies also reported different unique and distinctive features of the tonsillar descent (6). In our case, the patient was suffering from Chiari type 1, which is most common among all the Chiari types, and it mostly affects the females, with 0.5 to 3.5% of the population. In this type, the bones of the sull base were often underdeveloped, which results in posterior fossa volume reduction (7) and is frequently associated with syringomyelia. The management of the Arnold Chiari malformation depends upon the severity and should be corelated clinically. Asymptomatic patients without having hydrocephalus or syrinx can be managed conservatively with a six-month follow-up period. However, the diagnosis should be done early to have a better prognosis. If the patients are symptomatic with symptoms like myopathy, severe headache and neck pain, medullary compression, and syringomyelia, they usually require a ventriculoperitoneal shunt. Cranial cervical decompression surgery can also be performed in case of the presence of syrinx (8). In this case, foramen magnum decompression surgery was done, and the surgery was successful. A post-operative check was done for several days, and patient vitals and all the other parameters were stable, and there was no sign of any infection. There was optimal pain control during the surgery due to the epidural anesthesia, and the patient was able to get up after 24 hours of surgery.

Conclusion

Arnold Chiari malformation is common nowadays. However, many times patients are undiagnosed due to the lack of symptoms. However, early diagnosis is important to manage the symptoms and also to improve the quality of life. Also, the physicians should consider the possibility that Arnold Chiari malformation in cases of acute respiratory failure is unexplainable.

Acknowledgements: We thank the patients for allowing us to share her case

Ethical Approval: Ethical approval were taken as per international standards

Conflicts of Interests: None declared

Funding: The authors receive no funding for this work

- **1.** Ansorge R. Chiari Malformation [Internet]. WebMD. 2022. Available from: https://www.webmd.com/brain/chiarimalformation-symptoms-types-treatment
- **2.** Rodríguez-Blanque R, Almazán-Soto C, Piqueras-Sola B, Sánchez-García JC, Reinoso-Cobo A, Menor-Rodríguez MJ,

- Cortés-Martín J. Chiari Syndrome: Advances in Epidemiology and Pathogenesis: A Systematic Review. J Clin Med. 2023 Oct 23;12(20):6694. doi: 10.3390/jcm12206694. PMID: 37892831; PMCID: PMC10607306.
- **3.** Rodríguez EM. Chiari Malformations: A Review of the Current Literature. 2023 Mar 1; Available from: https://doi.org/10.37191/mapsci-actcr-2(1)-20
- 4. Maurya MR, Ravi R, Pungavkar SA. A case report of Arnold Chiari type 1 malformation in acromesomelic dwarf infant. Pan Afr Med J. 2021 Jan 18;38:58. doi: 10.11604/pamj.2021.38.58.27295. PMID: 33854687; PMCID: PMC8017357.
- Maurya MR, Ravi R, Pungavkar SA. A case report of Arnold Chiari type 1 malformation in acromesomelic dwarf infant. Pan African Medical Journal [Internet]. 2021 Jan 1;38. Available from: https://doi.org/10.11604/pamj.2021.38.58.27295
- Park RJ, Unnikrishnan S, Berliner J, Magnussen J, Liu S, Stoodley MA. Cerebellar Tonsillar Descent Mimicking Chiari Malformation. Journal of Clinical Medicine [Internet]. 2023 Apr 9;12(8):2786. Available from: https://doi.org/10.3390/jcm12082786
- Hidalgo JA, Tork CA, Varacallo M. Arnold-Chiari Malformation. [Updated 2023 Sep 4]. In: StatPearls [Internet]. Treasure Island (FL): StatPearls Publishing; 2024 Jan-. Available from: https://www.ncbi.nlm.nih.gov/books/NBK431076/
- Cociasu I, Pastia M, Davidescu I, Buraga I, Popescu BO. CHIARI MALFORMATION TYPE I CASE REPORT AND REVIEW OF LITERATURE. Romanian Journal of Neurology [Internet]. 2016 Sep 30;15(3):138–142. Available from: https://doi.org/10.37897/rjn.2016.3.6

Journal of Emergency Medicine Case Reports

Disproportionate Short Stature in Nail Patella Syndrome: Clinical, Radiological, and Genetic Insights

📵 Yüksel Yaşartekin¹, 📵 Onur Dirican², 📵 Abbas Ali Husseini³⁴, 📵 Ayşe Derya Buluş¹, 📵 Uğur Ufuk Işın¹, 📵 Mehmet Ali Ergün⁵

¹Pediatric Endocrinology, Ankara Atatürk Sanatoryum Training and Education Hospital, Ankara, Türkiye

Abstract

This case report presents an 11-year-old male with disproportionate short stature as a rare manifestation of Nail Patella Syndrome (NPS). NPS is an autosomal dominant disorder caused by mutations in the LMX1B gene and is typically characterized by nail dysplasia, skeletal anomalies, and renal complications. The patient exhibited classical features of NPS, including dysmorphic traits, nail abnormalities, and radiological evidence of patellar aplasia and iliac horns. Genetic analysis identified a p.Arg221Ter pathogenic variant in LMX1B, confirming the diagnosis. Importantly, comprehensive evaluations excluded endocrine, renal, and systemic causes for short stature, suggesting it as a direct, albeit uncommon, feature of NPS. This case underscores the importance of genetic testing in diagnosing atypical presentations and highlights the need for tailored management, including regular monitoring for renal and ophthal-mological complications. By documenting short stature in NPS, this report expands its clinical spectrum, contributing valuable insights to the understanding of this complex condition. Keywords: Chronic pancreatitis, complication, ductal rupture

Keywords: Childhood anomalies, nail-patella syndrome, short stature

Introduction

Nail Patella Syndrome (NPS), also known as hereditary osteoonychodysplasia or Turner-Kieser syndrome, is a rare genetic condition with an incidence ranging from 4.5 to 22 per million people (1,2). It is inherited in an autosomal dominant manner, affecting both genders equally. The causative mutations are found in the LIM homeobox transcription factor 1-beta (LMX1B) gene, located on chromosome 9q33.3 (3-5). This gene encodes a transcription factor that is essential for the development of dorsal limb structures, podocyte morphogenesis and fusion with the glomerular basement membrane, and the formation of the anterior chamber of the eye (6). The LMX1B gene is also involved in the development of dopaminergic and serotonergic neurons in the midbrain and posterior parietal cortex, as well as spinal interneurons in the central nervous system (7).

Clinical manifestations of NPS include short stature, particularly rhizomelic shortening, and less frequently, hydrocephalus, true megalencephaly, midface hypoplasia, trident hand configuration, and joint hyperextension (8,9). NPS was first described in 1820 by Chatelain (10) and

later insights into its familial and hereditary nature were provided by Pye-Smith in 1883 and Little in 1897 (11). The clinical definition of NPS is based on a tetrad of features: nail dysplasia, hypoplastic or absent patella, radial head dislocation, and iliac horns with bony prominences on the iliac bones (1,2).

Nail abnormalities affect approximately 98% of NPS patients, primarily involving the thumbs and index fingers symmetrically. These abnormalities include dystrophy, hypoplasia, or nail absence, often accompanied by longitudinal or horizontal stripes that may change color and be separated by longitudinal cracks. An abnormal lunula, which may be triangular or absent, is a pathognomonic feature of NPS, most noticeable on the thumbs [12]. Soft tissue complications include renal dysplasia, muscle weakness, hearing loss, shaky gait, and winging of the scapula. Renal involvement is seen in 30-50% of NPS cases and significantly affects prognosis. Proteinuria, with or without hematuria, is common and may progress to end-stage renal disease over time (13,14).

While the classical features of NPS are well-documented, disproportionate short stature remains an uncommon and underexplored manifestation. Although reports linking NPS

Corresponding Author: Onur Dirican e-mail: odirican@gelisim.edu.tr Received: 07.11.2024 • Revision: 25.11.2024 • Accepted: 25.12.2024 DOI: 10.33706/jemcr.1497983

©Copyright 2020 by Emergency Physicians Association of Turkey - Available online at www.jemcr.com

Cite this article as: Yaşartekin Y, Dirican O, Ali Husseini A, Buluş AD, Işin UU, Ergün MA. Disproportionate Short Stature in Nail Patella Syndrome: Clinical, Radiological, and Genetic Insights. Journal of Emergency Medicine Case Reports. 2025;16(1): 7-11

²Department of Pathology Laboratory Techniques, Vocational School of Health Services, Istanbul Gelişim University, İstanbul, Türkiye

³Life Science, and Biomedical Engineering Application and Research Center, Istanbul Gelisim University, İstanbul, Türkiye

⁴Vocational School of Health Services, Istanbul Gelisim University, Istanbul, Türkiye

⁵Medical Genetic, Gazi University Hospital, Ankara, Türkiye

to short stature are scarce in the literature, we present a rare case of disproportionate short stature in a patient with Nail Patella Syndrome. NPS is frequently associated with renal anomalies; however, this case is notable for contributing to the limited knowledge of its association with growth abnormalities. By reviewing this presentation in the context of short stature and dysmorphic features, this report aims to enhance understanding of the broader phenotypic spectrum of NPS and its implications for clinical management.

Case Report

A patient with short stature, monitored in our outpatient clinic from the age of 9 to 11 years, was evaluated. His birth weight was 3600 grams, with no reported postpartum complications. On physical examination, the patient's height was 130 cm (<3rd percentile), body weight was 40.9 kg (25th–50th percentile), and he was at Tanner stage 2. Dysmorphic features were observed (Figure-1a). The patient's maternal height was 148.2 cm, paternal height was 158 cm, and mid-parental height was calculated at 159.75 cm. His parents, who are cousins, had no known medical conditions or dysmorphic features.

Laboratory and Hormonal Investigations

Laboratory screening included a growth hormone stimulation test using L-Dopa and clonidine, which showed normal results. Specifically, theL-Dopa stimulation was 9 ng/mL, clonidine stimulation was 13 ng/mL, thyroid-stimulating hormone (TSH) was 1.2 mIU/L, and free T4 was 1.3 ng/dL. These values were within normal reference ranges,

indicating no growth hormone deficiency, normal thyroid function, and no evidence of hypothyroidism.

Clinical Examination and Radiological Findings

The patient exhibited frail upper limbs with straight, grooved fingernails, predominantly affecting the 1st and 2nd metacarpals (Figure-1b). Similar abnormalities were observed in the toenails (Figure-1c). Both elbows showed a full range of motion, with no crepitation and unrestricted pronation and supination. A careful assessment of the lower extremities revealed equal leg lengths and squareness of the knees, with no signs of ligament laxity. The feet were noted to be in a valgus position.

Radiographs of the knees revealed complete absence of the patella on both sides, along with moderately hypoplastic lateral femoral condyles (Figure-2a). Pathognomonic posterior iliac horns, characteristic of Nail Patella Syndrome (NPS), were evident (Figure-2b).

Genetic Analysis

Given the patient's dysmorphic features, short stature, and abnormal radiographic findings, genetic analysis was pursued. Sanger sequencing identified a pathogenic variant in the LMX1B gene: NM 002316.3(LMX1B):c.661C>T (p.Arg221Ter), associated with dbSNP identifier rs121909487. This mutation is known to cause phenotypic features such as nail dysplasia, swan-neck toes, and patellar dysplasia, consistent with the clinical presentation. The same mutation was not observed in parental analysis. Along with clinical examination of the parents, who do not display any features characteristic of NPS, suggests the mutation is likely de novo.

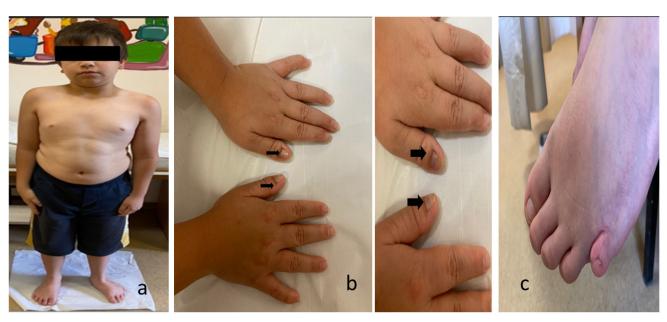


Figure 1. Characteristics of Nail-Patella Syndrome with Short Stature: a; Dysmorphic features observed during physical examination. b; Clinical image illustrating subtle discoloration and dystrophy of nails, with mild ridges primarily evident in the thumbs, index, and middle fingers on both sides. c; Presentation of dystrophic toenails associated with Nail-Patella Syndrome.



Figure 2. Radiographic Features of Nail-Patella Syndrome with Short Stature: a; X-ray depicting the absence of patellae. B; Pelvic X-ray revealing the presence of bilateral iliac horns.

Follow-up and Diagnosis Confirmation

At the final evaluation at age 11, the patient's height remained 130 cm, corresponding to -2.01 SD. NPS was diagnosed based on the identified genetic mutation, as well as the clinical and radiological findings. Kidney and ophthalmological screenings were performed to assess any potential complications. No evidence of renal disease, such as proteinuria or hematuria, was found, and renal ultrasound results were normal. Ophthalmological examination also revealed no abnormalities.

Discussion

Documented cases of Nail Patella Syndrome (NPS) associated with short stature are scarce, with limited literature specifically addressing this rare manifestation. This case report enhances the existing knowledge by identifying disproportionate short stature as an uncommon feature of NPS, underscoring the critical role of genetic testing, and offering a comprehensive clinical, radiological, and diagnostic profile to guide future research and clinical management.

The patient demonstrated disproportionate short stature, a rare feature in Nail Patella Syndrome (NPS). A previous case series reported two pediatric patients with NPS presenting for short stature evaluation, underscoring that while this association is uncommon, it is documented in the literature (15). Clinical manifestations of NPS are highly variable, and although short stature is not a defining characteristic, it has been observed in some cases alongside classical NPS features. For example, a study described a family with multiple affected members exhibiting varying degrees of short stature in conjunction with the typical symptoms of NPS [16,17]. In certain cases, endocrine factors such as hypothyroidism have been implicated as contributing factors to growth impairment in children with NPS [12]. The

role of genetic variations in the LMX1B gene has also been highlighted, as exemplified by Alvarez Martin et al., who identified a c.728G>C (p.Trp243Ser) mutation in a patient presenting with short stature (16). Similarly, Lindelöf H et al. reported an inversion of the LMX1B gene in a Swedish family, all members of whom exhibited varying degrees of short stature, further expanding the genetic spectrum associated with this phenotype (17). Recently, Jang J et al. reported a deletion variant in the LMX1B gene, specifically NM_001174147.2(LMX1B):c.641_887-556delinsGG, p.(Lys214Argfs*11), associated with Nail Patella Syndrome (NPS); however, short stature was not identified as a typical characteristic in their findings (18).

Normal laboratory and hormonal profiles along with no evidence of proteinuria, hematuria, renal abnormalities, or ophthalmological issues was found during comprehensive screenings. These results indicate that the patient's short stature is unlikely due to systemic conditions such as growth hormone deficiency, thyroid dysfunction, or renal complications, which are common contributors to growth impairment. Additionally, the absence of proteinuria, hematuria, renal abnormalities, and ophthalmological issues suggests that the patient does not exhibit some of the more typical complications associated with Nail Patella Syndrome (NPS). This reinforces the conclusion that the disproportionate short stature observed in this case is a rare and isolated manifestation of NPS rather than a secondary effect of other systemic abnormalities.

Despite full genetic penetrance in familial contexts, Nail Patella Syndrome (NPS) exhibits a variable degree of clinical manifestation. The syndrome lacks specific diagnostic criteria; however, it is often diagnosed based on a combination of clinical and imaging findings (18-22). In cases where clinical evaluation is inconclusive, genetic testing provides a valuable diagnostic tool (23). LMX1B, a member of the LIM-homeodomain family of transcription factors, plays a critical role in establishing ventral-dorsal body patterning during embryonic development (13).

NPS warrants careful attention due to its association with complications carrying significant morbidity and mortality, particularly chronic renal failure, with a reported incidence of 5%, and glaucoma. Renal involvement, characterized by hematuria and proteinuria that can progress to the nephrotic range during childhood or adolescence, occurs in approximately 40% of cases (23). Annual urine testing, including microalbuminuria/creatinine ratio assessment, and regular blood pressure monitoring are recommended. Ophthalmological complications affect 35% of NPS patients (6), with primary open-angle glaucoma, normal-tension glaucoma, ocular hypertension, and isolated glaucomatous optic disc lesions more frequently manifesting at a younger age compared to the general population. Annual ophthalmological evaluations are therefore advised (24). In the present case, renal and ophthalmological evaluations revealed no abnormalities.

NPS is a condition that must not be overlooked due to its association with complications of significant morbidity, including chronic renal failure and glaucoma. Renal involvement, evident in 40% of cases, underscores the need for vigilant monitoring. Nail dysplasia and patellar aplasia or hypoplasia are hallmark features of NPS (21). Thumb anomalies are particularly severe; in this case, the patient exhibited longitudinal protrusions. While toenail involvement is less common, the presence of triangular lunulae is considered pathognomonic (21). In this case, toenails were not involved. Nail dysplasias in NPS can include horizontal or longitudinal protrusions, pitting, or longitudinal clefts. Patellar anomalies, such as absence, hypoplasia, or abnormal formation, are frequently associated with recurrent patellar subluxation, indicative of knee involvement. Elbow abnormalities, including restricted joint movement, capitellum hypoplasia, and radial head subluxation, can occur, often asymmetrically (25). Iliac horns, bony projections from the middle of the outer iliac fossa, are pathognomonic for NPS and present in over 80% of cases (22,26). Radiographs in this patient revealed complete absence of the patella bilaterally and moderately hypoplastic lateral femoral condyles (Figure-1b).

NPS manifestations extend to sensorineural hearing loss, gastrointestinal involvement, and neurological or vasomotor dysfunction. Skeletal deformities such as pes planus, pes equinovarus, pectus excavatum, and scoliosis have also been reported (27). Short stature in NPS patients is inconsistently described in the literature (23-27). While individuals with NPS are, on average, shorter than the general population, this difference is not statistically significant. A study of 89 British patients reported a mean final height of 170.9 cm (-0.77 SD) in males and 158.5 cm (-0.71 SD) in females[6,23-25,28,29]. Notably, only a few reports, including two Tunisian sisters with short stature (-4.0 SD) and a Spanish case of short stature associated with hypothyroidism, link short stature to NPS (12). In this case, the patient harbored a p.Arg221Ter pathogenic variant, a mutation previously described in the literature (28). The patient presented with disproportionate short stature (-2.01 SD) alongside nail and finger anomalies but demonstrated normal hormonal and bone age assessments.

Conclusion

This case highlights disproportionate short stature as a rare feature of Nail Patella Syndrome (NPS), expanding its clinical spectrum. The identification of the *LMX1B* pathogenic variant (p.Arg221Ter) underscores the role of genetic testing in diagnosing atypical presentations. The absence of systemic abnormalities suggests short stature may be a direct feature of NPS rather than a secondary complication. This report emphasizes the importance of comprehensive evaluations and monitoring to improve management and outcomes for individuals with NPS.

- BRIXEY AM, BURKE RM. Arthro-onychodysplasia; hereditary syndrome involving deformity of head of radius, absence of patellas, posterior iliac spurs, dystrophy of finger nails. Am J Med 1950;8:738–44. https://doi.org/10.1016/0002-9343(50)90098-2.
- Schulz-Butulis BA, Welch MD, Norton SA. Nail-patella syndrome. J Am Acad Dermatol 2003;49:1086–7. https://doi. org/10.1016/j.jaad.2002.01.001.
- **3.** Finsterer J, Stöllberger C, Steger C, Cozzarini W. Complete heart block associated with noncompaction, nailpatella syndrome, and mitochondrial myopathy. J Electrocardiol 2007;40:352–4. https://doi.org/10.1016/j.jelectrocard.2006.11.008.
- **4.** Lucas GL, Opitz JM, Wiffler C. The nail-patella syndrome: Clinical and genetic aspects of 5 kindreds with 38 affected family members. J Pediatr 1966;68:273–88. https://doi.org/https://doi.org/10.1016/S0022-3476(66)80159-2.
- Milla E, Gamundi MJ. Novel LMX1B mutation in familial Nail patella syndrome with variable expression of open angle glaucoma. 2007.
- 6. Bongers EMHF, Huysmans FT, Levtchenko E, de Rooy JW, Blickman JG, Admiraal RJC, et al. Genotype-phenotype studies in nail-patella syndrome show that LMX1B mutation location is involved in the risk of developing nephropathy. Eur J Hum Genet 2005;13:935–46. https://doi.org/10.1038/sj.ejhg.5201446.
- 7. Ding YQ, Yin J, Kania A, Zhao ZQ, Johnson RL, Chen ZF. Lmx1b controls the differentiation and migration of the superficial dorsal horn neurons of the spinal cord. Development 2004;131:3693–703. https://doi.org/10.1242/dev.01250.
- Shiang R, Thompson LM, Zhu YZ, Church DM, Fielder TJ, Bocian M, et al. Mutations in the transmembrane domain of FGFR3 cause the most common genetic form of dwarfism, achondroplasia. Cell 1994;78:335–42. https://doi. org/10.1016/0092-8674(94)90302-6.
- Bellus GA, Hefferon TW, Ortiz de Luna RI, Hecht JT, Horton WA, Machado M, et al. Achondroplasia is defined by recurrent G380R mutations of FGFR3. Am J Hum Genet 1995;56:368–73.
- **10.** Guidera KJ, Satterwhite Y, Ogden JA, Pugh L, Ganey T. Nail patella syndrome: a review of 44 orthopaedic patients. J Pediatr Orthop 1991;11:737–42.
- **11.** Bongers EMHF, Gubler M-C, Knoers NVAM. Nail-patella syndrome. Overview on clinical and molecular findings. Pediatr Nephrol 2002;17:703–12. https://doi.org/10.1007/s00467-002-0911-5.
- **12.** Goecke C, Mellado C, García C, García H. Short stature and hypothyroidism in a child with Nail-Patella Syndrome. A case report. Rev Chil Pediatr 2018;89:107–12. https://doi.org/10.4067/S0370-41062018000100107.
- **13.** McIntosh I, Dunston JA, Liu L, Hoover-Fong JE, Sweeney E. Nail patella syndrome revisited: 50 years after linkage. Ann Hum Genet 2005;69:349–63. https://doi.org/10.1111/j.1529-8817.2005.00191.x.
- **14.** Lemley KV. Kidney disease in nail-patella syndrome. Pediatric Nephrology 2009;24:2345–54. https://doi.org/10.1007/s00467-008-0836-8.

- **15.** Haddad S, Ghedira-Besbes L, Bouafsoun C, Hammami S, Chouchene S, Ben Meriem C, et al. Nail-patella syndrome associated with short stature: a case series. Case Rep Med 2010;2010. https://doi.org/10.1155/2010/869470.
- **16.** Álvarez-Martín N, Gamundi MJ, Hernan I, Carballo M, Isabel Luis-Yanes M, García-Nieto V. Nail-patella syndrome. A case with a de novomutation in the LMX1B gene not previously described. Nefrologia 2013;33:585–6. https://doi.org/10.3265/Nefrologia.pre2013.Apr.12006.
- 17. Lindelöf H, Horemuzova E, Voss U, Nordgren A, Grigelioniene G, Hammarsjö A. Case Report: Inversion of LMX1B A Novel Cause of Nail-Patella Syndrome in a Swedish Family and a Longtime Follow-Up. Front Endocrinol (Lausanne) 2022;13. https://doi.org/10.3389/fendo.2022.862908.
- **18.** Jang J, Im H, Lee H, Sung H, Cho SI, Lee J-S, et al. A Family With Nail-Patella Syndrome Caused by a Germline Mosaic Deletion of *LMX1B*. Ann Lab Med 2024;44:625–7. https://doi.org/10.3343/alm.2024.0140.
- **19.** Levy M, Feingold J. Estimating prevalence in singlegene kidney diseases progressing to renal failure. Kidney Int 2000;58:925–43. https://doi.org/10.1046/j.1523-1755.2000.00250.x.
- **20.** Letts M. Hereditary onycho-osteodysplasia (nail-patella syndrome). A three-generation familial study. Orthop Rev 1991;20:267–72.
- **21.** THOMPSON EA, WALKER ET, WEENS HS. Iliac horns; an osseous manifestation of hereditary arthrodysplasia associated with dystrophy of the fingernails. Radiology 1949;53:88–92. https://doi.org/10.1148/53.1.88.
- **22.** Fong EE. "Iliac Horns" (Symmetrical Bilateral Central Posterior Iliac Processes). Radiology 1946;47:517–8. https://doi.org/10.1148/47.5.517.

- 23. Álvarez-Martín N, Gamundi MJ, Hernan I, Carballo M, Luis-Yanes MI, García-Nieto V. Nail-patella syndrome. A case with a de novo mutation in the LMX1B gene not previously described. Nefrologia 2013;33:585–6. https://doi.org/10.3265/Nefrologia.pre2013.Apr.12006.
- **24.** Mimiwati Z, Mackey DA, Craig JE, Mackinnon JR, Rait JL, Liebelt JE, et al. Nail-patella syndrome and its association with glaucoma: a review of eight families. Br J Ophthalmol 2006;90:1505–9. https://doi.org/10.1136/bjo.2006.092619.
- **25.** Sweeney E, Fryer A, Mountford R, Green A, McIntosh I. Nail patella syndrome: a review of the phenotype aided by developmental biology. J Med Genet 2003;40:153–62. https://doi.org/10.1136/jmg.40.3.153.
- **26.** Pinette MG, Ukleja M, Blackstone J. Early prenatal diagnosis of nail-patella syndrome by ultrasonography. J Ultrasound Med 1999;18:387–9. https://doi.org/10.7863/jum.1999.18.5.387.
- 27. Figueroa-Silva O, Vicente A, Agudo A, Baliu-Piqué C, Gómez-Armayones S, Aldunce-Soto MJ, et al. Nail-patella syndrome: report of 11 pediatric cases. J Eur Acad Dermatol Venereol 2016;30:1614–7. https://doi.org/10.1111/jdv.13683.
- **28.** Dreyer SD, Zhou G, Baldini A, Winterpacht A, Zabel B, Cole W, et al. Mutations in LMX1B cause abnormal skeletal patterning and renal dysplasia in nail patella syndrome. Nat Genet 1998;19:47–50. https://doi.org/10.1038/ng0598-47.
- **29.** Beals RK, Eckhardt AL. Hereditary onycho-osteodysplasia (Nail-Patella syndrome). A report of nine kindreds. J Bone Joint Surg Am 1969;51:505–16.

Journal of Emergency Medicine Case Reports

A Case of Pneumopericardium after Pericardiocentesis Conservatively Managed with Multimodality Imaging Methods

Abstract

Pneumopericardium is characterized by the presence of air or gas in the pericardial sac, which can occur spontaneously, due to trauma, underlying disease, or iatrogenically. Following pericardiocentesis, pneumopericardium is a rare but notable complication, and management depends on the hemodynamic status of the patient. A 57-year-old male with lung adenocarcinoma underwent pericardiocentesis for cardiac tamponade. After the procedure, pneumopericardium was diagnosed using multimodal imaging, including chest X-ray, CT scan, and echocardiography. The patient remained hemodynamically stable and was managed conservatively with close follow-up. In stable patients, a conservative approach to pneumopericardium may be appropriate. Multimodal imaging is crucial for diagnosis and management, and timely intervention is necessary in cases of hemodynamic instability.

Keywords: Pericardial disease, pericardial effusion, pericardiocentesis, pneumopericardium

Introduction

Pneumopericardium is defined by the presence of air or gas in the pericardial sac. It can occur spontaneously, following trauma, as a result of an underlying disease, or iatrogenically (1). In most cases, pneumopericardium is asymptomatic and resolves spontaneously without the need for invasive treatment. However, in some cases, especially when complications like cardiac tamponade arise, immediate intervention may be required (1). The management of pneumopericardium is largely dependent on the patient's hemodynamic status and the presence of any accompanying conditions such as pneumothorax (2). Pneumopericardium after pericardiocentesis is an uncommon yet significant complication. In this case report, we describe the diagnosis of pneumopericardium after pericardiocentesis using multimodal imaging techniques and discuss the management approach, emphasizing the conservative treatment approach due to the patient's stable hemodynamics.

Case Report

A 57-year-old male patient who received carboplatin and paclitaxel chemotherapy for lung adenocarcinoma underwent pericardiocentesis due to cardiac tamponade. A 6-FR sheath and a pigtail catheter were used and approximately 250 cc

of fluid was drained immediately. A total of 1200 cc of fluid was drained in 36 hours.

Then we removed the pericardiocentesis system and the patient was in a stable condition both clinically and in terms of vital signs. On a follow-up chest X-ray (Figure-A) taken on the same day after pericardiocentesis, the pericardial layer was distinguishable (red arrow) and a radiolucent area is noted between the heart and the pericardial layer. On the thorax CT (Figure-B) taken with the suspicion of pneumopericardium, it was observed that the pericardial space is filled with air (blue arrow), and accumulation is particularly observed in the anterior region, probably due to the low density of air. Accompanying pneumothorax was not observed on CT. In echocardiography (echo) (Figure-C), air bubbles are observed in the pericardial space and they are seen to change location from beat to beat.

The patient was taken back to the ICU for observation and daily echo monitoring. We discharged the patient after 6 days because there was no deterioration in the patient's clinical condition and the echo appearance was stable on follow-ups.

In this case, a conservative approach was preferred because the patient was hemodynamically stable and diastolic collapse of the right ventricle was not detected. During follow-up, it was observed that the air appearance disappeared. On the thorax CT (Figure-D) taken 4 months after pericardiocentesis, no air is seen between the pericardial

Corresponding Author: Serhat Kesriklioğlu e-mail: drserhatk@gmail.com

Received: 01.11.2024 • **Revision:** 21.12.2024 • **Accepted:** 29.12.2024

DOI: 10.33706/jemcr.1577878

©Copyright 2020 by Emergency Physicians Association of Turkey - Available online at www.jemcr.com

Cite this article as: Gürbüz AS, Kesriklioğlu S, Şahin AT. A Case of Pneumopericardium after Pericardiocentesis Conservatively Managed with Multimodality Imaging Methods. Journal of Emergency Medicine Case Reports. 2025;16(1): 12-13



Figure 1 A-B-C-D. A: After pericardiocentesis, the pericardial layer can be distinguished (red arrow), B: The pericardial space is filled with air (blue arrow), C: In echocardiography, air bubbles are observed in the pericardial space, D: On thoracic CT four months after pericardiocentesis, no residual air was seen between the pericardial layers but the effusion recurred.

layers. Despite this, the effusion recurred and the patient was referred for pericardioperitoneal window surgery.

No significant increase in effusion was observed after the successful surgical procedure. However, eight months after the first contact, metastatic foci were detected in the brain and the patient passed away due to non cardiac reasons.

Discussion

In the treatment of malignant pericardial effusions, options such as pericardiocentesis, percutaneous balloon pericardiotomy, catheter drainage or surgical pericardial window opening can be chosen (3).

In the pericardial fluid sample taken from the patient, the fluid was found to be exudative in nature, but no evidence of metastasis was observed. It should be noted that in cancer patients, pericardial fluid may be related to direct metastatic tumor spread, obstruction of lymphatic channels, or treatment-related factors (4).

Recurrence of malignant effusions varies depending on the type of primary cancer (5). Considering that the risk of recurrence is higher, especially in lung adenocarcinoma, pericardial window surgery may be considered at the first stage.

Pneumopericardium may develop due to trauma, mechanical ventilation, infection, invasive intervention, or may occur spontaneously (6). Pneumopericardium is an uncommon occurrence following pericardiocentesis. Pneumopericardium after pericardiocentesis may result from excessive dilation of the skin orifice, air leakage due to a defect in the drainage system, or a pleuropericardial fistula (7).

Pneumopericardium may be asymptomatic, or it may cause symptoms such as chest discomfort, difficulty

breathing, syncope, or even cause cardiac tamponade. It is diagnosed by the presence of air in the pericardial space. Detecting pneumopericardium can be achieved through Standard chest X-rays, CT scans, or echocardiography (1).

In the management of the patient, the hemodynamic status, possible association of accompanying pathologies such as pneumothorax and self-limitation of pneumopericardium should be taken into consideration. The process of differentiate between diagnosis should involve pneumomediastinum, where air is generally not present around the base of the heart and is not restricted solely to the heart, but may extend into the upper mediastinum and neck (8). We preferred a conservative approach because our patient's hemodynamics were stable, no pneumothorax was detected, and we saw that pneumopericardium was self-limiting during the follow-up during his stay (2).

Conclusion

In patients with stable hemodynamics, a conservative approach can be considered at the first stage. Being familiar with these images of pneumopericardium is important to prevent unnecessary interventions.

- **1.** Choi WH, Hwang YM, Park MY, Lee SJ, Lee HY, Kim SW, et al. Pneumopericardium as a complication of pericardiocentesis. KoreanCircJ2011;41(5):280-2.doi:10.4070/kcj.2011.41.5.280.
- **2.** Triantafyllis AS, Zamfir T, Kontogiannis N. Pneumopericardium as a complication of pericardiocentesis. CMAJ 2023; 195(5): E193-E194. doi: 10.1503/cmai.221137.
- Mudra SE, Rayes D, Kumar AK, Li JZ, Njus M, McGowan K, et al. Malignant pericardial effusion: A systematic review. CJC Open 2024; 6(8): 967-972. doi: 10.1016/j.cjco.2024.05.003.
- **4.** Ekmekçi C, Ekmekçi S, Dere Y, Adalı Y, Ekinci S, Çabuk AK, et al. Could the cytological evaluation of pericardial effusions illuminate our path? *İzmir Tepecik Eğitim Hastanesi Dergisi* 2018; 28(2): 99-103. doi: 10.5222/terh.2018.099.
- **5.** Kim SH, Kwak MH, Park S, Kim HJ, Lee HS, Kim MS, et al. Clinical characteristics of malignant pericardial effusion associated with recurrence and survival. Cancer Res Treat 2010; 42(4): 210-216. doi: 10.4143/crt.2010.42.4.210.
- 6. Polistina GE, Lanza M, DiSomma C, Annunziata A, Fiorentino G. A rare evolution to pneumopericardium in a patient with COVID-19 pneumonia treated with high flow nasal cannula. Medicina (Kaunas) 2021; 57(10): 1122. doi: 10.3390/medicina57101122.
- **7.** Palma A, Henriques C, Silva PV, Pires A. Pneumopericardium and pleural effusion: a rare complication of pediatric pericardiocentesis. BMJ Case Rep 2020; 13(11): e236308. doi: 10.1136/bcr-2020-236308.
- **8.** Brander L, Ramsay D, Dreier D, Peter M, Graeni R. Continuous left hemidiaphragm sign revisited: a case of spontaneous pneumopericardium and literature review. Heart (BrCardSoc) 2002; 88(4): e5. doi: 10.1136/heart.88.4.e5.

Journal of Emergency Medicine Case Reports

A Rare Case Requiring Emergency Tracheotomy, Isolated Tracheal Mucormycosis: It is Important to Suspect First

Abstract

Mucormycosis is asignificant opportunistic infection, ranking as the third most common invasive fungal infection after candidiasis and aspergillosis. A 67-year-old male patient was evaluated for progressive shortness of breath. Intubation tube could not be passed through the subglottic region. An emergency trache-otomy was performed. After the tracheal incision, a gray mass and necrotic material were encountered in the tracheal lumen. The tracheal biopsy result was mucormycosis. In emergency services, situations requiring emergency airway access due to difficult intubation can often be linked to central airway obstruction. For immunocompromised diabetic patients, it is important to consider the possibility of a fungal infection in the tracheobronchial tree that may need immediate treatment. To facilitate a quick differential diagnosis, perioperative frozen examinations of the obstructive mass are recommended.

Keywords: Emergency tracheotomy, invasive fungal infection, tracheal mucormycosis, upper airway obstruction

Introduction

Situations necessitating emergency airway access due to difficult intubation or inadequate ventilation are rare but life-threatening and cricothyrotomy is the quickest method to secure airway access in emergency situations (1). However, in a 'can not intubate can not oxygenate' situation arising from central airway obstruction, cricothyrotomy does not bypass the central airway and an open surgical tracheotomy or bronchoscopy becomes imperative as soon as possible (2).

Mucormycosis is asignificant opportunistic infection, ranking as the third most common invasive fungal infection after candidiasis and aspergillosis but, isolated tracheal involvement is rare (3).Patients with immunosuppression, diabetes mellitus (DM), or hematological malignancy are particularly at a higher risk. The prompt diagnosis and treatment of potential invasive fungal infections in at-risk patients can be lifesaving (3).

In this article, we present a case of isolated tracheal mucormycosis that necessiated emergency tracheotomy. We discuss the management of the airway and the importance of urgent treatment of invasive fungal infection.

Case Report

A 67-year-old male patient was admitted to the nephrology department of a tertiary care institution due to chronic renal failure (CRF) and uncontrolled DM. It was discovered that the patient had been undergoing an immunosuppressive treatment for the past six months due to chronic inflammatory bowel disease. Subsequently, the patient was referred to the Otorhinolaryngology department of our hospital due to the development of progressive dysphonia and cough. Indirect laryngoscopy revealed slight edema and mucoid material on the vocal cords. Treatment for acute laryngitis commenced with third-generation Cephalosporin (1gr/day), antireflux, and antimucolytic therapy. Given the ongoing COVID-19 pandemic, we conducted the COVID-19 polymerase chain reaction (PCR) test on the patient. COVID-19 PCR on nasopharyngeal swab was negative for four times. The patient, whose shortness of breath worsened rapidly within five days, was treated with meropenem, linezolid, and colistin upon the recommendation of an infectious diseases specialist, and hemodialysis was performed due to hyperkalemia. His condition continued to deteriorate leading to clinical hypercapnic respiratory acidosis. Intubation was attempted,

Corresponding Author: Bilge Tuna e-mail: opdrbilgetuna@gmail.com Received: 19.03.2024 • Revision: 30.09.2024 • Accepted: 30.12.2024 DOI: 10.33706/jemcr.1442492

©Copyright 2020 by Emergency Physicians Association of Turkey - Available online at www.iemcr.com

Cite this article as: Tuna B, Tüzemen G, Kıyım Altıntaş E. A Rare Case Requiring Emergency Tracheotomy, Isolated Tracheal Mucormycosis: It is Important to Suspect First. Journal of Emergency Medicine Case Reports. 2025;16(1): 14-18

but severe resistance prevented the passage of endotracheal tube deep enough through the subglottis. Considering an intratracheal obstruction, emergency tracheotomy was performed. During the tracheotomy, as the incision made on the trachea through the intercartilaginous space, necrotic material was encountered in the tracheal lumen. Direct laryngoscopy indicated a normal laryngeal passage, but a grey mass, 2.5 cm below the vocal cords, narrowed the trachea from the anterior part. Initially suspecting a tumor or a granulomatous mass occlusion, perioperative tracheal biopsies were taken for differential diagnosis. The patient was then transferred to the intensive care unit and underwent bronchoscopy due to recurrent airway obstruction with necrotic material. During the operation, necrotic materials were removed from the tracheal lumen by fiberoptic bronchoscopy, and the main bronchi were found to be normal. Postoperatively, serum blood tests, blood culture and radiological imaging were conducted. C-reactive protein was high with accompanying leukocytosis in the hemogram. Blood culture identified Enterococcus faecium, while sputum culture and serum tests for galactomannan antigen, c-ANCA and p-ANCA were negative. Laryngeal computed tomography (CT) displayed an endotracheal mass (Figure -1) located from the inferior part of the cricoid cartilage to the third tracheal ring. Paranasal sinus CT revealed maxillary and ethmoidal inflammation findings, while thoracal and cranial CT scans revealed normal results.

The tracheal biopsy result was finalized 11 days after the tracheotomy, with the pathologist reporting mucormycosis (Figure-2). Liposomal Amphotericin B (Amp B) was administered intravenously, starting with 1mg/kg/day and gradually increasing to 5 mg/kg/day. Functional endoscopic sinus surgery was conducted to investigate another potential focus of fungal infection. Nonspecific inflammation was observed during sinus surgery, and Candida nonalbicans was identified in the sinonasal biopsy culture. However, the recurrence of accumulated intratracheal necrotic material and hypercapnic respiratory acidosis persisted, ultimately resulting in the patient's demise on the twentieth day of tracheotomy. As no other focus was identified during the direct examination of the larynx with laryngoscopy and broncoscopy and normal findings were observed on laryngeal, cerebellar and thoracal CT scans, the case was classified as isolated tracheal mucormycosis.

(Permission to use the patient's data was obtained from the patient's wife and the local ethics committee of our hospital.)

Discussion

In this report, we present a male patient with DM and CRF, who could not be intubated due to tracheal obstructive pathology and underwent emergency tracheotomy. Initially,

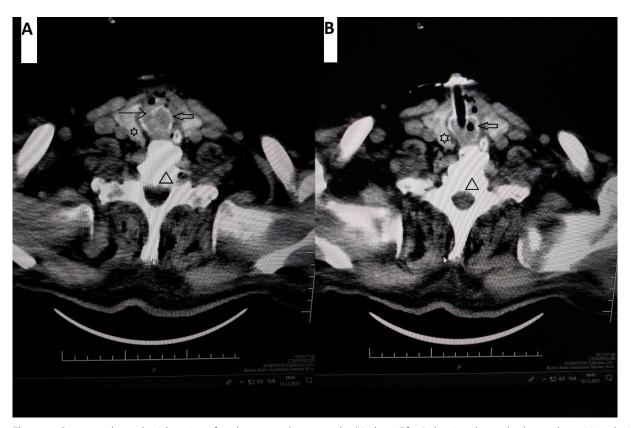


Figure 1. Contrast-enhanced axial section of neck computed tomography (Neck 5.0 Bf 37) showing the tracheal cannula positioned within the trachea. Line arrow: soft tissue mass in the anterior part of tracheal lumen. Hollow arrow: third tracheal ring. Star: right lobe of thyroid gland. Triangle: body of seventh Cervical vertebra.

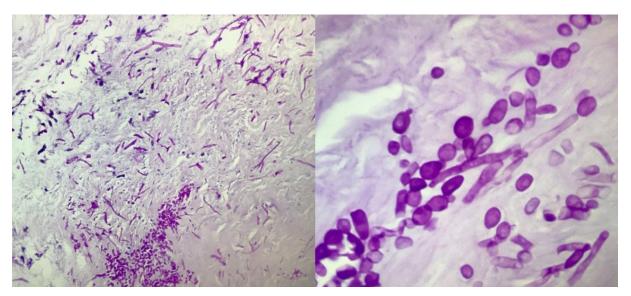


Figure 2. Photomicrographs showing large aseptate fungal hyphae in necrotic ground consistent with mucormycosis morphology (hematoxylin and eosin stain, magnification x100 and magnification x400).

it was suspected that the cause of tracheal obstruction might be a tumoral or granulomatous pathology. Perioperative biopsies were taken from the grey mass and necrotic materials in the trachea. The patient was treated with broad-spectrum antibiotics and during this period, the patient underwent repeated bronchoscopies due to the reaccumulation of obstructive necrotic material. Eleven days after tracheotomy, histopathological diagnosis of mucormycosis was reported, and antifungal treatment was initiated. Unfortunately, the patient passed away twenty days after the tracheotomy procedure. When we noticed the obstruction in the trachea, we suspected a tumor and proceeded with biopsies. Unfortunately, it took eleven days for the biopsy results to be reported, which also caused a delay in initiating the mucor treatment. Since the primary focus persisted in the trachea, the recurrence of necrotic material and airway obstruction continued. By performing a frozen section analysis and ensuring airway safety, healthcare professionals can accurately diagnose invasive infections that need prompt attention. If a mucor infection is present, it is important to surgically remove the affected areas completely and start antifungal treatment as soon as possible. This timely intervention can potentially change the course of the patient's clinical outcome. The most important thing to consider in such patients is to maintain a high level of suspicion.

Neoplasia and post-intubation tracheal stenosis are common causes of airway obstruction. CT of the neck and chest using intravenous contrast in the inspiratory and expiratory phases is the gold standard imaging for suspected tracheal obstruction (4). Causes of tracheal obstruction include granulomatous infections and, rarely, fungal infections (5). It is important to ensure airway patency immediately. Most guidelines for the management of central airway obstruction recommend awake endotracheal intubation, tracheotomy,

laryngeal mask airway, or suspension laryngoscopy, with a high likelihood of requiring subsequent rigid bronchoscopy (6-8). Endotracheal intubation is particularly important due to the high risk of turning critical airway obstruction into a complete blockage during rapid sequence induction and paralysis. Additionally, airway manipulation during tracheal intubation can lead to inflammation and hemorrhage (9). However, it is possible that the expiratory positive airway pressure in non-invasive ventilation serves to stent an anatomical stenosis, preventing collapse during expiration. This mechanism may also reduce the work of breathing and enhance alveolar ventilation, facilitating the washout of hypercapnia (10). Chen et al. recommended the successful use of non-invasive ventilation for patients with distal and critical airway stenosis, particularly those experiencing hypercapnic respiratory failure. This approach allows for significant time and logistical planning before bronchoscopic intervention (2).

Tracheobronchial involvement of mucormycosis has been previously reported, and bronchoscopy is an important diagnostic method indiagnosis (11,12). Grey granulation tissue obstructing the airways can be directly visualized (3). A systematic review by He et al. documented 60 cases of mucormycosis in the tracheobronchial tree, with only 10 exhibiting tracheal involvement along with bronchial involvement (12). However, isolated tracheal mucormycosis is exceptionally rare (11,13).

Conditions that need evaluation in the differential diagnosis of focal necrotic involvement of the trachea encompass granulomatous diseases (tuberculosis, Wegener's granulomatosis), neoplastic diseases (chondrosarcoma, adenoid cystic carcinoma), and long-term intubation trauma (5). While sputum culture can serve as a preliminary diagnosis method for infectious granulomatous diseases, negative results are typically obtained and the accurate diagnosis is usually achieved through histopathology. In thecase

of mucor infections, biopsies may show tissue invasion with characteristic broadnonseptate hyphae with rightangled branching (3). In the literature, the most common comorbidity in isolated tracheal mucormycosis cases is DM, and the association with H1N1 pneumonia was reported in only one case (13). Damaraju et al. reported that among the 26 cases of isolated airway invasive mucormycosis identified in the literature, 4 cases were associated with COVID-19 infection (14). Amid the ongoing COVID-19 pandemic, we conduted a COVID-19 PCR test on our patient, vielding negative results. Patients with COVID-induced mucormycosis have been reported during the pandemic. In addition to the ongoing risks of mucormycosis, such as uncontrolled DM and immunosuppression, new factors such as excessive use of steroids and antibiotics couped with hypoxia have emerged during the pandemic. Despite both surgical and medical treatments, the mortality rate remains high (15). A multidisciplinary approach is required to prevent mucormycosis in COVID-19 patients, and the risk of mucormycosis should be consistently considered.

Early diagnosis and antifungal treatment are pivotal determinants of outcome, but above all a high degree of suspicion is required. Surgery plays a crucial role in treating the disease. Among antifungal agents, parenteral Amp B is the preferred treatment, but it can reduce the glomerular filtration rate, and hence Liposomal Amp B is recommended in cases with impaired renal function (16). In some cases, Amp B was administered through inhalation or bronchoscopic instillation (14,17,18). The placement of an airway stent and the use of a transbronchial microtube to drip Amp B have been recommended to quickly relieve airway obstruction and extend the duration of drug action, thereby enhancing the antifungal therapeutic effect (17,18). When tracheotomy or bronchoscopy was managed, the presence of necrotic material in the trachea should raise suspicion of mucormycosis, and perioperative frozen examination may be recommended for early diagnosis, especially in diabetic patients with compromised immune systems. Frozen section analysis is an effective method for promptly managing invasive fungal infections, particularly in situations where decisions need to be made during surgery (19).

Extensive endotracheal involvement is more difficult to manage, and resection and anastomosis have also been performed in the past (5). Due to the limited number of tracheal mucormycosis cases, the timing of surgery is a matter of debate. More data isneeded to decide whether surgery should be performed immediately at the time of diagnosis or whether surgical resection would be better after a course of medical treatment.

Conclusion

In the emergency service, situations necessitating emergency airway access due to difficult intubation may be releated with central airway obstruction. When emergency tracheotomy or bronchoscopy is required, it should be kept in mind that an obstructive greymass or necrotic material in the trachea may be a focus of fungal infection requiring rapid treatment. Perioperative frozensection examination may be recommended to accelerate the differential diagnosis of potential invasive fungal infection in immunocompromised diabetic patients. Early management supported with a confirmed frozen section may be life-saving for immunocompromised patients with fungal infections.

- **1.** Milner S, Bennett J. Emergency cricothyrotomy. Journal of Laryngology and Otology 1991; 105: 883–5.
- Win Cheng LT, Sim TB, Kuan WS. Noninvasive ventilation as a temporizing measure in critical fixed central airway obstruction: A cese report. J Emerg Med 2018 May;54(5):615-618. DOI:10.1016/j.jemermed.2017.12.059.
- **3.** Luo LC, Cheng DY, Zhu H, Shu X, Chen W-B. Inflammatory pseudotumoral endotracheal mucormycosis with cartilage damage. Eur Respir Rev 2009 Sep;18(113):186-9. DOI: 10.1183/09059180.00000709.
- **4.** Aquino SL, Shepard JA, Ginns LC, et al. Acquired tracheomalacia: detection by expiratory CT scan. J Comput Assist Tomogr 2001;25:394-9.
- Wolf O, Gil Z, Leider-Trejo L, Khafif A, Biderman P, Fliss DM. Tracheal mucormycosis presented as an intraluminal soft tissue mass. Head Neck 2004 Jun;26(6):541-3. DOI:10.1002/ hed.20055.
- 6. BMJ Best practice. Central airway obstruction. Available at: 2017. http://bestpractice.bmj.com/best-practice/monograph/1069.html. Accessed June 4, 2017.
- **7.** Ernst A, Feller-Kopman D, Becker HD, et al. Central airway obstruction. Am J Respir Crit Care Med 2004;169:1278-97.
- **8.** Theodore PR. Emergent management of malşgnancy-releated acute airway obstruction. Emerg Med Clin North Am 2009;27:231-41.
- **9.** Patel A, Pearce A. Progress in management of the obstructed airway. Anesthesia 2011;66(Suppl 2):93-100.
- **10.** Kinnear W, Watson L, Smith P, et al. Effect of expiratory positive airway pressure on tidal volume during noninvasive ventilation. Chron Respir Dis 2017;14:105-9.
- **11.** Fallahi MJ, Nikandish R, Ziaian B, Shahriarirad R. Near-Complete tracheal obstruction due to mucormycosis: A report of two cases. Clin Case Rep 2022 Aug 24;10(8):e6278. DOI:10.1002/ccr3.6278.
- **12.** He R, Hu C, Tang Y, Yang H, Cao L, Niu R. Report of 12 cases with tracheobronchial mucormycosis and a review. Clin Respir J 2018 Apr;12(4):1651-1660. DOI:10.1111/crj.12724.
- **13.** Mohindra S, Gupta B, Gupta K, Bal A. Tracheal mucormycosis pneumonia: a rare clinical presentation. Respir Care 2014 Nov;59(11):e178-81. DOI:10.4187/respcare.03174.
- **14.** Damaraju V, Agarwal R, Dhooria S, et al. Isolated tracheobronchial mucormycosis: Report of a case and systematic review of literatüre. Mycoses. 2023 Jan;66(1):5-12. DOI:0.1111/myc.13519.

- **15.** Dilek A, Ozaras R, Ozkaya S, Sunbul M, Sen El, Leblebicioglu H. COVID-19-associated mucormycosis: Case report and systematic review. Travel Med Infect Dis 2021 Aug 26;44:102148. DOI:10.1016/j.tmaid.2021.102148.
- **16.** Greenberg RN, Scott LJ, Vaughn HH, Ribes JA. Zygomycosis (mucormycosis): emerging clinical importance and new treatments. Curr Opin Infect Dis 2004;17(6):517-525.
- **17.** Damaraju V, Agarwal R, Prabhakar N, et al. Isolated tracheal mucormycosis in diabetes mellitus and bronchoscopic management. Lung India. 41(3):p 226-227, May–Jun 2024.
- **18.** Chen GJ, Chen XB, Rao WY, et al. Airway necrosis and granulation tissue formation caused by Rhizopus oryzae leading to severe upper airway obstruction: a case report. Front Cell Infect Microbiol. 2024 Mar 4:14:1366472. DOI:10.3389/fcimb.2024.1366472.
- **19.** Papagiannopoulos P, Lin DM, Al-Khudari S, et al. Utility of intraoperative frozen sections in surgical decision making for acute invasive fungal rhinosinusitis. Int Forum Allergy Rhinol 2017;7(5):502-507. DOI:10.1002/alr.21918.

Journal of Emergency Medicine Case Reports

An Interesting Cause of Abdominal Pain in the Emergency Room

Naim Hikmet Kalkan¹, Muhammet Gökhan Turtay¹, Mustafa Çifçi Qifçi Qif

Abstract

In this case report, we aimed to present a case of abdominal pain who presented to the emergency department with complaints of abdominal pain and constipation for 2 days. A 76-year-old woman presented to our emergency department with complaints of abdominal pain and constipation for 2 days. Physical examination revealed a distended abdomen, diffuse tenderness and hypoactive bowel sounds. Abdominal plain abdominal radiography showed approximately 10-12 similarly shaped hyperdense foreign bodies (long dimension measured 15 mm and short dimension measured 5 mm) in the left lower quadrant and left upper quadrant. Abdominal computed tomography showed similarly shaped nodular foreign bodies with dense calcific-metallic characteristic foreign bodies with different locations and contents, one in the stomach and the others in the colon. The next day, black stools were added to her complaints. Endoscopy and colonoscopy performed 3 days later revealed no nodular foreign body with calcific-metallic appearance. The patient was followed up by the gastroenterology department and discharged after her general condition improved. It should be kept in mind that atypical eating disorders may underlie high-density calcific-metallic nodular lesions in the stomach and intestines in patients presenting with abdominal pain.

Keywords: Abdominal pain, constipation, eating disorders, radiology

Introduction

Abdominal pain is a common cause of emergency department admissions. It is challenging and high of readmissions. Careful evaluation and systematic approach towards the cause prevents overlooking potentially serious conditions (1).

Many blood tests and imaging methods, including direct radiography, ultrasound and tomography, are used in diagnosis.

In this case report, we aimed to present an interesting case of abdominal pain who presented to the emergency department with complaints of abdominal pain and constipation for 2 days.

Case Report

A 76-year-old woman presented to our emergency department with complaints of abdominal pain and constipation for 2 days. In her anamnesis, she stated that she had frequent constipation complaints before. Her medical history included asthma, hypertension (HT), anxiety, total abdominal hysterectomy + bilateral salpingooopherectomy

(TAH+BSO), and appendectomy. On physical examination, the patient's general condition was good, oriented, cooperative and vital signs were fever: 36.5°C, pulse rate: 103/min, blood pressure: 110/70mmHg, respiratory rate: 16/min. The abdomen was distended, there was diffuse tenderness and bowel sounds were hypoactive (2-4/ min). Blood tests revealed white blood cell count (WBC) 13,800 /mm3, hemoglobin count (Hgb) 6.9 g/dL, lactate 2.5 mmol/L. She was otherwise normal. Standing plain abdominal radiography (SPAR) revealed approximately 10-12 similarly shaped hyperdense foreign bodies (long dimension measured 15 mm and short dimension measured 5 mm) in the left lower quadrant and left upper quadrant (Figure-1). Subsequently, abdominal computed tomography (CT) performed for differential diagnosis showed nodular foreign bodies with dense calcific-metallic character with different locations and contents, one in the stomach and the others in the colon, measuring 15 mm at the most prominent location, with a density of 2500-3000 HU (Figure-2). General surgery and internal medicine consultations were requested. General surgery did not consider emergency surgery and recommended outpatient clinic. The patient left the hospital voluntarily without waiting for internal medicine consultation.

Corresponding Author: Muhammet Gökhan Turtay e-mail: mgturtay@gmail.com

Received: 24.12.2024 • **Revision:** 30.12.2024 • **Accepted:** 05.01.2025 **DOI:** 10.33706/jemcr.1606684

©Copyright 2020 by Emergency Physicians Association of Turkey -Available online at www.jemcr.com Cite this article as: Kalkan NH, Turtay MG, Çiftçi M, Yıldırım B. An Interesting Cause of Abdominal Pain in the Emergency Room. Journal of Emergency Medicine Case Reports. 2025;16(1): 19-21

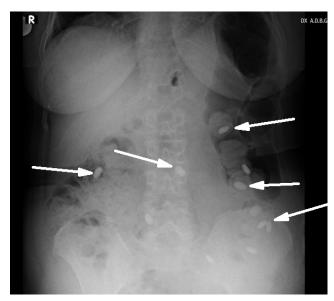


Figure 1. Hyperdense foreign bodies on standing plain abdominal radiography

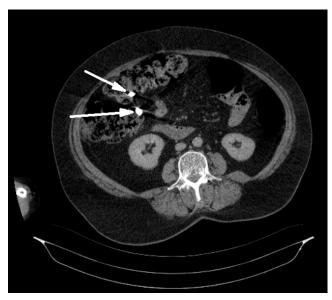


Figure 2. Nodular foreign bodies with dense calcific-metallic character on abdominal computed tomography

The next day, the patient's complaints increased and vomiting and black stools were added to her complaints. The patient's general condition was poor and rectal examination revealed melena. In blood tests, leukocytosis, anemia and lactate level deteriorated further (Hgb: 4.9 g/dL, lactate: 5.8 mmol/L). The patient was consulted to the gastroenterology department considering gastrointestinal system (GIS) bleeding. The patient was hospitalized in the intensive care unit with a prediagnosis of GI bleeding for further investigations and treatment. Endoscopy performed 3 days later revealed no nodular foreign body with calcific-metallic appearance, but a large submucosal mass on the posterior wall of the gastric antrum-corpus junction, approximately 6

cm in size, with 2 white exudate ulcers of 6-7 mm in size. This vision was consistent with gastrointestinal stromal tumor (GIST). The patient showed clinical improvement in the follow-up and was discharged with a recommendation for general surgery outpatient clinic control.

Discussion

Abdominal pain is a common condition that accounts for 5-10% of emergency department admissions. However, 20% to 30% of patients with abdominal pain leave the hospital without a definitive diagnosis (2).

Laboratory and imaging methods are used in the diagnosis and differential diagnosis of diseases underlying abdominal pain (3-6). In our case, we ordered blood tests for abdominal pain. Blood tests revealed leukocytosis, anemia and elevated lactate levels. SPAR and CT scans were then ordered as imaging modalities. SPAR revealed approximately 10-12 similarly shaped hyperdense foreign bodies in the left lower and left upper quadrants. Subsequently, an abdominal CT scan confirmed the presence of foreign bodies in the stomach and colon and excluded other possible causes of abdominal pain.

Afterwards, the patient developed melena and gastroenterology department was consulted considering GIS bleeding. Endoscopy and colonoscopy were performed by the gastroenterology department 3 days later. No nodular foreign bodies with calcific-metallic appearance were detected in endoscopy and a large submucosal lesion compatible with GIST with 2 white exudate ulcers was observed on the posterior wall of the gastric antrum - corpus junction. When the patient's previous examinations were examined, endoscopy and colonoscopy performed 5 months ago due to the patient's anemia showed no metallic appearance and SPAR performed 6 months ago showed no hyperdense image.

We considered atypical eating disorder as the cause of the foreign bodies present in the patient. Atypical eating disorders are eating behaviour disorders that lead to medical, social and psychological problems and affect quality of life negatively (7,8). Epidemiologic risk factors for atypical eating disorders include cultural characteristics, gender, age, mental disorders in the family, family lifestyle, socioeconomic class, personality role, previous psychological disorders, genetics, sexual orientation and occupation (9). Atypical eating disorder are based on a type of self-injurious behaviour involving the ingestion of non-nutritive objects. The result is usually excretion of the ingested substances through feces, but rarely requires surgical intervention (10). Our patient was an elderly woman with anxiety disorder and psychiatric drug use. We thought that the patient had atypical eating disorder and the high-density calcific-metallic nodular lesions on imaging to be excreted through feces.

Conclusion

It should be kept in mind that patients presenting with abdominal pain may have atypical eating disorders underlying the high-density calcific-metallic nodular lesions seen in the stomach and intestines in imaging methods.

- 1. Kacprzyk A, Stefura T, Chłopaś K, Trzeciak K, Załustowicz A, Rubinkiewicz M, et al. Analysis of readmissions to the emergency department among patients presenting with abdominal pain. BMC Emerg Med. 2020;20(1):37.
- **2.** Watkins JW 4th, Lewis ZB. Diagnoses of Exclusion in the Workup of Abdominal Complaints.EmergMedClin North Am. 2021 Nov;39(4):851-863.
- **3.** Lameris W, van Randen A, Dijkgraaf MG, Bossuyt PM, Stoker J, Boermeester MA.Optimization of diagnostic imaging use in patients with acute abdominal pain OPTIMA: Design and rationale. BMC Emerg Med 2007;7:9.
- **4.** Loo JT, Duddalwar V, Chen FK, Tejura T, Lekht I, Gulati M. Abdominal radiograph pearls and pitfalls for the emergency department radiologist: a pictorial review. Abdom Radiol (NY) 2017;42(4):987-1019.

- 5. Scheinfeld MH, Mahadevia S, Stein EG, Freeman K, Rozenblit AM. Can lab data be used to reduce abdominal computed tomography (CT) usage in young adults presenting to the emergency department with nontraumatic abdominal pain?. Emerg Radiol 2010; 17:353–60.
- 6. Hardwick RH, Armstrong CP. Synchronous upper and lower gastrointestinal endoscopy is an effective method of investigating iron-deficiency anaemia. Br J Surg 1997;84:1725-8
- Treasure J, Duarte TA, Schmidt U. Eating disorders. Lancet 2020;395(10227):899-911.
- **8.** Delaney CB, Eddy KT, Hartmann AS, Becker AE, Murray HB, Thomas JJ. Pica and rumination behavior among individuals seeking treatment for eating disorders or obesity. Int J Eat Disord 2015;48(2):238-48.
- **9.** Lam CL, Pan PC, Chan AW, Chan SY, Munro C. Can the Hospital Anxiety and Depression (HAD) Scale be used on Chinese elderly in general practice?. Fam Pract 1995;12(2):149-54.
- **10.** Ariza-Fernández JL, Úbeda-Muñoz M, Redondo-Cerezo E. Endoscopic retrieval of multiple large sharp foreign bodies from the stomach. Testing the limits of endoscopy. Gastroenterol Hepatol 2017;40(2):95-6.

Journal of Emergency Medicine Case Reports

Testicular Infarction: A Rare Complication of Epididymo-Orchitis

¹Department of Urology, Kemer State Hospital, Antalya, Türkiye

²Department of Radiology, Kemer State Hospital, Antalya, Türkiye

Abstract

Epididymo-orchitis is a common urological disorder in clinical practice. Testicular infarction is a rare complication of epididymo-orchitis with a few reports in the literature. A 53-year-old male presented to the Emergency Department complaining of right scrotal pain. The patient was diagnosed with acute epididymitis and treated with ciprofloxacin for 2 weeks without clinical improvement. Subsequently, he was admitted to the Urology Department. A color Doppler ultrasonography revealed an enlarged right epididymis with increased vascularity. No significant color flow signal was observed in the right testis except for a small parenchymal area of the posterior part. The patient underwent scrotal exploration, and the ischemic changes of the testis were observed. Therefore, a right orchiectomy was performed. We present here a case of testicular infarction secondary to epididymo-orchitis. If clinical findings do not improve despite appropriate conservative treatment, patients should be under close follow-up for reassessment with ultrasonography, and more aggressive conservative therapy could be prescribed to prevent complications that may lead to testicular loss.

Keywords: Complication, epididymitis, epididymo-orchitis, testicular infarction

Introduction

Epididymo-orchitis, which is an inflammation of the epididymis and testis, is a common condition in clinical practice. The symptoms include scrotal pain accompanied by swelling, fever, and lower urinary tract symptoms. The diagnosis is made based on clinical symptoms, physical examination, and Doppler (capital D) ultrasonography. This condition can be managed with anti-microbial and anti-inflammatory treatment. However, testicular infarctions have rarely been reported as a complication of epididymo-orchitis in literature (1). Here in, we discuss a case of epididymo-orchitis that progressed to abscess formation with testicular infarction.

Case Report

A 53-year-old male presented to the Emergency Department complaining of right scrotal pain. The patient was diagnosed with acute epididymitis and treated with ciprofloxacinfor 2 weeks without clinical improvement. He subsequently developed right scrotal swelling, dysuria, and fever and was admitted to the Urology Department. He had a history of hypertension and

previously treated left-sided epididymitis but denied having a history of trauma, drug use, urethral discharge, or prior sexually transmitted diseases. His vital signs were normal. The physical examination revealed an erythematous right hemiscrotum and a swollen right testicle with severe tenderness. Laboratory tests showed leukocytosis (21.7x103 /mm3) with neutrophil predominance (84.9%), elevated C-reactive protein (119 mg/L), and elevated creatinine level (1.48 mg/dL). A color doppler ultrasonography (CDU) showed an enlarged right epididymis with increased vascularity. No significant color flow signal was observed in the right testis except for a small parenchymal area of the posterior part (Figure-1). The patient underwent emergent scrotal exploration. There were severe adhesions of the scrotal wall. No evidence of torsion of the spermatic cord was identified. Ischemic changes and discoloration of the testis were observed, and the discoloration persisted (Figure-2). A right orchiectomy was performed. Histopathological findings were hemorrhagic infarction, abscess formation, and infiltration by inflammatory cells. After surgery, the patient's condition improved significantly, and the laboratory results returned to normal dramatically. He was discharged home on post-operative day 3 with oral antibiotics and anti-inflammatory drugs.

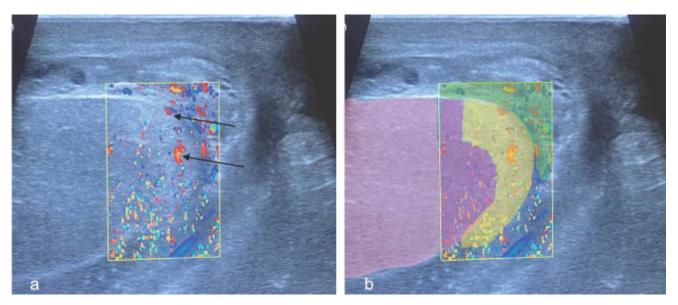


Figure 1. Testicular ultrasound images of the patient with segmental infarction. (a) The color Doppler (with capital D) box is placed to cover the necrotic and non-necrotic parts of the testicle and epididymis. (b) The epididymis is seen in the Doppler box withing the green area; increased blood flow in this area suggests epididymitis. The dark and pink sections are necrotized testicle; there is no Doppler flow in the necrotized testis (the dark pink part), only noise. The light pink area is the testicle in B-mode; hypoechoic and granular appearance is indicative of infarction. The yellow area represents the normal testicle with preserved echogenicity and color doppler flow (arrows).



Figure 2. Intraoperative view of the testicle

Discussion

Epididymo-orchitis is the most common cause of acute scrotal pain in adults (2). The main treatment strategy for epididymo-orchitis includes antibiotics and analgesics. Testicular infarction secondary to epididymo-orchitis is extremely rare, and a few reports have been published in the urologic literature (1,3). The pathogenesis of abscess formation and testicular infarction remains unknown. The true incidence of this severe complication is still obscure and may be underestimated. Several mechanisms have been proposed to contribute to testicular infarction due to epididymo-orchitis. These are compression of the testicular artery by the inflammatory process of the spermatic cord, endothelial dysfunction caused by bacterial toxins, and

increased venous resistance leading to thrombosis (4). Although the exact mechanism of testicular infarction due to epididymo-orchitis is not well understood, early diagnosis and intervention are crucial to prevent testicular loss.

Color Doppler ultrasonography is essential along with clinical findings and physical examination to confirm the diagnosis of testicular infarction. The differential diagnosis of acute scrotal pain includes epididymitis, testicular torsion, torsion of appendix testis, testicular malignancy, and urolithiasis. CDU is the primary modality used to assess the blood flow of the testis and epididymis in the differential diagnosis (5). Alternatively, contrast-enhanced ultrasound provides visualization of testis parenchyma and vascular enhancement with high sensitivity and can be performed quickly at the patient's bedside (6). In this case, the patient's first CDU showed normal testicular blood flow and increased epididymal blood flow. Despite ciprofloxacin and analgesics treatment, an increase in epididymal blood flow was observed in the CDU performed after 2 weeks, accompanied by increased scrotal pain and swelling in the scrotum, but vascularization was absent in most of the testicle. If clinical findings do not improve despite appropriate conservative treatment, patients should be under follow-up for reassessment with ultrasonography to prevent complications that may lead to testicular loss.

Acute epididymitis is a common diagnosis in men with scrotal pain. Due to anatomical proximity, the inflammation extends from the epididymis and can easily spread to the testis. Epididymo-orchitis can be seen in all age groups. The most common pathogens responsible for epididymo-orchitis are Neisseria gonorrhoeae and Chlamydia trachomatis in men under 35 years of age, while Escherichia

coli and other gram-negative bacteria in older men (7). A urine culture should be obtained to determine proper treatment. Nevertheless, urine culture is often unable to reliably detect the pathogenic microorganism (8). Empiric therapy should be prescribed based on age and sexual activity. Empirical antibiotics include ceftriaxone plus doxycycline for suspected sexually transmitted pathogens and fluoroquinolones for suspected urinary tract pathogens (7). In our case, the patient had a history of epididymitis treated with fluoroquinolone without any complications a year ago. After that, the patient was diagnosed with acute epididymitis again and fluoroquinolone treatment for 2 weeks did not yield positive results. Considering the increasing incidence of fluoroquinolone resistance and the possible coexistence of atypical pathogens, empirical treatment can be expanded in patients with recurrent urinary tract infections.

Conclusion

Epididymo-orchitis leading to testicular infarction is extremely rare. Situations predisposing to testicular infarction secondary to epididymo-orchitis may include a history of scrotal trauma, instrumentation, anatomical variations, recurrent urinary tract infections, and immunosuppressive conditions. Patients with these risk factors should be under close follow-up and more aggressive conservative therapy could be prescribed. Additionally, repeated CDU of the testes should be performed. Clinicians must be aware of the serious consequences such as the progression of the infection and testicular infarction in patients with severe nonresolving epididymitis despite appropriate conservative treatment.

Ethics Approval and Consent to Participate:

This case report was presented in accordance with the Declaration of Helsinki. Written informed consent was obtained from the patient involved in this study.

Acknowledgment

None

Funding

This research received no external funding.

Conflict of Interest

The authors declare no conflict of interest.

- 1. Fehily SR, Trubiano JA, McLean C,Teoh BW, Grummet JP, Cherry CL et al. Testicular loss following bacterial epididymoorchitis: Case report and literatu rereview. Can Urol Assoc J 2015;9(3-4):E148-E 151.
- Marcozzi D, Suner S. The non traumatic, acute scrotum. Emerg Med Clin North Am 2001;19(3):547-568.
- **3.** Devlies W, Seghers M, Dilen K. Case report on secondary testicular necrosis due to fulminant epididymitis: ultrasonographic evaluation and diagnosis. BMC Urol 2020;20(1):115.
- **4.** Gandhi J, Dagur G, Sheynkin YR, Smith NL, Khan SA. Testicular compartment syndrome: an overview of pathophysiology, etiology, evaluation, and management. Transl Androl Urol 2016;5(6):927-934.
- **5.** Mc Adams CR, Del Gaizo AJ. The utility of scrotal ultrasonography in the emergent setting: beyond epididymitis versus torsion. Emerg Radiol 2018;25(4):341-348.
- 6. Lung PF, Jaffer OS, Sellars ME, Sriprasad S, Kooiman GG, Sidhu PS. Contrast-enhanced ultrasound in the evaluation of focal testicular complications secondary to epididymitis. AJR Am J Roentgenol 2012;199(3):W345-W 354.
- **7.** Trojian TH, Lishnak TS, Heiman D. Epididymitis and orchitis: an overview. Am Fam Physician 2009;79(7):583-587.
- Kim SD, Kim SW, Yoon BI, Ha US, Kim SW, Cho YH et al. The Relationship between Clinical Symptoms and Urine Culture in Adult Patients with Acute Epididymitis. World J Mens Health 2013;31(1):53-57.

Journal of Emergency Medicine Case Reports

Current Approaches in the Treatment and Reconstruction of Frostbite Injuries: A Case Report

ihtişam Zafer Cengiz¹, in Fatma Çakmak²

¹Department of Plastic Reconstructive and Aesthetic Surgery, Atlas University Hospital, İstanbul, Türkiye

²Department of Emergency Medicine Atlas University Hospital, İstanbul, Türkiye

Abstract

Frostbite injuries, though uncommon in temperate climates like Turkey, are prevalent in high-altitude, cold regions such as Eastern Anatolia. These injuries are particularly observed in vulnerable populations exposed to prolonged cold, such as refugees. Pathophysiologically, tissue damage occurs due to ice crystal formation—either intracellularly, leading to direct cell death, or extracellularly, causing osmotic dehydration. Early intervention with rewarming protocols is crucial to prevent severe complications, including necrosis and amputations. A 24-year-old male refugee walked on snowy roads for a week, resulting in frostbite injuries to his hands and feet. On admission, he presented with bullae formation, prolonged capillary refill, and weak circulation in the extremities. Initial management included anti-inflammatory (ibuprofen), anticoagulant (heparin), and peripheral vasodilation (pentoxifylline) therapies. After one week, demarcation lines developed, necessitating amputation of affected fingers. A reverse sural artery flap was used to reconstruct the deep tissue defect in the left calcaneal region, while a full-thickness skin graft was applied to the right calcaneal defect in a secondary procedure. Frostbite injuries require prompt diagnosis and intervention to minimize tissue loss and complications. In delayed cases, amputations and reconstructive surgeries become inevitable. A multidisciplinary approach, including medical and surgical management, is critical for successful outcomes. Preventive measures, education of at-risk populations, and timely access to healthcare are essential in reducing frostbite-related morbidity.

Keywords: Frostbite injury, revers flow, sural flap

Introduction

Although Turkey is generally located in a warm climate zone, frostbite injuries can frequently be encountered, particularly in regions with high altitudes and harsh cold weather conditions, such as the Eastern Anatolia Region. These cold-induced injuries are more commonly observed in certain at-risk populations. These risk factors can be remembered using an acronym often referred to in English as the "I's of Frostbite": Intoxicated (individuals under the influence of alcohol or drugs), Incompetent (those with impaired mental health), Infirm (elderly or debilitated individuals), Insensate (people with sensory deficits), Inducted (those in compulsory military service or war conditions), Inexperienced (individuals unfamiliar with cold climates), and Indigent (homeless or economically disadvantaged individuals) (1).

The pathophysiology of frostbite is evaluated through two distinct mechanisms: rapid and slow freezing. In cases of rapid freezing, intracellular ice crystals form, which cause direct cellular damage and result in cell death (2). In slow freezing, ice crystals form extracellularly, leading to osmotic fluid shifts and cellular dehydration. This crystallization process typically begins at approximately -2.2°C. Clinically,

bullae formation in affected areas is observed within 6 to 24 hours following exposure (3).

The complications of frostbite vary depending on the duration and severity of exposure. If not diagnosed and treated promptly, it can result in severe and irreversible damage. While cold-induced tissue injuries initially present as superficial, they can progress to involve deeper tissues, leading to necrosis, the need for amputation, and long-term functional deficits. Early diagnosis plays a critical role in the management of frostbite injuries. Recognizing symptoms such as pallor, hardness, loss of pain sensation, and bullae formation necessitates rapid intervention (4).

In the treatment of frostbite, the primary goal is to maintain the patient's systemic temperature while restoring circulation to the affected areas. This is typically achieved through immersion in water at 40-42°C, if possible. However, care must be taken during central rewarming, as peripheral vasodilation can cause cooled blood to return to the core, thereby increasing the risk of systemic hypothermia. Additionally, massaging or rubbing the affected extremities must be avoided, as intracellular ice crystals can cause irreversible tissue damage (5).

Early diagnosis and appropriate intervention are key to minimizing tissue loss and reducing the risk of complications in frostbite injuries.

Corresponding Author: İhtişam Zafer Cengiz e-mail: drizafercengiz@hotmail.com

Received: 18.12.2024 • **Revision:** 30.12.2024 • **Accepted:** 09.01.2025

DOI: 10.33706/jemcr.1603900

©Copyright 2020 by Emergency Physicians Association of Turkey -Available online at www.jemcr.com Cite this article as: Cengiz İZ, Çakmak F. Current Approaches in the Treatment and Reconstruction of Frostbite Injuries: A Case Report. Journal of Emergency Medicine Case Reports. 2025;16(1): 25-27

A 24-year-old male patient, a refugee from Pakistan, crossed the border into Turkey on foot. According to the patient's account, he had to walk on snowy roads for approximately one week. Upon arrival, the patient was in good general condition, conscious, oriented, and cooperative. Initial warming protocols had been applied at an external center. In the emergency department, hand finger circulation was assessed as weak, and capillary refill time was prolonged. Bullae were present on the dorsal aspect of the feet and calcaneal regions. The patient reported pain in the extremities. Ibuprofen and ampicillin treatments were initiated, and the extremities were placed in static splints. Tetanus prophylaxis was administered. Heparin therapy was started to prevent microthrombosis, and pentoxifylline treatment was initiated to improve blood flow to the extremities through peripheral vasodilation. The patient was admitted to the burn treatment center.

After one week of follow-up, demarcation lines developed on the fingers. Amputation was performed at the mid-phalanx level for the 3rd, 4th, and 5th fingers of the right hand and at the distal phalanx level for the 3rd and 4th fingers of the left hand. Following debridement, a deep tissue defect was observed in the left calcaneal region, while a superficial deep tissue defect was identified in the right calcaneal region (Figure-1). Due to the deeper necrosis in the left calcaneal region, reconstruction was performed with a reverse sural artery flap by the plastic surgery team (Figure-2). For the right calcaneal region, a full-thickness skin graft harvested from the surplus tissue in the pedicle of the sural flap was applied during the second session (Figures-3,4).

Discussion

Frostbite injuries can lead to severe complications, particularly in individuals at risk due to prolonged exposure to cold temperatures (6). As observed in our case, refugees are especially vulnerable to frostbite injuries due to inadequate protection and prolonged exposure to freezing conditions. Early initiation of rewarming protocols and appropriate medical treatment in the early stages of frostbite is crucial to limiting the progression of complications (5). However, in delayed cases, tissue necrosis may advance, necessitating amputations and reconstructive surgical interventions.

From a pathophysiological perspective, tissue damage in frostbite occurs primarily due to the formation of intracellular or extracellular ice crystals (3). In our case, the presence of bullae and circulatory disturbances in the hands and feet were consistent with the classical clinical findings of frostbite. The administration of ibuprofen aimed to reduce tissue damage through its anti-inflammatory effects, while therapies such as heparin and pentoxifylline were effective in preventing microthrombi and improving peripheral blood flow.



Figure 1. A deep tissue defect was observed in the left calcaneal region



Figure 2. The left calcaneal region, reconstruction was performed with a reverse sural artery flap





Figure 3,4. The right calcaneal region, a full-thickness skin graft harvested from the surplus tissue in the pedicle of the sural flap was applied

From a reconstructive surgical standpoint, soft tissue repair is a critical step in managing tissue defects caused by frostbite. In this case, the reverse sural artery flap applied to the left calcaneal region is a commonly used method for extensive and deep tissue defects, and it was successfully implemented. On the right side, repair was achieved using a full-thickness skin graft harvested from the pedicle portion

of the sural flap in a secondary session, aiming for both functional and aesthetic improvement (7). This highlights the importance of a multidisciplinary approach in managing tissue losses effectively.

Conclusion

Frostbite injuries are a significant health concern, particularly for individuals exposed to cold weather conditions for prolonged periods, and they can lead to severe complications. Early diagnosis and appropriate treatment play a critical role in minimizing tissue damage and the need for amputations. As demonstrated in our case, delayed intervention may render amputation and reconstructive surgery inevitable. With a multidisciplinary approach, successful outcomes can be achieved in the treatment of tissue defects caused by frostbite. To prevent such injuries, it is of utmost importance to educate at-risk individuals, implement protective measures against cold exposure, and ensure timely presentation to healthcare centers.

Ethic Approval

This case report has been written in an anonymous manner; therefore, confidential and detailed data about the patient have been removed. Editors and reviewers may have access to and review these detailed data, which are fully supported by the editors and reviewers.

- Xu X, Tikuisis P, Gonzalez RR. Thermoregulatory modeling of cold survival. Mil Med. 2003;168(7):556-565.
- Wilson PW. Mechanisms of cold injury. J Appl Physiol. 2000;89(3):1456-1464.
- **3.** Imray CHE, Grieve A, Dhillon S. Cold damage to the extremities: Frostbite and non-freezing cold injuries. Postgrad Med J. 2009;85(1007):481-488.
- **4.** Mills WJ, Whaley R, Block JE. Frostbite: Cold-induced tissue injury and its treatment. J Trauma Acute Care Surg. 2004;56(6):1334-1340.
- **5.** McIntosh SE, Opacic M, Freer L, Leemon D. Wilderness Medical Society practice guidelines for the prevention and treatment of frostbite. Wilderness \& Environ Med. 2011;22(2):156-166.
- **6.** Manganello JA, Lyons BA. Frostbite injuries: Risk factors, clinical features, and management. J Emerg Med. 2018;45(4):567-573.
- Sulaiman AR, Adhikari A, Balasubramanian D. Reverse sural artery flap in distal extremity reconstruction: A clinical review. Plast Reconstr Surg Glob Open. 2020;8(3):e2784.

Journal of Emergency Medicine Case Reports

Abdominal Aortic Aneurysm Presenting with Acute Pancreatitis: A Rare Clinical Manifestation

Mevlana Gül¹, Yunus Emre Ek¹

¹Ataturk University, Faculty of Medicine, Emergency Department, Erzurum, Türkiye

Abstract

Acute pancreatitis is an inflammatory condition of the pancreas associated with elevated pancreatic enzymes, mainly caused by gallstones and alcohol abuse. In this article, we report a rare case of an 82-year-old woman with abdominal aortic aneurysm (AAA) and acute pancreatitis. The patient presented with typical acute pancreatitis symptoms and AAA was diagnosed by further imaging. This study underlines the mechanical and inflammatory interactions between AAA and acute pancreatitis and highlights the potential for concurrent presentation, including aneurysm rupture or recurrent pancreatitis. It emphasises the importance of advanced imaging and multidisciplinary management in handling such cases. Further research is recommended to uncover pathophysiological interactions and optimise clinical outcomes for such comorbid conditions.

Keywords: Abdominal aortic aneurysms, acute abdomen, pancreatitis

Introduction

Acute pancreatitis (AP) is an inflammatory condition of the pancreas, characterised by abdominal pain and elevated levels of pancreatic enzymes in the blood (1,2). A comprehensive understanding of the etiology of acute pancreatitis is imperative for the effective management and prevention of the disease. The most prevalent aetiological factors contributing to acute pancreatitis are gallstones and alcohol misuse, which collectively account for approximately 75% of cases. Hypertriglyceridaemia (HTG) and endoscopic retrograde cholangio pancreatography (ERCP) have also been identified as significant aetiological factors (3,4). Genetic mutations, including those in the PRSS1 and SPINK1 genes, have been identified as risk factors for the development of pancreatitis. Autoimmune pancreatitis, although rare, is another recognised cause. Certain medications, including azathioprine and valproicacid, have been implicated in causing pancreatitis, though they account for less than 5% of cases (5). The etiology of pancreatitis encompasses a wide spectrum of factors, including hypercalcemia, infections, and abdominal trauma. Acute pancreatitis is characterised by the failure

of protective mechanisms against trypsinogen activation, resulting in the activation of enzymes within the pancreas, auto-digestion, and subsequent inflammation. This process can lead to both local and systemic inflammatory responses, which may ultimately result in multiple organ dysfunction syndrome (MODS) (6,7).

Case Report

An 82-year-old female patient presented to the emergency department with abdominal pain and nausea. The patient had been living with hypertension for 15 years, hypothyroidism for 15 years, and cardiomegaly and atrial fibrillation for 2 years. A physical examination of the patient revealed that their general condition was satisfactory. Their level of consciousness was clear, and they were oriented and coherent. Vital signs revealed a blood pressure of 137/69 mmHg, a pulse rate of 87/min, and finger tip oxygen saturation of 91%, all of which were stable. Abdominal examination revealed tenderness in the upper quadrants of the abdomen. There was no history of previous abdominal operation and no scar. Biochemical tests showed amylase 1152 U/L, lipase 2215 U/L, total bilirubin 0.61, and liver function tests

and other parameters were within the normal range. White blood cell count was 11,000/μL, haemoglobin was 12.7 and platelet count was 184,000/µL. The gallbladder exhibited signs of distension, with a diameter measuring 35 mm, and the presence of freefluid was observed in the pericholecystic region. Millimetre-sized hyperdense appearances were detected within the gallbladder lumen (possibly indicating calculi). Intra- and extrahepatic bile ducts are normal (Figure-1). The pancreas manifested normal dimensions and anatomical location, with a smooth internal structure and lobulated external contours. Contamination was observed in the fatty planes of the peripancreatic area (Figure-2). Contrasted computed tomography imaging revealed that the dilatation of the aneurysm measured up to 76 mm at its widest point at the level of the terminal abdominal aorta (Figur- 3). The patient was seen in the gastroenterology, general surgery and cardiovascular surgery (CVS) clinics. Urgent surgical intervention was considered and the patient was admitted to the CVS clinic for aneurysm surgery. The postoperative image, taken one month after the operation, is illustrated in Figure-4.

Discussion

Abdominal aortic aneurysm (AAA) is a serious vascular condition characterised by the dilation of the abdominal aorta. In the event of rupture, this condition can lead to life-threatening complications. It is most prevalent in older adults, particularly men over the age of 65, and is associated with high mortality rates upon rupture. It is important to note that AAAs are frequently asymptomatic and are often discovered incidentally through imaging studies such as ultrasound, CT, or MRI (8). Surgical repair, either through open surgery or endovascular aortic repair, is the primary treatment for large or symptomatic aneurysms. Elective



Figure 1. Normal intra-extrahepatic bile ducts



Figure 2. Contamination in the fatty planes of the peripancreatic area



Figure 3. The aneurysm at the level of the terminal abdominal aorta

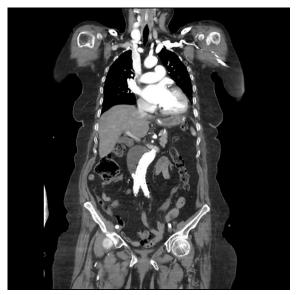


Figure 4. The postoperative image, one month after the operation

repair is generally considered to be the preferred option due to its proven track record of superior outcomes when compared with emergency repair. However, it should be noted that both methods carry an inherent risk of morbidity and mortality (9). A natomically and medically speaking, an AAA and pancreatitis are two distinct pathologies that can result in intricate clinical scenarios. AAA is characterised by the dilatation of the abdominal aorta, and if it were to rupture, would result in a potentially fatal outcome. Conversely, pancreatitis is an inflammation of the pancreas that can range in severity from mild to severe. The intersection of these conditions gives rise to distinctive challenges in terms of diagnosis and management. The co-occurrence of AAA and pancreatitis is an uncommon occurrence, but one that can present significant clinical challenges. A review of the literature reveals a small number of documented cases where these conditions coexist, often complicating the clinical picture and management strategies (10-12). The presence of pancreatitis in patients with AAA has been demonstrated to increase the risk of complications and mortality. For instance, acute pancreatitis (AP) has been associated with increased mortality in cases of ruptured AAA. As is evident in the present case, the presence of AAA has been demonstrated to exertpressure on the pancreatic duct, thereby inducing the onset of pancreatitis. This mechanical compression has been shown to result in recurrent episodes of acute pancreatitis, as documented in case reports (11,13). The inflammatory processes involved in pancreatitis have the potential to exacerbate the weakening of the aortic wall, thereby leading to aneurysm rupture. Elevated pancreatic enzymes, such as elastase, have been shown to degrade the aortic wall thereby contributing to aneurysm formation or rupture (12,14). The diagnosis of pancreatitis in the context of AAA can be challenging due to the presence of overlapping symptoms and the potential for atypical presentations. Consequently, the employment of advanced imaging techniques in conjunction with a meticulous clinical evaluation is imperative for an accurate diagnosis. Further research is required to enhance our understanding of the pathophysiological interactions between aneurysm of the abdominal aorta (AAA) and pancreatitis. This includes the exploration of the role of inflammatory mediators and the impact of surgical interventions on these conditions.

Conclusion

Acute pancreatitis is a multifaceted condition involving numerous aetiological factors. Gallstones and alcohol use are the most common aetiological factors, but other important contributing and known factors include hypertriglyceridemia, ERCP, genetic predispositions and drugs. It is likely that vascular factors may also play a role among these causes and a comprehensive understanding of the underlying pathophysiological mechanisms is imperative for the development of improved management strategies and optimisation of patient outcomes.

- 1. Lankisch, P.G., M. Apte, and P.A. Banks, Acute pancreatitis. The Lancet, 2015. 386(9988): p. 85-96.
- **2.** Williams, A., et al., Comprehensive Review Of The Etiologic Causes Of Acute Pancreatitis.
- **3.** Bálint, E.R., et al., Assessment of the course of acute pancreatitis in the light of aetiology: a systematic review and meta-analysis. Scientific Reports, 2020. 10(1): p. 17936.
- **4.** Walkowska, J., et al., The pancreas and known factors of acute pancreatitis. Journal of clinical medicine, 2022. 11(19): p. 5565.
- **5.** Forsmark Ch, E., S.S. Vege, and C.M. Wilcox, Acute Pancreatitis. N Engl J Med, 2017. 376(6): p. 598-9.
- **6.** Wang, G.-J., et al., Acute pancreatitis: etiology and common pathogenesis. World journal of gastroenterology: WJG, 2009. 15(12): p. 1427.
- **7.** Bhatia, M., et al., Pathophysiology of acute pancreatitis. Pancreatology, 2005. 5(2-3): p. 132-144.
- **8.** Chen, J., L. Hu, and Z. Liu, Medical treatments for abdominal aortic aneurysm: an overview of clinical trials. Expert Opinion on Investigational Drugs, 2024. 33(9): p. 979-992.
- **9.** Lin, J., et al., Status of diagnosis and therapy of abdominal aortic aneurysms. Frontiers in cardiovascular medicine, 2023. 10: p. 1199804.
- **10.** Veraldi, G., et al., Treatment of abdominal aortic aneurysms associated with pancreatic tumors: personal experience and review of the literature (1967-2006). International angiology, 2008. 27(6): p. 539.
- **11.** Chen, C.-Y., et al., Abdominal aortic aneurysm compression is probably responsible for the recurrent episodes of acute pancreatitis: case report. Hepato-gastroenterology, 1999. 46(28): p. 2625-2627.
- **12.** Wachal, K., et al., Acute pancreatitis complicated by rupture of abdominal aortic aneurysm. Gastroenterology Review/ Przegląd Gastroenterologiczny, 2016. 11(2): p. 136-138.
- **13.** Lieder, H.R., et al., Acute occlusion of the ductus pancreaticus due to abdominal aortic aneurysm: uncommon cause of silent sever acute pancreatitis—a case report and review of the litterature. Case Reports in Internal Medicine, 2016. 3(1).
- **14.** Drissi, M., et al., Severe acute pancreatitis after surgical treatment of a ruptured abdominal aortic aneurysm. Annals of Vascular Surgery, 2009. 23(6): p. 785. e5-785. e7.

Journal of Emergency Medicine Case Reports

Lead Ingestion in a Child: A Case Report

¹Department of Emergency Medicine, Yalova University, Yalova, Türkiye

²Department of Emergency Medicine, Mustafa Kemal Paşa State Hospital, Bursa, Türkiye

Abstract

Lead is a heavy metal capable of causing significant toxic effects in children during acute exposure. Accidental ingestion of lead-containing objects is a common cause of acute poisoning in the pediatric population. Acute lead exposure can manifest with a broad clinical spectrum, ranging from neurological and gastrointestinal symptoms to life-threatening complications. This report presents the case of a 6-year-old boy who accidentally ingested a lead object. The patient presented to the emergency department approximately 20 minutes after the incident. His vital signs were stable, physical examination findings were unremarkable, and laboratory results were normal. An upright abdominal X-ray revealed an oval-shaped radiopaque object located in the duodenum. On the fourth day, the blood lead level was measured as 14 µg/dL, and no symptoms were observed. In conclusion, the rapid passage of the lead object through the gastrointestinal tract may have played a role in limiting toxicity. This case contributes to the literature regarding the management and clinical course of acute lead poisoning in children.

Keywords: Acute lead, childhood lead toxication, lead ingestion poisoning

Introduction

Lead is a toxic heavy metal with rapid effects during acute exposure, posing serious health risks, particularly in children. Accidental ingestion of lead-containing objects constitutes a significant proportion of acute poisoning cases in the pediatric population. Lead is readily absorbed through the gastrointestinal tract, entering systemic circulation within a short period and potentially causing systemic toxicity. Acute lead poisoning can present with a diverse clinical spectrum, including neurological symptoms, gastrointestinal distress, hematological abnormalities, and life-threatening complications. Early diagnosis and intervention are critical to mitigating potential fatal outcomes (1,2).

The common sources of acute lead exposure in children include ingestion of toys, batteries, bullets, or other objects containing lead. Such incidents are more frequent among younger children due to their natural curiosity. The movement of lead objects through the gastrointestinal tract is a critical determinant of the severity of toxic effects. The duration of exposure and the time the object remains in the gastrointestinal system are key factors influencing toxicity (3). While objects that pass rapidly through the stomach may have limited toxic effects, those that remain for prolonged periods are associated with higher absorption rates and increased toxicity risk.

In this context, the management of acute lead exposure requires close monitoring of clinical symptoms and, when necessary, planning for endoscopic or surgical intervention within a multidisciplinary approach. (4). In this report, we aim to contribute to the literature by presenting the case of a 6-year-old boy who accidentally ingested a lead object.

Case Report

A 6-year-old boy was brought to the emergency department of a secondary care hospital with a complaint of accidental ingestion of a lead object approximately 20 minutes prior. The patient had no significant medical history, regular medication use, or history of previous surgeries. At presentation, his vital signs were as follows: blood pressure 100/70 mmHg, heart rate 70 bpm, body temperature 36 °C, and oxygen saturation 99%. Physical examination findings were unremarkable. Laboratory tests, including complete blood count, liver and renal function tests, and INR, were within normal limits.

An upright abdominal X-ray (Figure-1) revealed an ovalshaped radiopaque object located in the duodenal region. The patientdid not receive activated charcoal treatment. He was referred to a tertiary care center for further evaluation of potential toxic effects and, if deemed necessary, endoscopic removal of the object. At the tertiary center,

Corresponding Author: Mustafa Ümit Can Dölek
e-mail: umitcandolek@gmail.com
Received: 26.11.2024 • Revision: 30.01.2025 • Accepted: 05.02.2025
DOI: 10.33706/jemcr.1591635
©Copyright 2020 by Emergency Physicians Association of Turkey Available online at www.jemcr.com

Cite this article as: Dölek MÜC, Karanfil A, Altındaş AG. Lead Ingestion in a Child: A Case Report. Journal of Emergency Medicine Case Reports. 2025;16(1): 31-33



Figure 1. Lead X-ray Image.

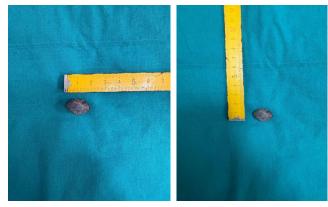


Figure 2. Extracted Lead Material

no endoscopic procedure was performed, and the patient was monitored without any intervention. At 32 hours post-exposure, the object was reported to have passed through the gastrointestinal tract and was excreted in the stool (Figure-2). The retrieved lead object weighed 19 grams, with dimensions of 17 mm, 12 mm, and 6 mm.

On the fourth day, follow-up assessments revealed normal complete blood count, liver and renal function tests, and electrolyte levels. Blood lead levels, measured using the AAS/ICP-MS method, were found to be 14.1 $\mu g/dL$. During this period, the patient remained asymptomatic, and no abnormalities were detected on diagnostic evaluations. The patient was scheduled for follow-up at the hematology outpatient clinic to monitor for potential long-term effects.

Discussion

Lead poisoning has been recognized as an occupational disease since ancient times. Hippocrates first documented this condition in a lead worker presenting with abdominal pain (5). Today, lead exposure occurs through diverse routes, including occupational exposure, accidental ingestion, or environmental contamination. Lead exposure may occur via inhalation, dermal contact, or gastrointestinal absorption.

In the presented case, exposure occurred via accidental ingestion, a common scenario also reported in cases involving indoor shooting ranges, leaded wine pitchers, jewelry manufacturing, and consumption of contaminated food o rmedications (6-9).

While the diagnosis in this case was straight forward, the underlying etiology in patients presenting with acute abdominal symptoms can be challenging to determine (10). In our case, the lead object was promptly identified. Despite the absence of symptoms, acute lead exposure has been associated with a wide range of clinical manifestations, including acute abdomen, anemia, encephalopathy, brain herniation, and fatal outcomes (11-13). The lack of symptoms in this case may be explained by the rapid gastric emptying of the object, which minimized the time it remained in the gastrointestinal system.

A study by Offor et al. in 2017 demonstrated that activated charcoal reduced liver enzymes, oxidative stres markers, and pro-inflammatory cytokines in male albino rats exposed to lead acetate, suggesting protective effects on liver and kidney tissues (14). However, human studies indicate that activated charcoal does not bind to lead, and its use in acute lead poisoning is not recommended (15). Furthermore, it is well-documented that children's gastrointestinal mucosa is more sensitive to lead absorption, with higher absorption rates compared to adults. (11)

In our case, the rapid transit of the ingested lead object through the gastrointestinal tract and it sexcretion in stool likely played a critical role in preventing toxic effects. Nevertheless, the measured blood lead level of 14 μ g/dL is note worthy despite the absence of acute symptoms. Similarly, in the case reported by Gopinath et al., no symptoms were observed in a patient with a blood lead level of 14 μ g/dL. Although asymptomatic in the acute phase, this level may pose long-term risks for cognitive and neurological development.

Conclusion

The pediatric population is particularly vulnerable to lead poisoning. In this case, despite the absence of activated charcoal administration, the blood lead level of $14~\mu g/dL$ did not result in acute symptoms. This case highlights the importance of prompt recognition and management of lead exposure in children, emphasizing the need for long-term monitoring for potential cognitive and neurological effects.

- **1.** Ahamed M, Siddiqui MohdKJ. Environmental lead toxicity and nutritional factors. Clinical Nutrition. 2007;26(4):400-408. doi:10.1016/j.clnu.2007.03.010
- 2. Needleman H. Lead Poisoning. Annu Rev Med. 2004;55(1):209-222. doi:10.1146/annurev.med.55.091902.103653

- Shannon M, Graef JW. Lead Intoxication in Children with Pervasive Developmental Disorders. Journal of Toxicology: Clinical Toxicology. 1996;34(2):177-181. doi:10.3109/15563659609013767
- **4.** Jacobs DE, Clickner RP, Zhou JY, et al. The prevalence of lead-based paint hazards in U.S. housing. Environ Health Perspect. 2002;110(10). doi:10.1289/ehp.021100599
- **5.** Browder AA, Joselow MM, Louria DB. The problem of lead poisoning. Medicine (Baltimore). 1973;52(2):121-139. doi:10.1097/00005792-197303000-00002
- 6. Gordon JN, Taylor A, Bennett PN. Lead poisoning: case studies. Brit J Clinical Pharma. 2002;53(5):451-458. doi:10.1046/j.1365-2125.2002.01580.x
- 7. Barber T, Jacyna M. Acute lead intoxication from medications purchased online presenting with recurrent abdominal pain and encephalopathy. J R Soc Med. 2011;104(3):120-123. doi:10.1258/jrsm.2010.100334
- **8.** Jouglard J, de Haro L, Arditti J, Cottin C. [A wine pitcher, cause of lead poisoning]. Presse Med. 1996;25(6):243-246.
- Štěpánek L, Nakládalová M, Klementa V, Ferenčíková V. Acute lead poisoning in an indoor firing range. Med Pr. 2020;71(3):375-379. doi:10.13075/mp.5893.00930

- **10.** Gopinath B, Kappagantu V, Mathew R, Jamshed N. Acute lead poisoning: a diagnostic challenge in the emergency department. BMJ Case Rep. 2021;14(1):e239740. doi:10.1136/bcr-2020-239740
- **11.** Berkowitz S, Tarrago R. Acute Brain Herniation From Lead Toxicity. Pediatrics. 2006;118(6):2548-2551. doi:10.1542/peds.2006-1733
- **12.** Corradi M, Goldoni M, Sabbadini F, Mutti A. [Acute lead poisoning: a singular case of hemolytic anemia and lead colic]. Med Lav. 2011;102(3):243-249.
- **13.** Barber T, Jacyna M. Acute lead intoxication from medications purchased online presenting with recurrent abdominal pain and encephalopathy. J R Soc Med. 2011;104(3):120-123. doi:10.1258/jrsm.2010.100334
- 14. Offor SJ, Mbagwu HOC, Orisakwe OE. Lead Induced Hepato-renal Damage in Male Albino Rats and Effects of Activated Charcoal. Front Pharmacol. 2017;8. doi:10.3389/ fphar.2017.00107
- **15.** Hon KL, Fung CK, Leung AK. Childhoodleadpoisoning: an overview. Hong Kong Med J. 2017 Dec;23(6):616-21. doi: 10.12809/hkmj176214. Epub 2017 Oct 13.

Journal of Emergency Medicine Case Reports

Importance of Recombinant Tissue Plasminogen Activator (rt-PA) Thrombolysis in Hyperacute Stroke Patients with Aphasia-A Case Report

Ahmad Luqman Bin Md Pauzi¹, Juliana Hashim²

¹Department of Medicine, Faculty of Medicine and Health Science, University Putra, Malaysia

²Department of Pediatrics, Hospital Tuanku Jaafar Seremban, Malaysia

Abstract

Thrombolysis in an acute stroke has been shown to have a good outcome measure in an eligible candidate. However, the benefits and effectiveness of it in acute minor stroke are still not well established. We describe a case of 35-year-old male who was presented with isolated aphasia with the total National Institutes of Health Stroke Scale of 3 out of 42. Magnetic Resonance Imaging of the brain showed a focal left middle cerebral artery territory infarction secondary to distal vessel occlusion at M4 segment distally on Diffusion-Weighted Imaging sequence which was mismatched on Fluid-Attenuated Inversion Recovery sequence. Recombined tissue plasminogen activator was administrated intravenously without any complication. The patient's condition improved gradually with positive outcome measures. Intravenous r-TPA can be a beneficial treatment for a subset of patients with minor stroke.

Keywords: Alteplase, isolated aphasia, minor stroke, thrombolysis

Introduction

When an acute stroke occurs within a window of less than 4.5 hours, intravenous recombinant tissue plasminogen activator (r-TPA) is still the primary treatment option. There is a chance of haemorrhages with this medication, particularly intracranially. Therefore, the treating physician can be reluctant or uneasy about thrombolyzing this group of patients, especially those who have shown up with mild symptoms like isolated aphasia, because the danger of injury to the patient is greater than the benefit of treatment (1). Furthermore, even in the absence of thrombolysis, this kind of small stroke has a favourable prognosis within three months of treatment and a 75% good recovery rate (2).

However, there is still controversy as to which category the patient with a minor stroke falls into. Most researchers or clinicians use the National Institutes of Health (NIHSS) Stroke Scale as a stroke assessment tool and dictate a score from 0 to 4 as an indicator of a mild stroke (3). This may lead to inaccuracies as the NIHSS score is weighted more towards the anterior circulation, specifically the left (dominant) hemisphere (4), compared to other areas such as the right hemisphere or the posterior circulation. Stroke affecting isolated area of language without motor or sensory involvement is rare and is often due to mimic strokes, such

as those of toxic or metabolic origin (5). However, if the stroke itself exhibits aphasia, this predicts an unfavourable prognostic marker for a minor stroke (6). This article describes the benefit of aggressive thrombolysis in a mild, disabling symptomatic stroke that has achieved a good outcome within a few months.

Case Report

We described a 35-year-old Malay man who had a history of chronic smoking who presented with generalised body weakness that caused him to fall in the bathroom. Subsequently, he develops aphasia and family members noticed the patient lost the ability to talk. Within two hours of the commencement of symptoms, he was in the emergency stroke unit. Upon the initial assessment, he was alert with expressive aphasia. Apart from that, there was no drift noted over his right and left upper or lower limbs. There were no noteworthy findings from any other neurological examinations. The patient was able to respond to the question by indicating the correct response on a sheet of paper during the NIHSS assessment for component no. 1b. He received a score of 1 for dysarthria and 2 for best language on component no. 10. The total NIHSS score was 3 out of 42.

Corresponding Author: Ahmad Luqman Bin Md Pauzi e-mail: ahmadluqman@upm.edu.my Received: 03.12.2024 • Accepted: 11.02.2025 DOI: 10.33706/jemcr.1592792

©Copyright 2020 by Emergency Physicians Association of Turkey - Available online at www.jemcr.com

Cite this article as: Bin Md Pauz AL, Hashim J. Importance of Recombinant Tissue Plasminogen Activator (rt-PA) Thrombolysis in Hyperacute Stroke Patients with Aphasia-A Case Report. Journal of Emergency Medicine Case Reports. 2025;16(1): 34-

The following clinical information was available at admission: thrombocyte count, 367 × 103 mm³ (normal: $130 \times 103-400 \times 103$ mm³), international normalised ratio (INR), 1.04, prothrombin time (PT), 11.3 seconds (control: 9.95–11.34 seconds), activated partial thromboplastin time (aPTT), 25.20 seconds (normal: 32.36-42.0 seconds), and creatinine level, 70 umol/L. The brain's magnetic resonance imaging (MRI) revealed an acute infarct in the left temporoparietal region, which corresponds to the left middle cerebral artery (MCA) territory. This was probably caused by a distal vessel occlusion (DVO), and the imaging from the Fluid-attenuated Inversion Recovery (FLAIR) sequence appears mismatched with the Diffusion-weighted Imaging (DWI) sequence. Additionally, there was no blooming effect that would indicate bleeding on the susceptibility-weighted imaging (SWI) sequence (Figure-1). Magnetic Resonance Angiography (MRA) also shows no evidence of major vessel blockage (Figure-2).

It was decided to thrombolyse the patient after considering the patient's clinical presentation of a minor stroke, which was based on the NIHSS score of 3 but was also linked to a disabling aphasic symptom. The clinical advantage of r-TPA in this sort of mild stroke was also taken into consideration. Informed consent was obtained before 0.9 mg/kg of r-TPA with alteplase was infused intravenously. During and after r-TPA administration, no immediate complications arise. On day four following thrombolysis, a repeat MRI revealed a

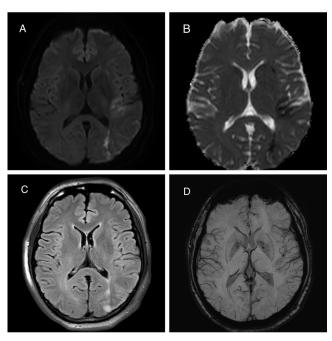


Figure 1. A-B: Showing diffusion weighted imaging (DWI) (Figure-1A) and the apparent diffusion coefficient (ADC) (Figure-1B) map showing acute infarct at the left temporo-parietal region, corresponding to the left middle cerebral artery (MCA) territory, likely due to a distal vessel occlusion (DVO).

C: No matched high signal intensity is noted on the fluid attenuated inversion recovery (FLAIR) sequence.

D: No blooming is seen on the susceptibility weighted imaging (SWI) sequence to suggest hemorrhage.

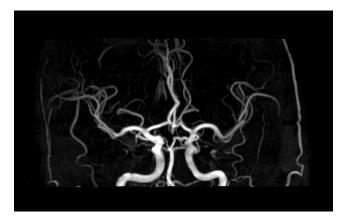


Figure 2. MRA demonstrated no obstruction of large vessel in carotid artery, anterior, middle, and posterior cerebral artery & basilar artery.

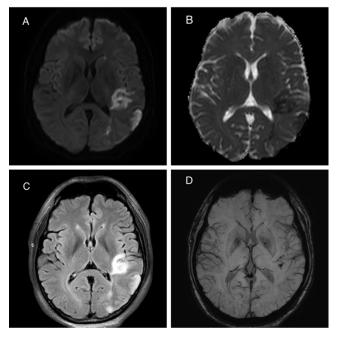


Figure 3. Repeated brain magnetic resonance imaging (MRI) on day 4 showing:

A-B: The DWI and ADC map shows established infarct at the left temporoparietal region.

C: Corresponding matched abnormal high signal intensity is more conspicuous on the FLAIR sequence.

D: Minimal microhaemorrhages are seen.

confirmed infarct at the left temporoparietal area on the DWI and ADC maps, accompanied with mild microhaemorrhages and an abnormally high signal intensity that was more noticeable on the FLAIR sequence (Figure-3). On the fifth day after his stay, he was released with a slight aphasia that was still present, but an improved NIHSS score to 1. He had a good score of 97 on the Western Aphasia Battery (WAB) when he was examined during the follow-up in the clinic one month's time.

Discussion

It is still debatable and not fully demonstrated whether thrombolysis is safe and appropriate in cases of acute mild stroke. Although there are currently numerous methods and scoring systems for defining a mild stroke, a study indicates that there are no appreciable changes in the outcome measure between patients who receive treatment and those who do not, with roughly 30% of the former experiencing a bad outcome within 90 days (7). An intracranial and extracranial vascular occlusive lesion on imaging, or a parameter in the NIHSS component such as leg weakness and extinction/inattention, may be the cause of the adverse outcome in a mild ischaemic stroke (8). Additionally, on vascular imaging, some people who have had small strokes may have substantial artery blockage. If thrombolysis is not performed at the time of the initial mild stroke presentation, this manifestation will have a poor outcome measure with a Modified Rankin Scale (MRS) score of less than or equal to 2 at three months after the stroke (9). It also carries a risk of having a full-blown stroke at any time. In addition, thrombolysis has been demonstrated to have positive results in cases of mild stroke, especially in cases of left hemisphere lesions, with an estimated 47% recovery rate with no lingering symptom s(10).

This patient is receiving thrombolytic therapy because, despite the NIHSS assessment indicating a mild stroke, greater attention should be paid to the evaluation of the patient's neurological deficiencies and how they affect functional impairment (11). According to research by Maas (2012), 57% of patients experienced a spontaneous improvement in their symptoms, which was followed by a 38% resolution. This kind of stroke has a recovery rate of about 90%, which raises doubts about the necessity of thrombolysis in certain situations (12). Nesi et al., however, conducted a prospective study on aphasic stroke patients with NIHSS scores below 6 who later received thrombolysis and reported a positive outcome and notable recovery after three months. This implies that thrombolysis should be administered to stroke patients with isolated aphasia in the early evaluation, irrespective of their NIHSS score (13). Additionally, this subgroup of patients showed improvements in their composite language score, language screening test, and NIHSS score following thrombolysis. Surprisingly, none of the thrombolysed patients experienced cerebral haemorrhage (14). Because of the extent of the neural network encompassing the language region, this subset of patients is at risk for a poor prognosis, which is why urgent treatment is necessary. Bridging the penumbra in a small infarct area enhances overall functional improvement. But, if conservatively handled, it can cause the infarcted area to spread into the language area, which would have a negative effect as it worsens (15).

Inlightof this data, in 2019 the American Heart Association (AHA) developed updated guidelines for treating hyperacute stroke patients who had mild symptoms. It has been proposed that, if qualified and within a reasonable time frame, patients with small strokes

who exhibit incapacitating symptoms that could interfere with their ability to return to work or engage in everyday activities should receive thrombolysis treatment (16). In this instance, the patient's NIHSS score significantly improved after thrombolysis, going from 3 to 1 upon discharge. His clinic follow-up revealed a good recovery with few signs of aphasia. The severity of aphasia was assessed at clinic follow-up using the Western Aphasia Battery (WAB) score. The main clinical components of language function—content, fluency, auditory comprehension, repetition, naming, as well as reading, writing, and calculation—will be evaluated by this WAB score. With a score of 97, the patient's WAB score after one month was considered modest.

Conclusion

Information about the use of thrombolysis in treating acute mild stroke with isolated aphasia is currently scarce. However, thrombolysis has been shown to have a positive effect in isolated aphasia because of the language area's extensive neural network, which provides a chance for higher functional improvement because of its collateral blood supply. To determine the best course of treatment and the safety profiles of rTPA thrombolysis in this single aphasic patient, more research is required.

- Werner Hacke, M.D., Markku Kaste, M.D., Erich Bluhmki, Ph.D., Miroslav Brozman, M.D., Antoni Dávalos, M.D., Donata Guidetti, M.D., Vincent Larrue, M.D., Kennedy R. Lees, M.D., Zakaria Medeghri, M.D., Thomas Machnig, M.D., Dietmar Schneider, M.D., Rüdiger von Kummer, M.D., et al. Thrombolysis with Alteplase 3 to 4.5 Hours after Acute Ischemic Stroke. N Engl J Med 2008; 359:1317-1329
- Krassen Nedeltchev, Benjamin Schwegler, Tobias Haefeli, Caspar Brekenfeld, Jan Gralla, Urs Fischer, Marcel Arnold, Luca Remonda, Gerhard Schroth, and Heinrich P. Mattle. Outcome of Stroke With Mild or Rapidly Improving Symptoms. Stroke. 2007;38:2531–2535
- 3. Urs Fischer, Adrian Baumgartner, Marcel Arnold, Krassen Nedeltchev, Jan Gralla, Gian Marco De Marchis, Liliane Kappeler, Marie-Luise Mono, Caspar Brekenfeld, Gerhard Schroth, and Heinrich P. Mattle. What Is a Minor Stroke? Stroke. 2010;41:661–666
- **4.** NIH Stroke Scale (NIHSS). NIH stroke scale International; 1999; http://www.nihstrokescale.org/.
- **5.** Casella G, Llinas RH, Marsh EB. Isolated aphasia in the emergency department: The likelihood of ischemia is low. Clin Neurol Neurosurg. 2017 Dec;163:24-26.
- **6.** Nesi M, Lucente G, Nencini P[,] Fancellu L, Inzitari D. Aphasia predicts unfavorable outcome in mild ischemic stroke patients and prompts thrombolytic treatment. J Stroke Cerebrovasc Dis. 2014 Feb;23(2):204-8.
- Ilana Spokoyny, MD, Rema Raman, PhD, Karin Ernstrom, MS, Pooja Khatri, MD, MSc, Dawn M. Meyer, PhD, RN, FNP-C,

- Thomas M. Hemmen, MD, PhD, and Brett C. Meyer, MD Defining Mild Stroke: Outcomes Analysis of Treated and Untreated Mild Stroke Patients. J Stroke Cerebrovasc Dis. 2015 June; 24(6): 1276–1281.
- **8.** Sato S, Uehara T, Ohara T, Suzuki R, Toyoda K, Minematsu K. Factors associated with unfavorable outcome in minor ischemic stroke. Neurology, 06 Jun 2014, 83(2):174-181.
- 9. Mirjam R. Heldner, Panagiotis Chaloulos-lakovidis, Leonidas Panos, Bastian Volbers, Johannes Kaesmacher, Tomas Dobrocky, Pasquale Mordasini, Marwan El-Koussy, Jan Gralla, Marcel Arnold, Urs Fischer, Heinrich P. Mattle & Simon Jung. Outcome of patients with large vessel occlusion in the anterior circulation and low NIHSS score. Journal of Neurology volume 267, pages 1651–1662(2020).
- **10.** Köhrmann M. Nowe T. Huttner H.B Engelhorn T. Struffert T- Kollmar R· Saake M· Doerfler A · Schwab S· Schellinger P.D Safety and Outcome after Thrombolysis in Stroke Patients with Mild Symptoms. Cerebrovasc Dis 2009; 27:160–166.
- 11. Matthias Wendt, Serdar Tütüncü, Jochen B Fiebach, Jan F Scheitz, Heinrich J Audebert, Christian H Nolte. Preclusion of ischemic stroke patients from intravenous tissue plasminogen activator treatment for mild symptoms should not be based on low National Institutes of Health Stroke Scale Scores. J Stroke Cerebrovasc Dis 2013 May;22(4):550-3.
- **12.** Maas MB, Lev MH, Ay H, Singhal AB, Greer DM, Smith WS, et al: The prognosis for aphasia in stroke. J Stroke Cerebrovasc Dis 2012; 21:350–357.
- **13.** Nesi M, Lucente G, Nencini P, Fancellu L, Inzitari D: Aphasia predicts unfavorable outcome in mild ischemic stroke

- patients and prompts thrombolytic treatment. J Stroke Cerebrovasc Dis 2014;23:204–208.
- **14.** C Denier, O Chassin, C Vandendries, L Bayon de la Tour, C Cauquil, M Sarov, D Adams, C Flamand-Roze. Thrombolysis in Stroke Patients with Isolated Aphasia. Cerebrovasc Dis 2016;41(3-4):163-9.
- **15.** Kremer, C., Kappelin, J., & Perren, F. (2014). Dissociation of severity of stroke and aphasia recovery early after intravenous recombinant tissue plasminogen activator thrombolysis. Journal of Clinical Neuroscience, 21(10), 1828–1830. doi:10.1016/j.jocn.2014.01.010
- 16. Guidelines for the Early Management of Patients with Acute Ischemic Stroke: 2019 Update to the 2018 Guidelines for the Early Management of Acute Ischemic Stroke: A Guideline for Healthcare Professionals from the American Heart Association/American Stroke Association. William J. Powers, MD, FAHA, Chair, Alejandro A. Rabinstein, MD, FAHA, Vice Chair, Teri Ackerson, BSN, RN, Opeolu M. Adeoye, MD, MS, FAHA, Nicholas C. Bambakidis, MD, FAHA, Kyra Becker, MD, FAHA, José Biller, MD, FAHA, Michael Brown, MD, MSc, Bart M. Demaerschalk, MD, MSc, FAHA, Brian Hoh, MD, FAHA, Edward C. Jauch, MD, MS, FAHA, Chelsea S. Kidwell, MD, FAHA, Thabele M. Leslie-Mazwi, MD, Bruce Ovbiagele, MD, MSc, MAS, MBA, FAHA, Phillip A. Scott, MD, MBA, FAHA, Kevin N. Sheth, MD, FAHA, Andrew M. Southerland, MD, MSc, FAHA, Deborah V. Summers, MSN, RN, FAHA, David L. Tirschwell, MD, MSc, FAHA, on behalf of the American Heart Association Stroke Council

Journal of Emergency Medicine Case Reports

Superior Trunk Block: A Novel Approach for Shoulder Reduction in Pregnancy

D Chun Chau Tan¹

¹Emergency and Trauma Department, Hospital Seberang Jaya, Jln Tun Hussein Onn, 13700 Seberang Jaya, Pulau Pinang, Malaysia

Abstract

Anterior shoulder dislocations are common in emergency settings but are rarely reported in pregnant patients, necessitating careful pain management to ensure maternal and fetal safety. While procedural sedation and analgesia are conventional approaches, they carry risks such as altered maternal consciousness, raising concerns during pregnancy. Ultrasound-guided regional anaesthesia offers a safer and more effective alternative, particularly nerve blocks. We present the case of a 30-year-old woman at 28 weeks of gestation with a recurrent anterior shoulder dislocation. An ultrasound-guided superior trunk block was performed, targeting the superior trunk of the brachial plexus, which is formed by the C5-C6 nerve roots, offering a promising alternative to the interscalene brachial plexus block for shoulder procedures in pregnant patients. This technique provided effective analgesia, facilitating successful closed manipulative reduction without maternal or fetal complications, thereby supporting its broader adoption in similar clinical context scenarios.

Keywords: Regional anesthesia, shoulder dislocation, superior trunk block

Introduction

Anterior shoulder dislocation (ASD) is the most common type of shoulder dislocation and a frequent presentation in emergency departments (ED). While reduction is essential in the early management of shoulder dislocations, it is a painful procedure. Its occurrence in pregnant patients is rare and not well documented, necessitating careful pain management to ensure maternal comfort, fetal safety, and procedural success.

Various approaches to managing shoulder dislocations include procedural sedation and analgesia (PSA), intra-articular analgesia (IAA), and ultrasound-guided regional anaesthesia (UGRA). While PSA is commonly used, its potential for adverse effects, such as compromised maternal consciousness, raises concerns in the emergency setting. Ultrasound-guided regional anaesthesia, including nerve block techniques, is increasingly recognised as a safer alternative in these scenarios.

The superior trunk block has emerged as a promising option for shoulder procedures, targeting the C5-C6 nerve roots. Compared to the interscalene brachial plexus block, it provides effective analgesia with a reduced risk of complications, such as hemidiaphragmatic paralysis (HDP). This makes it particularly suitable for pregnant patients requiring shoulder reduction.

In this case report, we describe the successful use of a superior trunk block (STB) for reducing ASD in a pregnant patient. It underscores the potential of this technique to provide effective analgesia with minimal risk, presenting it as a safer alternative for managing shoulder dislocations during pregnancy. Additionally, we review the literature comparing UGRA techniques with PSA and IAA for shoulder procedures.

Case Report

A 30-year-old Malay woman, gravida 2 para 1 at 28 weeks of pregnancy, presented to the ED with recurrent right shoulder dislocation. Her antenatal history included idiopathic generalised seizures, chronic hypertension, and maternal obesity. She reported a sudden onset of right shoulder pain after reaching for a water bottle from the back seat of her car with her right arm in abduction, external rotation, and extension. The pain, worsened by movement and partially relieved by supporting the arm, was not associated with numbness, tingling, or weakness. There were no associated abdominal pain, per vaginal bleeding, or changes in fetal movements. The patient had a prior history of right shoulder dislocation 10 weeks earlier due to a fall, which

Corresponding Author: Chun Chau Tan
e-mail: charlestan89@hotmail.com
Received: 09.12.2024 • Accepted: 20.02.2025
DOI: 10.33706/jemcr.1598621

Cite this article as: Tan CC. Superior Trunk Block: A Novel Approach for Shoulder Reduction in Pregnancy. Journal of Emergency Medicine Case Reports. 2025;16(1): 38-41

©Copyright 2020 by Emergency Physicians Association of Turkey - Available online at www.jemcr.com

was successfully reduced under procedural sedation and analgesia without complications.

On examination, the patient was alert and reported moderate pain with a pain score 5. Her vital signs were stable, with a blood pressure of 130/75 mmHg, heart rate of 83 bpm, oxygen saturation of 98% on room air, and a temperature of 36.7°C. The right shoulder inspection revealed a loss ofright shoulder contour with no obvious wound, swelling or bruises. The arm was held in abduction and a slight external rotation position with an arm sling. On palpation, there was tenderness over the right shoulder joint with step-off deformity, no crepitus and no warmth. Neurovascular status was intact, with normal sensation and distal pulses. The patient weighed 90 kg, with a BMI of 37.5 kg/m². Abdominal examination was consistent with the gestational age, showing a soft, non-tender abdomen and a gravid uterus palpated four fingerbreadths above the umbilicus. Other systemic examinations were unremarkable.

A right shoulder X-ray confirmed an anterior dislocation of the humeral head without associated fractures (Figure-1a). The diagnosis was right ASD requiring closed manipulative reduction (CMR). The patient was informed of the available analgesic options for the procedure. After a discussion of the risks and benefits, she agreed to a superior trunk block (STB) for shoulder reduction under ultrasound guidance. She was prepared in the resuscitation bay with standard monitoring applied and resuscitation drugs and equipment on standby.

STB was performed using a high-frequency linear probe (6-12 MHz) on a GE Logiq V ultrasound machine and a 22G × 80 mm Pajunk® SonoTAP® block needle under aseptic conditions. During the scanning phase, the C5 and C6 nerve roots were visualised at the cricoid cartilage level, positioned within the groove between the anterior and middle scalene muscles (Figure-2a). The STB targeted the superior trunk of the brachial plexus, after C5 and C6 nerve root convergence (Figure-2b) but before the branching of the suprascapular nerve (Figure-2c). 10 mL of 1% lignocaine was administered using an in-plane technique, with lateral to medial needle insertion relative to the transducer probe (Figure-3). Analgesia was effectively achieved within five minutes, with no subjective increase in breathing effort. Ipsilateral diaphragm sonography showed no paradoxical movement, confirming the absence of phrenic nerve paralysis.

The patient then underwent a successful CMR using the external rotation manoeuvre (ERM) without requiring rescue analgesia. She reported tolerable pain during the procedure, with a pain score of 2. A collar-and-cuff sling was applied, and a follow-up X-ray confirmed successful reduction (Figure-1b). The patient reported minimal pain (pain score 1) post-procedure and was discharged an hour later with an outpatient follow-up appointment. At a next-day telephone follow-up, the patient reported high satisfaction, minimal pain (pain score 1), and no block-related complications. Written informed consent was obtained from the patient.

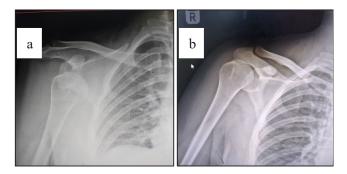


Figure 1. Right shoulder x-ray (anteroposterior view): (a) Pre-reduction right shoulder x-ray showed anterior right shoulder dislocation with the right humeral head dislocated anteriorly and inferiorly with the anteroinferior margin of the glenoid.(b) Post-reduction showed a normally aligned right humeral head with glenoid, and no fracture was seen.

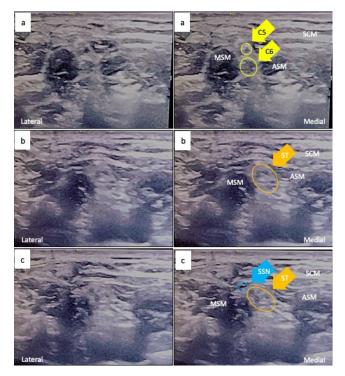
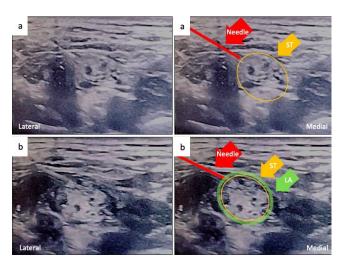


Figure 2. Sequential ultrasound images of the brachial plexus, captured from proximal-to-distal views. The image on the left is unlabelled, while the image on the right is annotated for clarity. Scanning phase of Right Superior Trunk Block (STB): (a) Brachial plexus at the interscalene block level with the classic "stoplight sign" form by C5 and C6 (Yellow) nerve root. (b) The superior trunk of brachial plexus (Orange) at the superior trunk block level. (c) Superior trunk view just distal to take-off of the suprascapular nerve, SSN (Blue).

C5: C5 nerve root; C6: C6 nerve root; MSM: Middle scalene muscle; ASM: Anterior scalene muscle; SCM: Sternocleidomastoid muscle; ST: Superior trunk of brachial plexus; SSN: Suprascapular nerve; Circles identify nerves and/or nerve group. Source: Original Image.

Discussion

Shoulder dislocation during pregnancy is uncommon but carries significant implications. It presents unique challenges due to physiological changes and the need to consider maternal and fetal well-being. The injury often results from a fall on an outstretched hand, with recurrent dislocations



Superior Trunk Block: A Novel Approach for

Shoulder Reduction in Pregnancy

Figure 3. Side-by-side ultrasound images of the brachial plexus at the level of Superior Trunk Block (STB). The image on the left is unlabelled, while the image on the right is annotated for clarity. (a) The needling phase of Right Superior Trunk Block: Regional anaesthesia (RA) needle (Red) advancement towards the superior trunk (ST) of the brachial plexus. (b) Local anaesthesia (LA) depositing phase of Right Superior Trunk Block: Injecting LA in the subparaneural space of the ST, demonstrating the characteristic 'halo' sign of successful deposition (Green).

occurring in about 20% of patients after an initial episode (1). Hormonal and physiological changes in pregnancy, particularly increased ligament laxity induced by relaxin, may predispose patients to shoulder instability and recurrent dislocations (2). In this case, the patient presented with atraumatic recurrent ASD 10 weeks after the first episode.

Severe pain from shoulder dislocation often triggers muscle spasms around the joint, impeding successful reduction. Achieving adequate muscle relaxation is essential for timely and effective management. A network meta-analysis compared the efficacy and safety of three methods—procedural sedation and analgesia (PSA), intra-articular anaesthesia (IAA), and ultrasound-guided regional anaesthesia (UGRA)-for the reduction of anterior shoulder dislocations. The analysis found no differences in reduction success rates and patient satisfaction among the methods. Notably, IAA and UGRA were associated with the absence of adverse respiratory events (3).

Procedural sedation and analgesia (PSA) remain a common approach in many emergency departments, employing agents such as midazolam, remifentanil, propofol, ketamine, and nitrous oxide. Evidence supports the judicious use of PSA in pregnant patients experiencing severe pain, distress, or requiring surgical intervention (4). While brief, low-dose PSA is generally considered safe in pregnancy, it requires careful consideration, particularly in third-trimester patients with underlying morbid obesity. Potential risks include unintentional deep sedation, respiratory or cardiovascular depression, pulmonary aspiration, and possible effects on the fetus, underscoring the need for meticulous monitoring and skilled administration (4). Furthermore, PSA is timeconsuming, necessitating prolonged monitoring during and after the procedure (5).

Given these concerns, alternative techniques such as IAA and UGRA are often considered for safer and more effective shoulder reduction. IAA has shown comparable effectiveness to PSA for ASD in the ED, with advantages such as fewer adverse events, shorter ED stays, and no significant differences in pain scores or ease of reduction (5, 6). However, IAA is typically administered using landmark guidance, which reported a misplacement rate of 41.1% in anterior dislocations. Although ultrasound guidance may improve accuracy, studies comparing landmark- and ultrasoundguided IAA are lacking (5). Despite its advantages, IAA has some trade-offs, including longer procedure times, slightly more challenging reductions, and lower patient satisfaction scores. The anxiety and fear associated with joint dislocations may also be difficult to address with IAA. Nevertheless, considering the risks associated with PSA, IAA emerges as a preferred option, particularly when minimising sedationrelated side effects is a priority (6).

UGRA has an advantage over PSA as it provides effective pain relief without systemic effects associated with sedation. With advanced training and expertise in point-of-care ultrasound, emergency physicians employ ultrasound-guided regional anaesthesia (UGRA) for managing acute pain in the emergency department (ED) (7). It is regarded as a safe and effective option for providing analgesia during the closed reduction of shoulder dislocations in the ED (5, 8). UGRA shortens ED discharge times and reduces the risk of adverse events and complications, especially respiratory-related issues (5, 8). Several techniques of nerve blocks can be used for the reduction of ASD, including interscalene brachial plexus block (ISB), superior trunk block (STB), supraclavicular brachial plexus block (SCB) and suprascapular nerve block (SSNB) (9).

ISB is the most commonly used peripheral nerve block for shoulder surgeries and procedures, targeting the brachial plexus at C5 and C6 nerve roots. This block is performed in the interscalene groove, located at the level of the cricoid cartilage between the anterior and middle scalene muscles. While effective, ISB has inherent risks, as the phrenic nerve is located within 2 mm of the brachial plexus at this level before diverging caudally (10). Consequently, phrenic nerve palsy and hemidiaphragmatic paralysis (HDP) are unavoidable side effects (9). These effects are particularly concerning during the third trimester of pregnancy, especially in patients with maternal obesity. The combination of reduced diaphragmatic function from HDP and the physiological respiratory changes of pregnancy can significantly compromise ventilation and oxygenation, posing a serious risk to maternal respiratory function.

Compared to the ISB, the SBT provides non-inferior surgical anaesthesia while preserving diaphragmatic function. Administering local anaesthetic at a more distal site reduces the risk of phrenic nerve paralysis (10). Targeting the superior trunk of the brachial plexus at a more inferolateral location, the STB provides effective pain relief while significantly reducing the incidence of HDP compared to the ISB (4.8% vs. 71.4%) (10-12). Additionally, the reduced proximal spread of local anaesthetic to the cervical sympathetic chain and recurrent laryngeal nerve results in a lower risk of Horner's syndrome and hoarseness of voice (10, 12).

The STB and ISB exhibited low complication rates overall, with no reports of major neurological complications, local anaesthetic systemic toxicity, or other adverse events (10). However, STB offers distinct advantages, including reduced needle-nerve contact due to its well-defined connective tissue sheath, a minimised risk of injury to the dorsal scapular and long thoracic nerves, increasing the likelihood of block success by identifying C5 nerve root anomalies, which present in 30-35% of cases (11). Additionally, STB was superior to ISB in providing better pain relief at rest 24 hours post-procedure (10,12).

A more distal block that can avoid HDP is the SCB, which targets the brachial plexus at the level of the tightly packed upper, middle, and lower trunks in the supraclavicular fossa. However, SCB provides anaesthesia to almost the entire upper extremity, which may be excessive for a shoulder reduction procedure. Adequate volume (approximately 20 mL) is required to ensure sufficient spread to the suprascapular nerve (SSN) proximally (12). The SSN arises from the superior trunk of the brachial plexus, located proximal to the division of the plexus into its cords (medial, lateral, and posterior) in the supraclavicular region. As a terminal branch, the SSN supplies up to 75% of the sensory input to the shoulder joint. The SSNB offers effective analgesia comparable to an ISB while avoiding the risk of phrenic nerve palsy (13).

The STB was an ideal choice for analgesia in this case, as it avoided sedation-related risks, preserved respiratory function, and accounted for fetal safety-critical considerations for pregnant patients. The procedure was highly successful, achieving smooth shoulder reduction in a single attempt. The patient was discharged in good condition and reported high satisfaction during follow-up, highlighting the efficacy and safety of the STB in managing shoulder dislocations during pregnancy.

Conclusion

The successful use of an STB for reducing ASD in a pregnant patient highlights the efficacy and safety of this technique. The STB provided effective analgesia, facilitating a smooth and successful closed manipulative reduction without any maternal or fetal complications. This case supports the broader adoption of the STB in similar clinical scenarios.

Source(s) of Support and FundingNone

Conflict of Interest Statement None declared.

- **1.** Gottlieb M. Shoulder dislocations in the emergency department: a comprehensive review of reduction techniques. The Journal of Emergency Medicine. 2020;58(4):647-66.
- **2.** Owens BD, Cameron KL, Clifton KB, Svoboda SJ, Wolf JM. Association between serum relaxin and subsequent shoulder instability. Orthopedics. 2016;39(4):e724-e8.
- **3.** Hayashi M, Kano K, Kuroda N, Yamamoto N, Shiroshita A, Kataoka Y. Comparative efficacy of sedation or analgesia methods for reduction of anterior shoulder dislocation: A systematic review and network meta analysis. Academic Emergency Medicine. 2022;29(10):1160-71.
- **4.** Neuman G, Koren G. Safety of procedural sedation in pregnancy. Journal of obstetrics and gynaecology Canada. 2013;35(2):168-73.
- **5.** Gawel RJ, Grill R, Bradley N, Luong J, Au AK. Ultrasound-guided peripheral nerve blocks for shoulder dislocation in the emergency department: a systemic review. The Journal of Emergency Medicine. 2023;65(5):e403-e13.
- 6. Sithamparapillai A, Grewal K, Thompson C, Walsh C, McLeod S. Intra-articular lidocaine versus intravenous sedation for closed reduction of acute anterior shoulder dislocation in the emergency department: a systematic review and meta-analysis. Canadian Journal of Emergency Medicine. 2022;24(8):809-19.
- Kumaran SNS, MEmMed MLH, binti Jusoh SR. Development of ultrasound guided regional anaesthesia in the emergency department, Hospital Kuala Lumpur. Med J Malaysia. 2024;79(3):371.
- **8.** Yao L, Dong W, Wu Z, Zhao Q, Mao H. Ultrasound-guided interscalene block versus intravenous analgesia and sedation for reduction of first anterior shoulder dislocation. The American Journal of Emergency Medicine. 2022;56:232-5.
- **9.** Campbell AS, Johnson CD, O'Connor S. Impact of Peripheral Nerve Block Technique on Incidence of Phrenic Nerve Palsy in Shoulder Surgery. Anesthesiology Research and Practice. 2023;2023(1):9962595.
- 10. Amaral S, Lombardi RA, Medeiros H, Nogueira A, Gadsden J. Superior Trunk Block Is an Effective Phrenic-Sparing Alternative to Interscalene Block for Shoulder Arthroscopy: A Systematic Review and Meta-Analysis. Cureus. 2023;15(11).
- 11. Laurent DB-S, Chan V, Chin KJ. Refining the ultrasound-guided interscalene brachial plexus block: the superior trunk approach. Canadian Journal of Anesthesia. 2014;61(12):1098-102.
- **12.** Kim DH, Lin Y, Beathe JC, Liu J, Oxendine JA, Haskins SC, et al. Superior trunk block: a phrenic-sparing alternative to the interscalene block: a randomized controlled trial. Anesthesiology. 2019;131(3):521-33.
- **13.** Mohanty CR, Gupta A, Radhakrishnan RV, Singh N, Patra SK. Ultrasound-guided low-volume anterior suprascapular nerve block for reduction of anterior shoulder dislocation in the emergency department: A case series. Turkish journal of emergency medicine. 2023;23(4):254-7.

Journal of Emergency Medicine Case Reports

Unexpected Ant Bites

Levent Şahin¹, Barış Kaban¹

¹KafkasUniversity, Faculty of Medicine, Department of Emergency Medicine, Kars, Türkiye

Abstract

Allergic reactions to insect bites and bee stings are frequently seen. In some cases, angioedema may be seen, more so in the pediatric age group. However, angioedema after an ant bite is a very rare condition. In this case, we present a young child who developed angioedema after being bitten by black ants on his penis and scrotum while playing in a rural area. The patient was given both prednisone and antihistamines and ice therapy in the emergency room. After the treatment, the patient's complaints of pain, swelling and redness decreased. With this study, we have shown that even ant bites in children can cause serious allergic consequences. Parents should be careful about serious allergic emergencies that may ocur after insect bites/stings in children.

Keywords: Angioedema, ant bite, child, penis

Introduction

Bees are the first in line among insect stings or bites seen in humans (1). The penis, in particular, has drawn the wrath of animals and insects (2). After bee stings, severe allergic reactions such as angioedema and even anaphylaxis are more common in the young and pediatric age group (1). The most common signs and symptoms are stinging pain, edema and redness at the site of the sting. A temporary, sometimes episodic, non-pitting edema involving the subcutaneous or submucosal tissue is called angioedema. Angioedema, seen in the pediatric age group, can also ocur after insectbites (3). When we conducted a literature review on the subject, we found that the published cases we remostly allergic reactions in the cornea, penis and mouth regions due to bee stings (4). However, cases with serious symptoms after ant bites are rare. This study will show that angioedema occurs in the penis and scrotum after the bite of black ants, which is quite rare.

Case Report

A 6-year-old boy, who went on a picnic with his family, suddenly reported pain in his penis and scrotum to his parents. When his mother and father opened their child's clothes and checked, they saw black ants. They saw redness

and slight swelling in the scrotum and penis. They brought the child to the emergency room because of his persistent pain, crying and restlessness. Approximately 60 minutes had passed since the incident when the patient arrived. The blood pressure, pulse, respiration and temperature values measured in triage were within normal ranges. In the physical examination, it was observed that the penis was painful and edematous, and the scrotum was both edematous and ecchymotic (Figure-1).

No pathology was detected in the blood values (hemogram, biochemistry) studied in the laboratory. No pathology was seen in the testicles in the superficial tissue ultrasound. The patient was given intravenous (i.v) antihistamine (1 mg/ kg pheniramin emaleate) in physiological serum and i.v. 2 mg/kg methylprednisolone was administered. Tetanus vaccination prophylaxis was also administered. Intermittent ice application was performed for the swelling in the penis and scrotum. After approximately 2 hours, there was a significant regression in the swelling and the pain decreased. The child, whose findings and complaints regressed during the control examination, was discharged with a prescription after a total of 8 hours of observation. The swelling and redness had completely disappeared in the child who came for control the next day; however, mild induration could be felt in the scrotum with palpation.



Figure 1. Skin findings on the penis and scrotum

Discussion

People can be bitten or stung by different insects. Bee stings are the most common among these (5). The most common signs and symptoms that ocur after insect bites are pain, redness and edema (1). Various allergic reactions can be seen in bee stings, ranging from mild skin findings to anaphylaxis. After ant bites, pain, swelling and redness may occur in the bitten area; however, angioedema is not an expected result. The areas bitten by ants are usually the limbs (6). In our case, the bite area was the penis and scrotum. More than half of the population living in areas where ants are densely populated are bitten every year (7). Since the region where the incident in our case occurred has cold climate conditions, the ant population is rare.

The reactions seen in more than half of the children exposed to insectstings/bites are usually limited to the skin. Although angioedema is observed in some cases, anaphylactic shock is almost non-existent (8). Delayed reactions such as coagulation disorders, vasculitis, peripheral neuropathy and serum sickness may be observed days after insectstings/bites (9). In our case, an early-type allergic reaction with pain and mild redness in the penis and scrotum was reported in the first minutes of the ant bite. On examination in the emergency room, angioedema was observed with increasing pain, but anaphylaxis was not observed. There was a local involvement limited to the penis skin and scrotum. Regardless of the cause, H1 and H2 antihistamines are preferred in the treatment of allergic angioedema to reduce itching and inflammation,

while steroids can also be added to thetreatment (10). The prognosis is usually good in ant bites among insectstings, and the patient's symptoms regress without the need for treatment. However, the possibility of anaphylaxis, although rare, cannot be ignored, and early diagnosis and treatment are of vital importance. The angioedema in our case rapidly regressed after the first treatment and was discharged.

Conclusion

To protect against possible insect bites/stings, long-sleeved clothing should be worn in open areas. Also, colorful clothing that looks like flowers should be avoided. Considering that they usually enter at ankle level, this entry way should be covered with socks. It should be kept in mind that angioedema may develop in children in the event of any insect bite despite all precautions, and a health institution should be consulted without delay.

- **1.** Ozkan A, Kaya M, Okur M, et al. Three-year-old boy with swelling and ecchymosis of the penis. Turk Arch Pediatr. 2011;46:267–268.
- **2.** Weiss HB, Friedman DI, Coben JH. Incidence of dog bite injuries treated in emergency departments. JAMA. 1998;279:51-3.
- **3.** Srividya KL, Vidyasagar P. Penile angioedema in a child: A nightmare to parents!.lndian Journal of Skin Allergy. 2022; 1(1): 21-23.
- Huseynov M, Cebisli A, Ozturk A, Sahin M, Ozdemir MA, Polat M. Pediatric Penile Bee Sting. Pediatric Emergency Care. 2022; 38(8): e1469-e1471.
- Golden DBK. Allergic reactions to insect stings. In: Biermann CW, Pearlman DS, Shapiro GG, Buse WW, (eds). Allergy asthma and immunology from infancy to adult hood. Philadelphia: Saunders. 1996:348-54
- **6.** Aliyu I. Ant sting at an unusual site. Muller Journal of Medical Sciences and Research. 2018; 9(1):38-39.
- Alsharani M, Alanazi M, Alsalamah M. Black ant stings caused by pachycondyla sennaarensis: A significant health hazard. AnnSaudi Med. 2009;29:207–11
- **8.** Kalyoncu AF, Demir AU, Ozcan U, Ozkuyumcu C, Sahin AA, Bariş YI. Bee and wasp venom allergy in Turkey. Ann Allergy Asthma Immunology. 1997;78:408-12.
- **9.** Bektaş S, Peker E, CağanE, ve ark. Arı sokmasını takiben konvulziyon geliştiren iki olgu sunumu. Tıp Araştırmaları Dergisi. 2010;8: 31-3.
- 10. Booker GM, Adam HM. Insect stings. Pediatr Rev. 2005;26:388-9.

Journal of Emergency Medicine Case Reports

Severe Pancytopenia Induced by Methotrexate in a Rheumatoid Arthritis Patient

¹Elazığ Fethi Sekin City Hospital, Department of Infectious Diseases, Elazığ, Türkiye

²Elazığ Fethi Sekin City Hospital, Department of Hematology, Elazığ, Türkiye

³Elazığ Fethi Sekin City Hospital, Department of Biochemistry, Elazığ, Türkiye

Abstract

Rheumatoid arthritis (RA) is a systemic autoimmune disease affecting approximately 1% of the global population, with a higher prevalence in women. Methotrexate (MTX), a disease-modifying antirheumatic drug (DMARD), is commonly used in RA treatment. However, improper or unsupervised use can lead to severe hematological toxicities, including pancytopenia. A 74-year-old female patient with RA presented with fever, oral ulcers, weakness, and difficulty swallowing. Physical examination revealed ecchymotic skin lesions and candidal plaques in the oral cavity. Laboratory results indicated severe pancytopenia (WBC: 1.1×10^{9} /L, RBC: 2.75×1012 /L, PLT: 9×10^{9} /L). Methotrexate intoxication was suspected. The patient received supportive treatment, including folate supplementation, Filgrastim for neutropenia, platelet apheresis, and erythrocyte transfusion. Scattergram analysis revealed marked alterations in neutrophil and eosinophil populations. Following treatment, hematological parameters normalized, but neutrophil volume changes persisted, suggesting a prolonged bone marrow effect of MTX. This case highlights the importance of routine complete blood count monitoring, including scattergram analysis, in RA patients receiving DMARD therapy. Laboratories should consider adjusting CBC measurement protocols in cases of suspected drug-induced pancytopenia to enhance diagnostic accuracy and early detection of hematological toxicity.

Keywords: Methotrexate toxicity, pancytopenia, rheumatoid arthritis

Introduction

Rheumatoid arthritis (RA) is a systemic autoimmune disease with a global prevalence of approximately 1%, and its prevalence increases with age. It is generally seen three to five times more frequently in women than in men, usually between the ages of 40 and 50. One of the treatment options is Methotrexate, which belongs to the group of disease-modifying antirheumatic drugs (DMARDs) (1).

Methotrexate is a folate antimetabolite that inhibits DNA synthesis, repair, and cellular replication. Methotrexate works by binding to and inhibiting dihydrofolate reductase, leading to reduced formation of reduced folate and inhibition of thymidylate synthase. This results in the inhibition of purine and thymidylic acid synthesis, thereby affecting DNA synthesis, repair, and cellular replication. Actively proliferative tissues are more sensitive to the effects of methotrexate (2).

In this case, a patient diagnosed with RA was initially started on Methotrexate therapy, which was later discontinued by the Rheumatology department. However, the patient continued to take and use her medications unsupervised for approximately 7 months, 2 tablets per day. As a result, the patient developed mouth ulcers, skin lesions, and pancytopenia. This case highlights the necessity of considering alternative modifying

agents or medications for geriatric patients who may not be able to take Methotrexate regularly and appropriately, or who cannot be monitored consistently. Additionally, in similar cases, it is emphasized that laboratory values, especially scattergram analyses of complete blood counts, should be used to assess the relationship with disease management.

Eosinophil elevation in rheumatoid arthritis is a very rare but documented condition. It remains controversial whether this increase is related to treatment independently of rheumatoid arthritis or if it is associated with the inflammatory processes caused by the disease (3). Additionally, there is literature indicating that the extent of eosinophilia may be a marker for the severity of rheumatoid arthritis, and that this change in value might be caused by disease-modifying antirheumatic drugs (4-9).

Rheumatoid arthritis is an incurable disease (1). In order to prevent the progression of the disease, disease-modifying drugs are used and efforts are made to prevent progression. Disease-modifying drugs for the disease are abbreviated as DMARDs and consist of Hydroxychloroquine, Sulfasalazine, Gold salts, D-penicillamine, Azathioprine, Cyclophosphamide, Methotrexate, Leflunomide, Tumor necrosis factor-α blockers, Anakinra Abatacept, and Rituximab. The most commonly used of these is Methotrexate (10).

corresponding Author: Hakan Ayyıldız
e-mail: hakan.ayyıldız@sbu.edu.tr
Received: 07.02.2025 • Revision: 20.02.2025 • Accepted: 24.02.2025
DOI: 10.33706/jemcr.1634492
©Copyright 2020 by Emergency Physicians Association of Turkey - Available online at www.jemcr.com

Cite this article as: Özgüler M, Koç A, Ayyıldız H. Severe Pancytopenia Induced by Methotrexate in a Rheumatoid Arthritis Patient. Journal of Emergency Medicine Case Reports. 2025;16(1): 44-47

A 74-year-old female patient presented to our hospital's emergency department with complaints of fever, widespread painful plaques in the mouth, oral intake disorder, weakness, and difficulty swallowing. During the patient's physical examination, prominent ecchymotic lesions on the body, widespread candidal plaques on the oral mucosa, areas of necrosis intermittently, and cryptic lesions on the tonsils were observed.

During the otolaryngologist's examination, crusting secondary to bleeding was observed at the nasal vestibules, while the nasal mucosa appeared normal. In the evaluation of the oral cavity, poor oral hygiene, aphthous ulcerative lesions on the bilateral buccal mucosa, and mucosal ulcerative aphthous lesions on the uvula and posterior oropharyngeal wall were observed. In the laryngeal evaluation, a yellowish plaque (suspected candidiasis) was noted on the laryngeal mucosa, while the bilateral vocal cords appeared normal and mobile. In addition, the patient's past history revealed that she had hypertension and asthma.

Due to the inability to measure the patient's complete blood count values, the presumptive (unconfirmed) diagnosis of the disease was communicated to the laboratory. A new sample was collected and analyzed in the laboratory using the reticulocyte mode on the device (Beckman Coulter DXH-800) by changing its routine working mode.

White Blood Cells: 1.1 (10⁹/L), Neutrophils: 0.1 (10⁹/L), Lymphocytes: 0.54 (10⁹/L), Eosinophils: 0.51 (10⁹/L), Monocytes: 0.01 (10⁹/L), Hemoglobin: 8.7 g/dL, Hematocrit: 24.6%, Platelets: 9 (10⁹/L) were detected (Figure-1 and

Table-1). In the peripheral smear examination, no atypical cells or blasts were observed, and leukopenia was noted. Platelet levels were consistent with the blood count.

Current findings were found to be compatible with intoxication due to uncontrolled methotrexate use. Other tests performed for the etiology of pancytopenia include Total Protein; 46 g/L (66-83), Albumin; 25 g/L (35-52), Urea; 69 mg/dL (12-50), Uric Acid; 4.96 mg/dL (2,6-6), AST; 10 U/L (5-40), ALT; 11 U/L (5-40), Total Bilirubin; 1.38 mg/dL (0,1-1,2), Direct Bilirubin; 0.40 mg/dL (0-0,3), Calcium; 7.5 mg/dL (8,8-10,6), Magnesium; 1.57 mg/dL (1,7-2,6), Phosphorus; 2.01 mg/dL(2,4-4,5), CRP; 333 mg/L (0-8), Erythrocyte sedimentation rate; 78 mm/h (0-30), Procalcitonin; 0.84 µg/L (0-0,1), vitamin B12; 351 ng/L (125-505), Folate; 8,66 µg/L (3,1-19,9)

The patient's radiological examinations revealed a normal chest X-ray. The patient, who had a fever and neutropenia, was started on Meropenem 3x1 g and Vancomycin 2x1 g with a preliminary diagnosis of febrile neutropenia. Methotrexate was discontinued and Folbiol 2x1 was initiated. Filgrastim 30 mIU (1×1 SC) was initiated with frequent hemogram monitoring. Due to the risk of bleeding, platelet apheresis was administered for thrombocytopenia, and erythrocyte suspension was infused for symptomatic anemia. On followup, platelet count was $58 (10^9/L)$, hemoglobin was 11 g/dL, and hematocrit was 32%.

Leukopenia improved during daily follow-ups after two days of Filgrastim. The patient, who had no fever after admission, had Meropenem and Vancomycin discontinued on the 3rd day, and the treatment was narrowed to Ciprofloxacin. The patient's laboratory values were found to be normal on the 5th day.

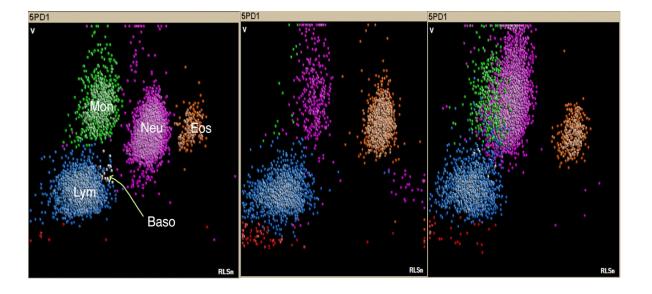


Figure 1. A-Scattergram of a normal patient B-Scattergram of at admission C-Scattergram of after treatment

Table 1: Complete blood count parameters at admission and after treatment

Parameter	At Admission	After Treatment	After Treatment 10th Day	Reference Value	Unit
WBC	1.1	9.5	9.2	3.6-11	10 ⁹ /L
RBC	2.75	3.66	3.77	3.8-5.3	10 ¹² /L
HGB	8.7	11.2	11.5	11.7-16	g /dL
НСТ	24.6	33.1	33.5	35-47	%
MCV	89.6	90.5	88.8	75.3-95.3	fL
МСН	31.7	30.6	30.6	24.3-33.2	pg
MCHC	35.3	33.8	34.5	32-36	g/dL
PLT	9	58	310	150-450	10 ⁹ /L
MPV	8.0	8.7	8.0	7-11.2	fL
PCT	0.05	0.17	0.25	0.1-0.5	mg/dL
PDW	18	18.8	18	8-18	fL
NEU#	0.14	6.58	5.5	1.9-8.2	10 ⁹ /L
LYM#	0.54	1.96	2.69	1.0-3.2	10 ⁹ /L
MON#	0.01	0.47	0.66	0.2-0.9	10 ⁹ /L
EOS#	0.51	0.38	0.32	0-0.5	10 ⁹ /L
BAS#	0.001	0.03	0.04	0-0.1	10 ⁹ /L
NEU%	12.76	68.89	59.77	40-75	%
LYM%	51.30	20.54	29.17	16-45.9	%
MON%	0.56	4.96	7.13	4.5-12.5	%
EOS%	35.35	5.29	3.46	0.5-7.0	%
BAS%	0.03	0.32	0.47	0.2-1.5	%
RDW-CV	14.3	14.9	15.2	12.3-17.7	%
RDW-SD	44.6	46.4	46.4	37-54	fL

Discussion

Hematologic toxicities associated with methotrexate use include, though rarely, anemia, leukopenia, or thrombocytopenia, which may occur without significant reductions in other cell lines, or the more rarely seen condition of pancytopenia may accompany them. To prevent these complications, the American College of Rheumatology (ACR) guidelines recommend monitoring and routine peripheral complete blood counts(11).

When comparing the two-dimensional scattergram of our patient's admission to the hospital (5PD1-RLSn) with the scattergram of a patient with normal complete blood count parameters, we observe that the levels of monocytes and basophils are completely erased (Green-White Dots), the levels of neutrophils are significantly reduced (Pink Dots), and the levels of eosinophils and lymphocytes are increased (Blue-Orange Dots). In the laser scattering method, the laser is directed to the cells passing through the flow system and the low-angle signal represents the cell volume information, the medium-angle and high-angle signal represents the cell nucleus information and the cytoplasm information. The obtained data is displayed in a 2- or 3-dimensional diagram of WBC subgroups. The system creates these images by instantaneously measuring cell volume (Y-axis) and the

light loss calculated according to the axis (X-axis).

The hematological differential parameter changes at the time of the patient's admission indicate that eosinophils have increased both in percentage and cell volume, while neutrophils have an increased cell volume but are significantly reduced in number. The post-treatment scattergram changes are observed in Figure-1C. Although the hemogram diff image is quite similar to that of a healthy patient, neutrophils are still larger in volume. These cell changes suggest that methotrexate affects the bone marrow, creating aberrant lines in the granulocyte series, and likely extends the half-lives of these lines (eosinophil lifespan 8-18 hours), indicating that although the total WBC has normalized, cell-based volume corrections have not yet occurred. Detailed analyses with flow cytometry, examining the clusters of differentiation (CD16, CD49, CD69, etc.) of granulocyte series cell lines, could contribute to explaining the pathophysiology of this effect.

Conclucion

In conclusion, the severity of the pancytopenic state at admission suggests that laboratories performing CBC measurements could enhance accuracy by modifying their procedures in similar cases. We believe that in systemic diseases like rheumatoid arthritis, if disease-modifying antirheumatic drugs (DMARDs) are used, it is important to routinely include scattergram analysis in hemogram follow-ups to detect possible side effects, especially intoxication, at an early stage.

- **1.** Ding Q, Hu W, Wang R, Yang Q, Zhu M, Li M, et al. Signalingpathways in rheumatoidarthritis: implicationsfortargetedtherapy. SignalTransductTargetTher. 2023;8(1). doi: https://doi.org/10.1038/s41392-023-01331-9.
- 2. Up To Date. Methotrexate drug information. Available from: https://www.uptodate.com/contents/methotrexate-drug-information?search=metotreksat&source=panel_search_result&selectedTitle=1~148&usage_type=panel&kp_tab=drug_general&display_rank=1#F194514. Accessed [11.11.2024].
- Emmanuel D, Parija SC, Jain A, Misra DP, Kar R, Negi VS. Persistent eosinophilia in rheumatoid arthritis: a prospective observational study. Rheumatol Int. 2019 Feb;39(2):245-253.
- **4.** Savolainen HA, Leirisalo-Repo M. Eosinophilia as a side-effect of methotrexate in patients with chronic arthritis. Clin Rheumatol. 2001;20:432-4.
- Aristizabal-Alzate A, Nieto-Rios JF, Ocampo-Kohn C, Serna-Higuita LM, Bello-Marquez DC, Zuluaga-Valencia GA. Successful multiple-exchange peritoneal dialysis in a patient

- with severe hematological toxicity by methotrexate: case report and literature review. J BrasNefrol. 2019;41(3):427-432.
- **6.** Lim AY, Gaffney K, Scott DG. Methotrexate-induced pancytopenia: serious and under-reported? Our experience of 25 cases in 5 years. Rheumatology (Oxford) 2005;44:1051-5.
- 7. Cancelliere N, Barranco P, Vidaurrázaga C, Benito DM, Quirce S. Subacute prurigo and eosinophilia in a patient with rheumatoid arthritis receiving infliximab and etanercept. J Investig Allergol ClinImmunol. 2011;21:248-9.
- **8.** Poliak N, Orange JS, Pawel BR, Weiss PF. Eosinophilic fasciitis mimicking angioedema and treatment response to infliximab in a pediatric patient. Ann Allergy Asthma Immunol. 2011;106:444-5.
- **9.** Morrisroe K, Wong M. Drug-induced hypereosinophilia relatedt o tocilizumab therapy for rheumatoid arthritis. Rheumatology (Oxford). 2015;54:2113-4.
- 10. Demirel A, Kirnap M. Romatoid artrit tedavisinde geleneksel ve güncel yaklaşımlar (Traditional and up-to-date treatment in rheumatoid arthritis). J Health Sci. 2010;19(1):74-84. Available from: https://dergipark.org.tr/tr/download/article-file/693023.
- 11. Fraenkel L, Bathon JM, England BR, StClair EW, Arayssi T, Carandang K, et al. 2021 American College of Rheumatology Guideline for the Treatment of Rheumatoid Arthritis. Arthritis Care Res (Hoboken). 2021 Jul;73(7):924-939. doi: 10.1002/acr.24596.