E-ISSN
e-ISSN 2149-9934
Volume: 11 **Issue:** 2 September 2020

Journal of Emergency Medicine Case Reports

EPAT
1999
Emergency Physicians Association of Turkey

Owner and Responsible Manager
Başar Cander
Department of Emergency Medicine, School of Medicine, University of Health Sciences, Kanuni Sultan Süleyman Training and Research Hospital, İstanbul, Turkey

Editors in Chief
Özlem BİLİR
Latif DURAN

Editorial Board
Emine EMEKTAR
Gülşah ÇIKRIKÇI IŞIK
Yunsur ÇEVİK
Hakan OĞUZTÜRK
Ayhan AKÖZ
Hızır AKDEMİR
Mücahit AVCİL
Atıf BAYRAMOĞLU
Arif Onur EDEN
Şahin ÇOLAK
Şükrü GÜRBÜZ
Ali DUMAN
Selçuk Eren ÇANAKÇI

Printing and Graphics Department

PUNTO AJANS

NEF12 Ofis
Maltepe Mah. Çırpıcı Yolu B Çk.
NEF12 B Blok Kat:14 Daire:148
Zeytinburnu / İstanbul / Türkiye
Telefon: 0212 963 09 46

Şişli Ofis
Merkez Mahallesi Hasat Sokak
No: 52 Şişli / İstanbul / Türkiye
Telefon: 0553 199 95 59
info@puntodizgi.com
www.puntoajans.com

Editorial

Dear colleagues,

Nowadays, when we are going through extraordinary conditions, we are in a front-line struggle in a biological war. While the efforts made for the future of the country are being burdened on our shoulders in this difficult time, while advising everyone to stay at a distance we continue to strive to fulfill our profession with the difficulty of serving our patients face to face. The anonymous heroes of the health army, who show sacrifice far from being expressed by words, also continue to produce science and research. Our case report journal which is one of the 4 journals of EPAT, is scanned in many national and international indexes and within the scope of ESCI, continues its publication life despite all difficulties. Emergency medicine has an extremely wide spectrum. Very different cases can be applied almost every day and these interesting cases make important contributions to the medical literature. This prestigious journal, which is very popular in the international arena and is the first in our country in this regard, has come to this day with the important contributions of many scientists. Since this issue, there has been a flag assign and our journal will continue to move forward with a new, young and dynamic editorial team. We would like to thank all our stakeholders who have worked so far, and wish success to our new team.

Prof. Dr. Başar Cander

Değerli Meslektaşlarımız

Olağandışı şartlardan geçtiğimiz bugünlerde adeta biyolojik bir savaşın içinde ön cephede sürekli bir mücadele içindeyiz. Bu zor zamanda ülkenin geleceği için yapılan çabalar omuzlarımıza yüklenirken, herkese mesafeli olmalarını tavsiye ederken biz hastalarımızla burun buruna hizmet vermenin güçlüğüyle mesleğimizi icra etmeye gayret göstermeye devam ediyoruz. Bu kelimelerle ifade edilmekten uzak fedakârlığı gösteren sağlık ordusunun isimsiz kahramanları bir taraftan da bilim üretemeye, araştırma yapmaya devam etmekteler. ATUDER'in sürekli yayın yapan 4 dergisinden biri olan ulusal ve uluslararası birçok indekste taranan ESCI kapsamındaki case report dergimiz de yayın hayatına tüm zorluklara rağmen devam etmektedir. Acil tıp son derece geniş bir spektruma sahiptir. Hemen her gün çok farklı vakalar başvurabilmekte ve bu ilginç vakalar tıp literatürüne önemli katkılar sunmaktadırlar. Uluslararası arenada da çokça rağbet gören ve bu konuda ülkemizde ilk olan bu saygın dergi, birçok bilim insanının önemli katkılarıyla bu günlere gelmiştir. Bu sayımızdan itibaren bir bayrak devri olmuştur ve dergimiz genç dinamik yeni bir editör ekibiyle ileriye doğru yürümeye devam edecektir. Bugüne kadar emek sarf eden tüm paydaşlarımıza teşekkür eder yeni ekibimize başarılar dileriz.

Prof. Dr. Başar Cander

Contents

Case Report

Journal of Emergency Medicine Case Reports

A Story of Extraordinary Abdominally Perforation

Muhammed Ekmekyapar[1], Şükrü Gürbüz[1], Serdar Derya[1], Hakan Oğuztürk[1], Neslihan Yücel[1], Muhammet Gökhan Turtay[1]
Abdullah Ercan[1], İsmail Okan Yıldırım[2]

[1]Department of Emergency Medicine, Inonu University, Malatya, Turkey
[2]Department of Radiology, Inonu University, Malatya, Turkey

Abstract

Pneumoperitoneum is the result of a gastrointestinal (GI) tract perforation in more than 90% of cases. Perforation of the stomach or duodenum caused by peptic ulcer is considered the most common cause of pneumoperitoneum. In this article, we present a case of pneumoperitoneum associated with sigmoid colon perforation, which is the result of the patient trying to create a laxative effect by inserting the hose attached to the wash basin into his rectum and opening the water afterwards. An 89 years old male patient was brought to the emergency department with complaints of abdominal pain starting about 2 hours ago. The patient's abdominal ultrasonography showed free fluid up to 10 cm in the lower abdomen. Posterior anterior chest X-ray and direct standing abdominal X-ray of the patient had subdiaphragmatic free air and images with free fluid, pneumoperitoneum and perforation were obtained in contrast enhanced abdominal tomography. As a result of surgery performed by general surgery it was seen approximately 30 cm proximal from the rectumthat there was a 4 cm perforation in the sigmoid colon. We must consider pneumoperitoneum, which is the result of colon perforation especially in elderly patients with acute abdominal pain, to be definitely among our differential diagnoses.

Key Words: Sigmoid colon perforation, pneumoperitoneum, acute abdomen

Introduction

More than 90 percent of pneumoperitoneum cases are the result of gastrointestinal perforations[1]. Perforation of the stomach or the duodenum due to peptic ulcer is admitted as the most common cause of pneumoperitoneum. It also occurs because of abdominal trauma or diverticular perforation[1]. The most common radiological finding in a patient with widespread peritonitis is free air under the diaphragm on chest X-ray. Most situations require immediate exploration and intervention. Iatrogenic colorectal perforations have frequently taken place in the surgical literature, due to occurring during colonoscopy and barium enema examinations[5]. Rarely, pneumatic injuries caused by the jokes made with compressed air sources has also been reported[6]. Rarely, pneumatic colon injuries caused by the jokes made for the fun using compressed air sources has also been reported[6].

Case report

An 89-year-old male patient was brought to the emergency room with the complaint of abdominal pain which started about 2 hours ago. According to the medical history taken from patient and his relatives, the patient has complained about chronic constipation for many years. When we detailed the medical history, we learned that the patient has complained about constipation for a long time, he has went to the toilet, attached the hose to the faucet, put the hose into his anus and opened the faucet to get the feces output by the help of the water. The patient said that he had done the same process for the abdominal pain began 2 hours ago. There are pain and tenderness in the abdominal examination of the patient. The patient's vital parameters including blood pressure, pulse, respiratory rate and fever were respectively measured as 90/52 mmHg, 74 / min, 20 / min fever and 36.4° C. In the lab tests of the patient, the hemogram parameters were like that: White blood cell (WBC): 11x10^9/ L, hemoglobin (HB): 14.5 g/dL, hematocrit (HTC): 42% and PLT: 427x10^9/L. The biochemical parameters were like that: glucose:113 mg/dL, sodium: 129 mmol/L, calcium: 8 mg/dL, total bilirubin: 1.52 mg/dL, direct bilirubin: 0.58 mg/d L and albumin: 2.8 g/ dL. C-reactive protein (CRP) was 0,401 mg/dL (normal range is 0 - 0.35 mg/ dL). Other parameters were normal. The patient's abdominal ultrasonography showed that there was approximately 10 centimeter free liquid in the lower abdomen. There was a diaphragmatic free air in the patient's chest and abdomen X-ray (Figure-1) and contrast-enhanced abdominal tomography showed images compatible with intraabdominal free fluid, pneumoperito-

Corresponding Author: Şükrü Gürbüz **e-mail:** sukrugurbuz@gmail.com
Received: 17.01.2019 • **Accepted:** 17.01.2020
DOI:10.33706/jemcr.514083

DOI:10.33706/jemcr.514083
Journal of Emergency Medicine Case Reports 2020; 11 (1):1-2

neum and perforation (Figure-2).The patient was consulted general surgery department and the he was undergone emergency surgery with the pre-diagnosis of pneumoperitoneum and perforation by general surgeons.

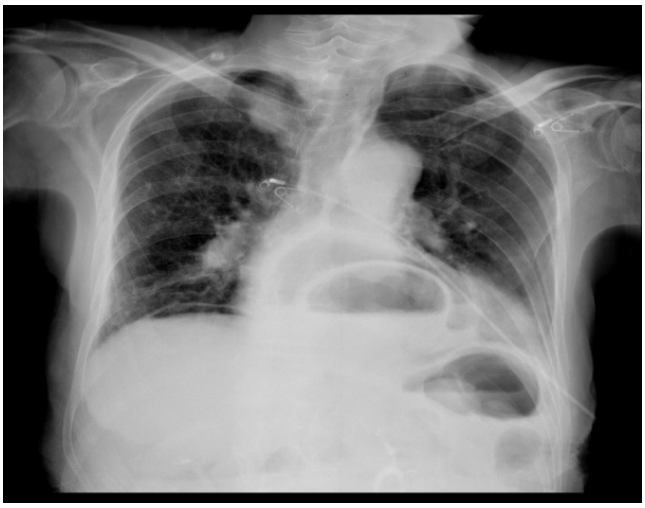

Figure 1: Free air under diaphragm on the chest X-ray

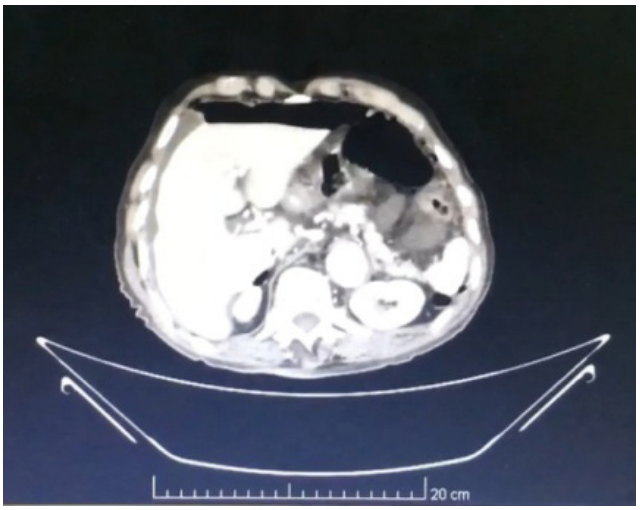

Figure 2 : Free fluid and air in the abdomen on the contrast-enhanced abdominal tomography

Discussion

More than 90 percent of pneumoperitoneum cases are the result of gastrointestinal perforations[1]. Perforation of the stomach or the duodenum due to peptic ulcer is admitted as the most common cause of pneumoperitoneum. It also occurs because of abdominal trauma or diverticular perforation[1]. Complaints of constipation and chronic laxative use increase with age, and at the same time, there is an increase in the frequency of enema applied in emergency services, in nursing homes or at home by the patient him- or herself[2-3]. Rectal tubes or similar instruments with varying length, diameter and stiffness are used for enema administrations performed with water or laxatives. The incidence of rectum and sigmoid colon perforations is increasing due to enema administrations performed by such tools[4]. Paran et al reported 13 patients who consulted their clinic within 3 years for colorectal perforation due to enema in the study conducted in Israel. Ten of them were elderly patients with chronic constipation who were staying at nursing homes and the enema was administered by nursing staff. The other 3 patients administered the enema him- or herself at home. Considering the elderly patients with chronic constipation in our country, pneumoperitoneum cases which are secondary to colonic perforation caused by enema administrations should be drawn attention and further studies are needed. To inform patients relatives or caregivers who are taking care of them about possible injuries will also be useful.

Conclusion

A 4 cm sigmoid colon perforation which is extending 30 cm proximal from rectum was found in the operation performed by general surgeons. Pneumoperitoneum, which is the result of colonic perforation, has been found to show a mortal course especially in elderly patients, and pneumoperitoneum, which is caused by the perforation of the sigmoid colon after a traumatic procedure performed by the patient in our case, is worthy of presentation.

References

1. Mularski RA, Ciccolo ML, Rappaport WD. Nonsurgical causes of pneumoperitoneum.West J Med 1999; 170: 41-46.
2. Harari D, Gurwitz JH, Avorn J, et al. Constipation: assessment and management in an institutionalized elderly population. J Am Geriatr Soc. 1994; 42: 947-952
3. Harari D, Gurwitz JH, Minaker KL. Constipation in the elderly. J Am Geriatr Soc. 1993; 41: 1130-40
4. Paran H, Butnaru G, Neufeld D, et al. Enema-induced perforation of the rectum in chronically constipated patients. Dis Colon Rectum 1999; 42: 1609-1612
5. Gedebou TM, Wong RA, Rappaport WD, et al. Clinical presentation and management of iatrogenic colonperforations. Am J Surg 1996; 172: 454-458
6. Suh HH, Kim YJ, Kim SK. Colorectal injury by compressed air - a report of two cases. J Korean Med Sci1996; 11: 179-82.

Case Report

Journal of Emergency Medicine Case Reports

Carotid Artery Dissection: A Case Report

Muhammed Ekmekyapar[1], Hakan Oğuztürk[1], Tuba Ekmekyapar[2], Serdar Derya[1], Şükrü Gürbüz[1]

[1]Emergency Medicine Department, Faculty of Medicine, Inonu University, Malatya, Turkey
[2]Neurology Department, Malatya Education and Research Hospital, Malatya, Turkey

Abstract

Carotid artery dissection, firstly described by Pratt-Thomas and Berger in 1947, typically begins in inner layer of artery wall, proceeds to the middle layer, and intramurally extends along length of the artery as a result of the pressure produced by blood stream. This study aims to report a case in which internal carotid artery dissection was diagnosed as secondary to trauma. A 24-year-old male patient was brought to the emergency room by 112 emergency service team after a motorcycle accident. Physical examination of the patient revealed a painful dermabrasion in his right neck region and other system examinations were normal. Carotid-vertebral color Doppler ultrasonography was performed on the patient because he had a right neck pain. Accordingly, an intimal flap appearance compatible with dissection was observed on the right internal carotid artery (ICA) proximal segment. Then, brain+cervical CT angiography was performed on the patient, and an appearance compatible with dissection was observed in the right ICA. Therefore, the patient was referred to neurology and neurosurgery consultation and accordingly admitted to neurosurgery intensive care unit. As a result, carotid artery dissection in addition to other intracranial pathologies should be considered among differential diagnoses for patients with head and/or neck pain complaints regardless of whether or not they have a trauma history.

Key Words: Carotid artery, trauma, dissection

Introduction

Carotid artery dissection was described for the first time by Pratt-Thomas and Berger (1947) after making autopsies of two death patients[1]. Arterial dissection typically begins in inner layer of artery wall, proceeds to the middle layer, and intramurally extends along length of the artery as a result of the pressure produced by blood stream. The most common type of dissection is external carotid artery dissection. The clinical condition may vary from mild symptoms (ipsilateral head and neck pain, Horner's syndrome) to more severe findings such as transient ischemic attack and cerebral ischemia. The causes of carotid artery dissection include rheumatic diseases, hypertension, migraine, fibromuscular dysplasia, and trauma[2]. This study aims to report a case admitted to emergency service after a trauma and diagnosed of an internal carotid artery dissection.

Case

A 24-year-old male patient was brought to the emergency room by 112 emergency service team after a motorcycle accident. Physical examination of the patient revealed a painful dermabrasion in his right neck region and other system examinations were normal. The patient's vital parameters were as follows; fever 36°C, pulse 75/min, TA 125/75 mmHg, and respiratory rate 18/min. The patient's hemogram and biochemistry values were normal. After the physical examination, the patient's radiological examinations were requested. There was no abnormal finding on his tomography, graphy and abdominal ultrasonography. Carotid-vertebral color Doppler ultrasonography was performed on the patient because he had a right neck pain. Accordingly, an intimal flap appearance compatible with dissection was observed on the right internal carotid artery (ICA) proximal segment. Then, brain+cervical CT angiography was performed on the patient, and an appearance compatible with dissection was observed in the right ICA (Figure-1). Therefore, the patient was referred to neurology and neurosurgery consultation and accordingly admitted to neurosurgery intensive care unit.

Discussion

An arterial dissection occurs as a result of the rupture of one of vein wall layers, usually the intima layer. The annual incidence of spontaneous carotid artery dissection in West-

Corresponding Author: Şükrü Gürbüz **e-mail:** sukrugurbuz@gmail.com
Received: 17.01.2019 • **Accepted:** 05.03.2020
DOI:10.33706/jemcr.514089

DOI:10.33706/jemcr.514089
Journal of Emergency Medicine Case Reports 2020; 11 (1):3-6

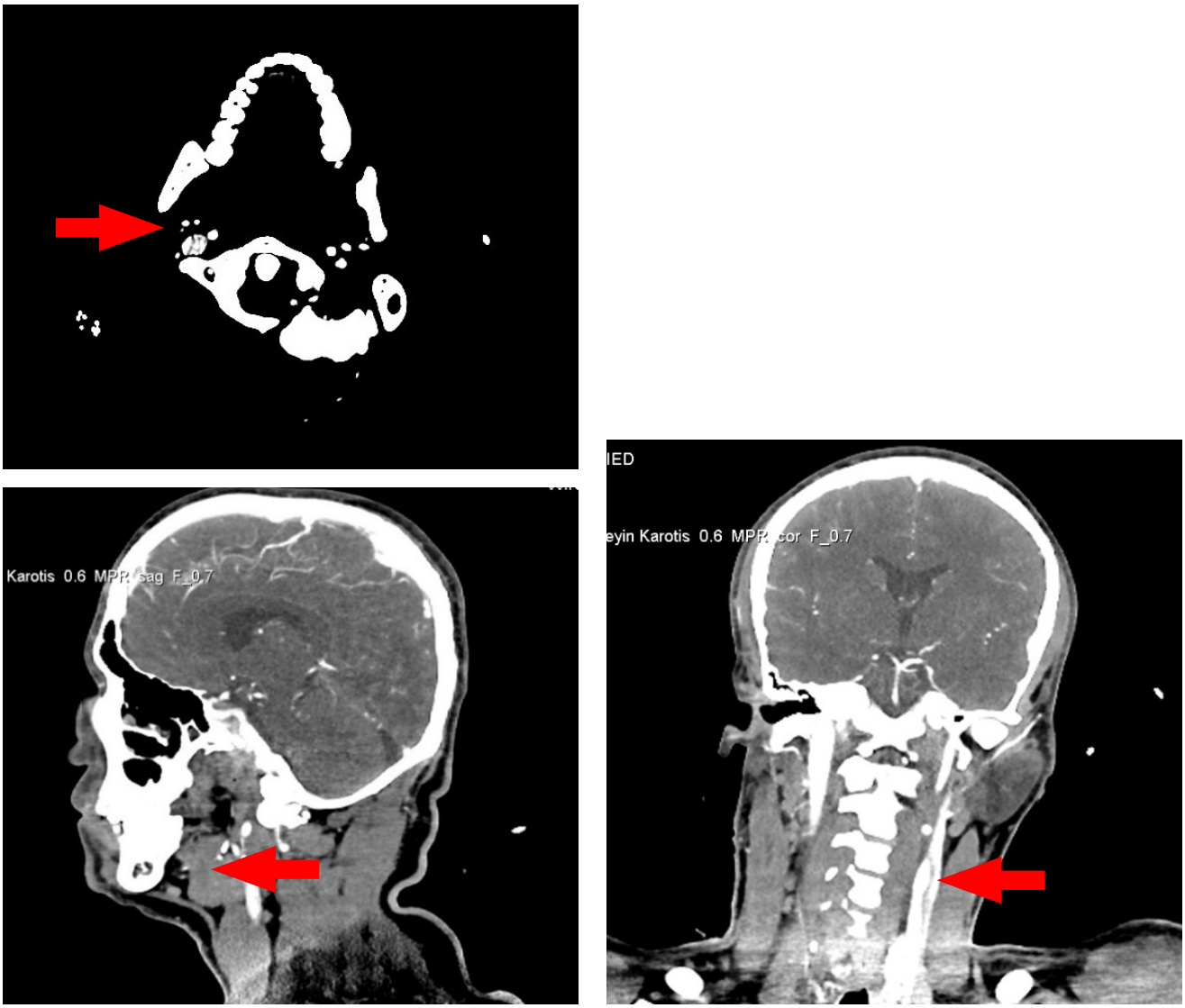

Figure 1: An appearance compatible with dissection in the patient's brain+cervical CT angiography

ern countries ranges from 2.5 to 3 per 100,000 individuals[3]. Spontaneous carotid artery dissection is an important cause of ischemic insemination in young adults and accounts for 10-25% of these cases[4]. It has a variety of local and clinical symptoms. The most common local symptoms of carotid dissection are acute head, face, or neck pains. The present study case had a dermabrasion in his neck region, but no additional complaints other than neck pain. Unilateral stroke and cerebral ischemic symptoms such as amaurosis fugax have been reported in 50-90% of the patients with arterial dissection[5]. The causes of carotid artery dissection can be classified as trauma including direct damage to the head and neck; underlying arteriopathy such as Marfan's syndrome; and spontaneous occurrence. Detecting the true frequency of traumatic ICA (internal carotid artery) dissection is difficult because of the absence of typical findings and sometimes asymptomatic course. Besides blunt trauma, artery stretch and exposure to hyperextension also cause intimal rupture. Various methods are used to diagnose carotid artery dissection. Firstly, computerized tomography is used to demonstrate the presence of cerebral hemorrhage, infarct, or cerebral edema, and also it is useful in determining whether there is any contraindication to the use of systemic anticoagulants. Doppler ultrasound is useful in diagnosing extracranial carotid artery dissections[6]. MR angiography is a minimally invasive diagnostic method with high sensitivity and specificity. Despite all, no matter how helpful these diagnostic methods are, arteriography is the gold standard in diagnosing of carotid artery dissections[7,8]. Regarding its treatment, if there is no contraindication in the acute phase, anticoagulation therapy is recommended for the risk of thrombosis. Surgical treatment or stenting may be considered as other treatment options in cases of an ongoing ischemic event despite anticoagulation treatment or in cases where anticoagulation therapy is contraindicated.

Conclusion

As a result, carotid artery dissection in addition to other intracranial pathologies should be considered among differential diagnoses for patients with head and/or neck pain complaints regardless of whether or not they have a trauma history.

References

1. Pratt-Thomas HR, Berger KE. Cerebellar and spinal injuries after chiropractic manipulation. JAMA. 1947;133:600–603.
2. Şengül G, Kadıoğlu HH, Aydın IH. Arteriyel diseksiyon sendromları. Turkiye Klinikleri J Surg Med Sci. 2006; 2: 163-5.
3. Schievink WI, Mokri B, Whisnant JP. Internal carotid artery dissection in a community Rochester, Minnesota, 1987-1992. Stroke 1993;24: 1678–80.
4. Schievink WI. Spontaneous dissection of the carotid and vertebral arteries. N Engl J Med 2001;344:898–906.
5. Arnold M, Bousser MG. Carotid and vertebral artery dissection. Pract Neurol 2005;5:100–9.
6. Srinivasan J, Newell DW, Sturzenegger M, Mayberg MR, Winn HR. Transcranial Doppler in the evaluation of internal carotid artery dissection. Stroke 1996 ;27(7):1226-30.
7. Desfontaines P, Despland PA. Dissection of the internal carotid artery: aetiology, symptomatology, clinical and neurosonological followup, and treatment in 60 consecutive cases. Acta Neurol Belg 1995;95(4):226-34.
8. Biousse V, D'Anglejan-Chatillon J, Touboul PJ, Amarenco P, Bousser MG. Time course of symptoms in extracranial carotid artery dissections. A series of 80 patients. Stroke 1995;26(2):235-9.

Case Report

Journal of Emergency Medicine Case Reports

An Uncommon Hyperosmolar Nonketotic Condition

Elif Çelikel[1], Uğur Bilgay Kaya[1], Birsen Uluca Kaya[2], Hasan Doğan[2], Afsin Emre Kayipmaz[1]

[1]Department of Emergency Medicine, Ankara City Hospital, Ankara, Turkey
[2]Department of Neurology, Ankara City Hospital, Ankara, Turkey

Abstract

Introduction: Hyperglycemia may present with neurological symptoms. This rare condition is called non-ketotic hemichorea-hemiballismus (HCHB). When investigating the causes of neurological symptoms in these cases, magnetic resonance imaging (MRI) imaging typically has a hyperintense appearance in T1 sections and a variable intense appearance in T2.

Case report: A 68-year-old female patient was admitted to the emergency department with speech disorder. Blood sugar elevation and cranial tomography revealed hyperdense appearance in the left caudate nucleus and lentiform nucleus. The patient was hospitalized with a preliminary diagnosis of HCHB. Magnetic resonance in hyperintense T1W sequences in the nucleus caudatus hood and corpus on the left, putamen, globus pallidus, Significant hypointense signal changes were observed in T2W sequences. The patient was admitted to the hospital for treatment and his blood sugar regulation was restored.

Conclusion: HCHB is a rare condition in emergency. After the regulation of blood sugar, neurological picture regression is observed. Blood sugar levels should be checked and appropriate treatment should be given in patients presenting with neurological symptoms.**Keywords:** Hyperglycemia, chorea, hemiballismus.

Introduction

The hyperosmolar hyperglycemic state (HHS) is the most serious acute hyperglycemic emergency. It is mostly seen within type 2 diabetes. It is a syndrome that presence hyperglycemia, hyperosmolarity, and dehydration in the absence of ketoacidosis. The mortality rate is high. It is a life-threating emergency. Patients may present polydipsia, polyuria, nausea, vomiting, lethargy. A wide variety of acute focal and global neurologic changes may be present delirium, coma, hemiparesis, and sensory deficits[1-2]. Hyperglycemia has been reported to cause intracranial pathology with intracranial acidosis, extracellular glutamate accumulation, brain edema, blood-brain barrier impairment[3]. The appearance of choreiform movements with hyperglycemia is called hyperglycemic nonketotic hemichorea-hemiballismus (HCHB). Especially in elderly, Asian, female patients, irregular, involuntary, unilateral abnormal movements accompanied by high blood glucose levels were observed[4-5-6]. In this study, we aimed to present a patient with speech disorder who accompanied hyperglycemic nonketotic conditions without loss of consciousness and choreiform movements.

Case report

A 68-year-old female patient presented with an urgent speech impairment. She answered only the questions asked, "I am fine." She had a history of hypertension, coronary artery disease, and diabetes. Light reflex was bilateral positive, pupils were isochoric, facial asymmetry and motor deficits were not present, deep tendon reflexes were bilateral flexor.

Her blood glucose level was 533 mg/dl, blood pressure was 150/89 mmHg, pulse rate was 78/minute, respiratory rate was 14/minute, fever was 36°C. Hemoglobin 12.8 g/dl, leukocyte count 4.4 thousand/µl, platelet count 132 thousand/µl, aspartate aminotransferase (AST) 15 (U/L), Alanine aminotransferase (ALT) 9 (U/L), urea 22 (mg/dL), creatinine 0.74 (mg/dL), blood glucose level 553 mg/dl, serum osmolarity 304 mOsm/kg, pH 7.45 and urine ketone was negative. Cranial tomography (CT) revealed hyperdense appearance in the left caudate nucleus and lentiform nucleus. In the emergency follow-up and treatment of the patient, his blood glucose level decreased to 273 mg / dL, and his control CT showed a marked decrease in hyperdensity of the left caudate nucleus and lentiform nucleus (Figure 1). The patient was admitted to the internal medicine department for further examination. During the follow-up in this department, the patient had status epilepticus seizure and she was intubated and taken to the intensive care unit. Midazolam and phenytoin were started for seizures. The patient was found to have a high fever during the follow-up in the intensive care unit. Cranial magnetic resonance imaging (MRI) performed at the time of hospitalization revealed hyperintense

Corresponding Author: Elif Çelikel **e-mail:** drelifkaya@gmail.com
Received: 02.10.2019 • **Accepted:** 17.01.2020
DOI:10.33706/jemcr.592152

DOI:10.33706/jemcr.592152
Journal of Emergency Medicine Case Reports 2020; 11 (1):7-8

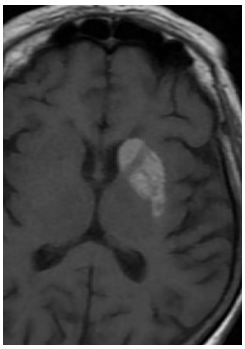

Figure 1

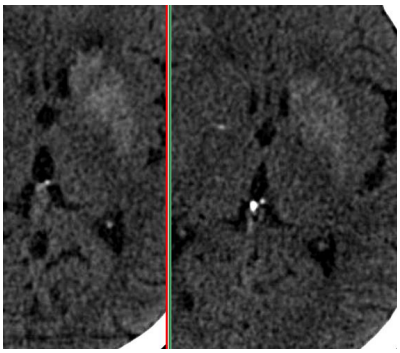

Figure 2

T1W sequences in the nucleus caudatus hood and corpus on the left, putamen, globus pallidus (Figure 2). Significant hypointense signal changes were observed in T2W sequences. The hyperintense areas identified in the GE sequence were not compatible with blood. Radiological findings of the patient with a pre-diagnosis of HCHB were interpreted as supporting the pre-diagnosis. The control MRI taken 10 days later revealed similar findings to the previous one. Electroencephalography showed a slight diffuse deceleration of bioelectric activity. The patient was extubated on the 5th day of intubation and followed up in the intensive care unit and then transferred to the ward. The patient whose general condition improved during the follow-up visits was discharged.

Discussion

HCHB is a rare condition[7]. It is usually unilateral involuntary, continuous and irregular movements, sometimes mimicking central nervous system diseases such as consciousness disorder. The mechanism is not clear. It has been reported that hyperglycemia changes the metabolic activity in the brain by causing hypoperfusion and abnormal movements are the result of this condition[4]. Basal ganglia have metabolically high activity and are affected by metabolic diseases, toxic substances, and neurodegenerative diseases[5]. MRI findings may also be seen due to calcium accumulation in neurons[8]. In MRI, hyperintense appearance in basal ganglia in T1 sections is attributed to secondary swelling of reactive astrocytes to hyperglycemia[9]. Hyperglycemia is associated with an improvement in these cases when treated with hyperglycemia. In these cases, cranial tomography shows hyperdense appearance in the basal ganglia, putamen and/or caudate nucleus on the opposite side of the lesion[9]. MRI typically has a hyperintense appearance in T1 sections and a variable intense appearance in T2[3-6]. It has been shown that petechial hemorrhages or calcifications may also cause these radiological features[7-8]. In the case reported by Hansford et al., it was reported that classical images were detected on CT and MRI without typical movements[10]. In our case, typical lesions were detected on CT and MRI without choreiform movements. These radiological findings may be confused with a stroke[11]. Measuring blood glucose levels in these patients is life-saving. HCHB, which can be seen in a wide range of symptoms, has a good prognosis with appropriate treatment[5]. In patients with known diabetes, irregular blood glucose level regulation, or newly detected high blood sugar levels, the emergency physician should consider the diagnosis of HCHB in abnormal choreiform movements.

References

1. Fayfman M, Pasquel FJ, Umpierrez GE. Management of Hyperglycemic Crises: Diabetic Ketoacidosis and hyperglycemic hyperosmolar state. Med Clin North Am. 2017 May;101 (3); 587-606
2. Pasquel FJ, Umpierrez GE. Hyperosmolar hyperglycemic state: A historic review of the clinical presentation, diagnosis, and treatment. Diabetes care 2014;37:31-24.
3. Hedge AN, Mohan S, Lath N, Lim T. Differential Diagnosis for Bilateral Abnormalities of the Basal Ganglia and Thalamus. RadioGraphics 2011;31:5-30. https://doi.org/10.1148/rg.311105041
4. Abd-Samat AH, Embong H. Hemichorea hemiballismus in a patient with hyperglycemic hyperosmolar state, a reversible neurological outcome of metabolic disorder. Hong Kong J. Emerg. Med.2017;24:93-95.
5. Herath HMMTB, Pahalagamage SP, Senanayake S. Case report of hyperglycemic nonketotic chorea with rapid radiological resolution. BMC Medical Imaging. 2017;17:54 https://doi.org/10.1186/s12880-017-0228-2
6. Cho HS, Hong CT, Chan L. Hemichorea after hyperglycemia correction. A case report and a short review of hyperglycemia-related hemichorea at the euglycemic state. Medicine 2018;97:10. DOİ:10.1097/MD:0000000000010076
7. Chu K, Kang DW, Kim DE, Park SH, Roh JK. Diffusion-Weighted and Gradient Echo Magnetic Resonance Finding of Hemichorea-Hemiballismus Associated With Diabetic Hyperglycemia. Hyperviscosity Syndrome?. ArchNeurol. 2002;59(3):448-452. Doi:10.1001/archneur.59.3.448
8. Yacoub HA. Abnormal Magnetic Resonance İmaging and Hemichorea Associated With Non-Ketotic Hyperglycemia. J NeurolRes. 2013;3(5):146-149. Doi:doi.org/10.402/jnr233e
9. Roy U, Das SK, Mukherjee A, Biswas D, Pan K, Biswas A, et al. Irreversible Hemichorea-Hemiballism İn a case of Nonketotic Hyperglycemia Presenting as the İnitial Manifestation of Diabetes Mellitus. Tremor Other Hyperkinet Mov. 2016;6. Doi:10.7916/D8QZ2B3F.
10. Hansford BG, Albert D, Yang E. Classic Neuroimaging findings of non-ketotic hyperglycemia on computed tomography and magnetic resonance imaging with absence of typical movement disorder symptoms (hemichorea-hemiballism). Radiology Case. 2013 Aug;7(8):1-9.Doi:10.3941/jrcr.v7i8.1470
11. Wintermark M, Fischbein NJ, Mukherjee P, Yuh LE, Dillon WP. Unilateral Putaminal CT, MR, and Diffusion Abnormalities Secondary to Nonketotic Hyperglycemia in the Setting of Acute Neurologic Symptoms Mimicking Stroke. AJNR Am J Neuroradiol. 2004;25(6):975-6.

Case Report

Journal of Emergency Medicine Case Reports

An Uncommon Complication: Hypoglossal Nerve Palsy After Carotid Endarterectomy

Ozgul Ocak[1], Hamit Celik[2], Behcet Varıslı[3]

[1]Department of Neurology, Canakkale Onsekiz Mart University, Canakkale, Turkey
[2]Department of Neurology, Buhara Hospital, Erzurum,Turkey
[3]Department of Emergency Medicine, Canakkale State Hospital, Canakkale, Turkey

Abstract

Carotid artery stenosis is the important causes of ischemic stroke, and it may result in death. Many complications can occur during carotid endarterectomy (CEA), which is the standard revascularization procedure for treating severe carotid artery stenosis. CEA is applied for prevention of stroke occurred by atherosclerotic plaque which is located at the common carotid artery bifurcation and, commonly, internal carotid artery. In addition to serious complications, such as stroke, infection, myocardial infarction, postoperative bleeding and death, non-fatal complications, such as cranial nerve palsy, can also occur rarely. Cranial nerve injury is a well-recognized complication' of carotid endarterectomy. Although major injuries are rare, in several series where thorough pre and postoperative neurologic examinations were performed, the incidence of cranial nerve dysfunction after carotid surgery ranged from 9.7% to 39% Here we present the an 80-year-old male patient with hypoglossal nerve palsy after carotid endarterectomy as it is a rare complication.

Keywords: hypoglossal nerve, palsy, carotid endarterectomy, complication

Introduction

Stroke is known as the third most commonly occurring death causality in the world and may result in long-term loss of labor especially in developed countries[1,2]. In total, 20%–25% of ischemic strokes are cause by atherosclerosis occurring in the carotid artery. Atherosclerotic plaque formation in the carotid artery is a pathological process that increases with age. Atheroma plaque usually begins in the posterior wall of the common carotid artery and progresses toward the internal carotid artery. An atheroma plaque is formed by endothelial damage and takes place in the intima. As the plaque increases, the internal carotid artery lumen is obliterated. The standard revascularization method for severe carotid artery stenosis is carotid endarterectomy, and cranial nerve palsy after endarterectomy has been reported in 3%–23% of cases[3].

The most common cranial nerve palsy occurring after carotid endarterectomy is the hypoglossal nerve palsy. It is thought to be the result of nerve retraction during surgery[4].

PURPOSE: We presented this case to indicate the rare occurrence of isolated hypoglossal nerve palsy after carotid endarterectomy.

Case Report

An 80-year-old male patient was admitted to the emergency department with a complaint of loss of power in the right arm and leg. The patient's medical history was remarkable for hypertension, coronary artery disease, and bypass surgery. Physical examination revealed arterial blood pressure of 135/80 mmHg; his pulse rate was 88 beats/min with a regular rhythm and body temperature was 36.6°C. Neurological examination revealed the following pathological findings: right central facial paralysis, right 4/5 hemiparesis, and extensor plantar response on the right side.

Cranial computed tomography performed in the emergency department revealed no acute hemorrhagic lesion. Diffusion magnetic resonance imaging (MRI) performed with the preliminary diagnosis of acute ischemic cerebrovascular disease revealed no diffusion restriction in the sections obtained from the infratentorial region that would suggest acute infarction. In the sections taken from the supratentorial region, acute infarct areas that were hyperintense on diffusion MRI examination and hypointense on apparent diffusion coefficient mapping were observed in the left parietal and occipital regions. The patient was referred to the neurology department

Corresponding Author: Özgül Ocak **e-mail:** dr_ozgul@hotmail.com
Received: 17.06.2019 • **Accepted:** 05.03.2020
DOI:10.33706/jemcr.573739

DOI:10.33706/jemcr.573739
Journal of Emergency Medicine Case Reports 2020; 11 (1):9-12

with the diagnosis of acute ischemic cerebrovascular disease. Carotid artery color Doppler ultrasonography revealed mixed-type plaque formation in the left internal carotid artery resulting in a stenosis of >70%. Routine biochemistry, hemogram, and hormone tests revealed no pathological findings, except a glucose level of 163.8 mg/dl. Anti-aggregant therapy was initiated with acetylsalicylic acid at 100 mg/day, and anti-hypertensive therapy was initiated with candesartan/hydrochlorothiazide at 16/12.5 mg/day. Consultation with a cardiovascular surgeon was performed for treating existing stenosis in the carotid artery. Endarterectomy was performed on the left carotid artery by a cardiovascular surgeon, and the patient was discharged.

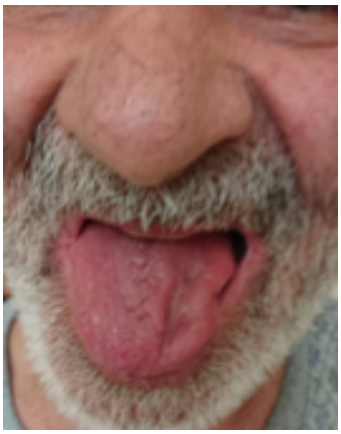

Figure 1. The patients tongue with difficulty in movement, swallowing and talk due to Isolated left N hypoglossus paralysis

After discharge, the patient presented to our outpatient clinic with complaints of restraint in tongue movements and swallowing difficulty in addition to his pre-existing neurological complaints (Fig. 1). In Cranial Magnetic Resonance (MR) images; Axial T2A and T1 weighted (Fig. 2) image had a normal brainstem and fluid attenuated inversion recovery (FLAIR) and T2-weighted image (Fig. 3) images left posterior circulation infarction.

Neurological examination revealed 4/5 hemiparesis in the right upper and lower extremities, extensor plantar response on the right side, atrophy in the left half of the tongue, and deviation of the tongue to the left side on protrusion. The patient was hospitalized in the neurology department with the diagnosis of hypoglossal nerve palsy. The patient was given physical therapy, and total parenteral nutrition was initiated because of swallowing and feeding difficulty. The ongoing anti-aggregant and anti-hypertensive therapies were continued. During the follow-up of the patient, it was observed that the severity of the existing tongue atrophy and swallowing difficulty decreased. The patient was discharged from the hospital on the 10th day of hospitalization with a recommendation of follow-up at an outpatient clinic.

Discussion

Stroke is the third most common cause of death in the world and the most common cause of fatal neurological events. Risk factors for ischemic stroke are divided into two groups: alterable and inalterable. Alterable risk factors include hyperlipidemia, hypertension, smoking, diabetes mellitus, alcohol, obesity, physical inactivity, heart disease, high homocysteine levels, carotid stenosis, presence of anti-cardiolipin antibodies, and the use of oral contraceptives, whereas unalterable risk factors include race, age, genetics, and gender.

Obstructive stroke is observed in approximately 85% patients who are hospitalized and treated for stroke, and ca-

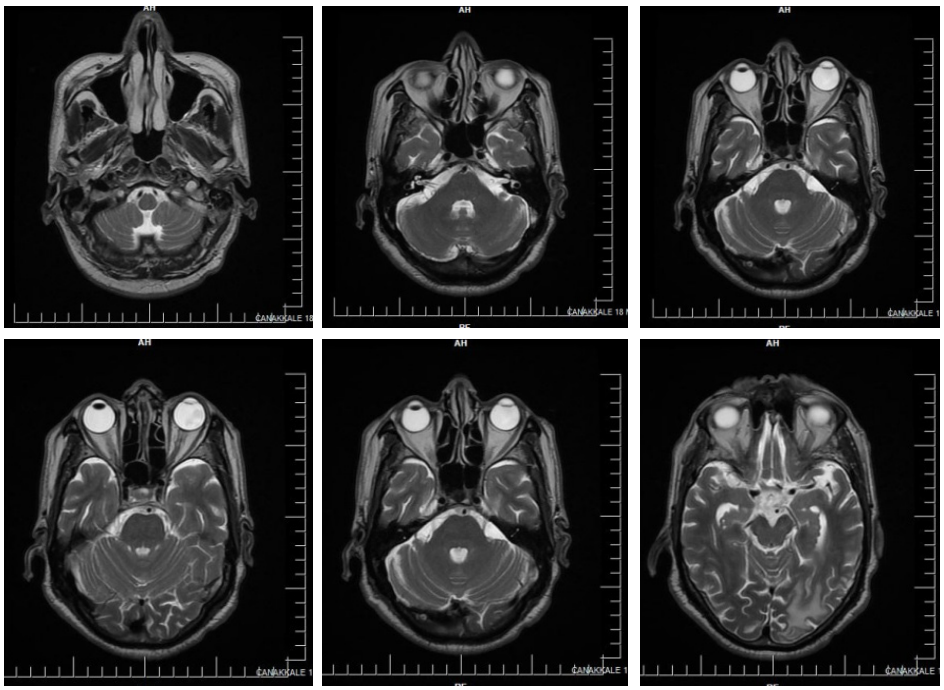

Figure 2. Brain magnetic resonance imaging (MRI) scan T2-weighted image of normal brainstem

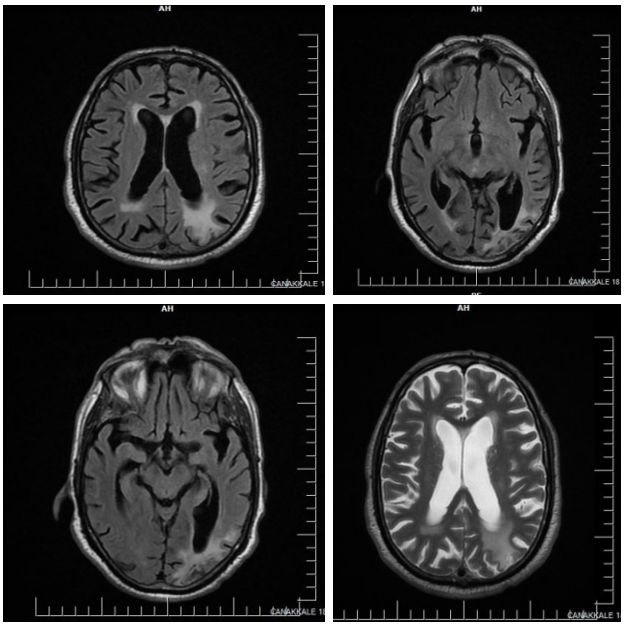

Figure 3. Fluid attenuated inversion recovery (FLAIR) and T2-weighted image images left posterior circulation infarction

rotid stenosis is responsible for 5%–12% of these attacks[5]. Carotid artery atherosclerosis is a systemic vascular disease due to possible complications of stroke and requires a multidisciplinary treatment. Carotid artery endarterectomy is currently accepted as the most effective method for preventing the development of stroke and transient ischemic attacks in patients with symptomatic/asymptomatic carotid artery stenosis[6]. Atherosclerotic plaques are most densely seen on the posterior wall of the carotid bifurcation in the internal carotid artery. Stroke occurs because of occlusion of the arteries by an embolus that detaches from the thrombus formed on the plaque or because of thrombosis of the atherosclerotic plaque resulting in occlusion of the vessel and decrease in brain perfusion[7].

The incidence of cranial nerve palsy after carotid endarterectomy has been reported to be between 3% and 23%[3]. The most common cranial nerve palsy after carotid endarterectomy is hypoglossal nerve palsy. The hypoglossal nerve is a pure motor cranial nerve innervating the extrinsic and intrinsic muscles of the tongue. The hypoglossal nerve follows a vertical course in the neck after leaving the anterior condylar foramen and it is in close proximity with the internal carotid artery and internal jugular vein. This proximity is also the cause of nerve palsy after surgeries performed on cervical veins[7]. Hypoglossal nerve palsy occurring after carotid endarterectomy is thought to result from nerve damage secondary to nerve retraction during surgery.

In our patient, hypoglossal nerve palsy was detected after carotid endarterectomy. This was probably due to axonal injury as a result of overstretching of the nerve while attempting to preserve the hypoglossal nerve during carotid artery exploration.

Conclusion

Isolated hypoglossal nerve palsy, which is a rare complication occurring after carotid endarterectomy, should be kept in mind, and careful neurological examination should be performed after the operation to avoid this complication from being unnoticed.

References

1. Liapis CD, Bell PR, Mikhailidis D, Sivenius J, Nicolaides A, Fernandes e Fernandes J, Biasi G, Norgren L. ESVS guidelines. Invasive treatment for carotidstenosis: indications, techniques. Eur J Vasc Endovasc Surg. 2009;37:1-19.
2. Thom T, Haase N, Rosamond W, Howard VJ, Rumsfeld J, Manolio T, et al. Heart disease and stroke statistics 2006 update: a report from the American Heart Association Statistics Committee and Stroke Statistics Subcommittee. Circulation 2006;113(6):e85-e151.
3. Cunningham EJ, Bond R, Mayberg MR, Warlow CP, Rothwell PM. Risk of persistent cranial nerve injury after carotid endarterectomy. J Neurosurg 2004;101(3):445-8.
4. Lusby RJ, Wylie EJ. Complications of carotid endarterectomy. Surgical Clinics of North America 1983;63:1293.
5. Bates ER, Babb JD, Casey DE et al. Consensus document on carotid stending: a report of the American Society of College of Cardiology FoundationTask Force on clinical expert consensus document. J Am Coll Cardiol 2007;49:126-170.
6. European Carotid Surgery Trialist's Collaborative Group MRC European carotid surgery trial: interim results for symptomatic patients with severe (70-99%) or with mild (0-29%) carotid stenosis. Lancet 1991;337:1235-1243.
7. De Borst GJ. Cranial nerve palsy should not be included within a primary composite end point in carotid surgery trials. Eur J Vasc Endovasc Surg 2014;47:583-4.
8. Bradley WG, Darof RB, Fenichel GM, Jankovic J. Neurology in clinical practice Vol-2, 4th Edition. Cranial neuropathies. Butter worth Heineman, Philadelphia; 2004. p. 2120-22.

Case Report

Journal of Emergency Medicine Case Reports

A Report of Recurring Pregnancy-İnduced Cushing's Syndrome

Amir Bahrami[1], Fatemeh Ghorbani[1], Nasser Aghamohammadzadeh[1,3], Vahideh Sadra[1], Nader Nader Djalal[4], Mitra Niafar[1,2]

[1]Endocrine Research Center, Tabriz University of Medical Sciences, Tabriz, Iran.
[2]Faculty of Medicine, Tabriz University of Medical Sciences, Tabriz, Iran.
[3]Imam Reza Hospital, Tabriz University of Medical Sciences, Tabriz, Iran.
[4]Department of Anesthesiology, Jacobs School of Medicine and Biomedical Sciences, University at Buffalo, Buffalo, NY 14203, USA

Abstract

Introduction: The incidence of pregnancy-induced Cushing's syndrome (CS) is very low. However, the diagnosis of CS in pregnant patients is very difficult because of an overlapping signs and symptoms.

Case report: Herein, a 20-year-old pregnant patient was reported that was afflicted by gestational CS during two pregnancies; termination of pregnancy was carried out in both cases. Bilateral adrenal hypertrophy was reported in magnetic resonance imaging. Following therapeutic abortion, signs of chemical evidence of CS was thoroughly regressed within a few months after each abortion without any treatment.

Conclusion: Due to the temporal relation between the occurrence of CS after each pregnancy the diagnosis of gestational hypercortisolism was made, and for further pregnancies bilateral adrenalectomy was recommended.

Key Words: Cushing`s syndrome, pregnancy, adrenal hypertrophy

Introduction

The functional physiology of several systems differs in pregnancy secondary to both fetal and maternal causes. The endocrine system is also affected by pregnancy-induced homeostasis among all other systems. Due to the associated infertility and inhibition of gonadotropin secretion, Cushing`s syndrome (CS) is rarely reported in a pregnant woman[1]. Moreover, aberrant expression of receptors of luteinizing hormone (LH) and human chorionic gonadotropin (HCG) in the adrenal membrane further decrease the likelihood of primary hypercortisolism syndrome in pregnancy. Regardless, development of CS during pregnancy poses serious threats to both the pregnant mother and the embryo, such as premature delivery and stillbirth[2, 3]. In the current study, we report the occurrence of CS in a young woman during two consecutive pregnancies that both developed during the first trimester of pregnancy. In both occasions, CS was fully remitted following termination of pregnancy without any further therapy.

Case Report

A 20-year-old woman with polycystic ovarian syndrome (PCOS) was successfully treated with clomiphene for infertility and pregnant in September of 2016 for the first time. She experienced out of proportion abdominal obesity, excessive striosis, exacerbation of hirsutism (Grade 20 on Ferriman-Gallwey scale)[4], increased facial hair and myasthenia during the first trimester of her pregnancy. Initial diagnosis of ovarian hyperstimulation syndrome (OHSS) was made and due to concomitant intrauterine growth retardation (IUGR), the pregnancy was electively terminated at the 8th week. All associated symptoms spontaneously regressed within 2-3 months after the abortion. The second pregnancy was induced with a pulse treatment of letrozole 5 mg, followed by 3 every other day injections of 100 units recombinant follicle-stimulating hormone (rFSH) in November 2017. Within the first 4 weeks into the second pregnancy, the patient developed recurrence of OHSS symptoms (Figure 1). In addition to the prior symptoms and progressive proximal myasthenia, the patient developed hypertension, round moon-like face, back hump, and suprasternal fat pad consistent with the typical features of CS.

In week 14 of pregnancy, the patient complained from acute-onset bilateral blindness and hypertension (blood pressure of 180/120 mmHg), which soon progressed into loss of consciousness and grand-mal seizure. She was admitted to the intensive care unit (ICU) with a preliminary diagnosis of intracerebral bleeding. Magnetic resonance imaging (MRI) of the brain was performed which ruled out intracerebral/pituitary hemorrhages/lesions. Admission laboratory testing

Corresponding Author: Mitra Niafar **e-mail:** dr.mniafar@gmail.com
Received: 16.10.2019 • **Accepted:** 17.01.2020
DOI:10.33706/jemcr.633840

DOI:10.33706/jemcr.633840
Journal of Emergency Medicine Case Reports 2020; 11 (1):13-16

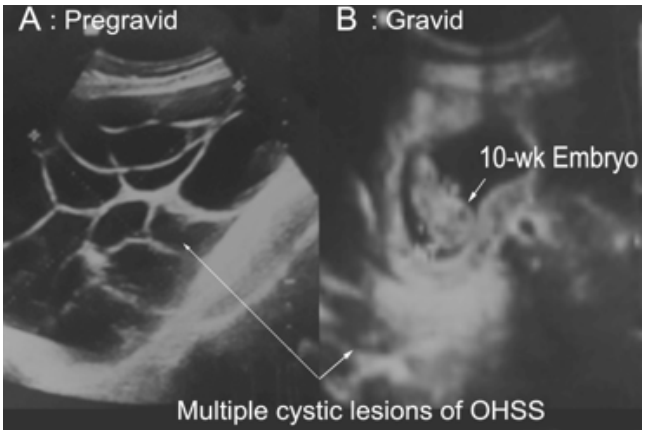

Figure 1. Abdominal ultrasound of the pelvis before the conception (A), showing a large ovary containing multiple cysts (OHSS) and (B) a normal intrauterine pregnancy.

included those to assess the function of the pituitary-adrenal axis (HPA) due to the presence of symptoms related to CS. This test battery included determination of serum concentrations of baseline cortisol and after low-dose dexamethasone suppression, serum adrenocorticotropic hormone (ACTH), renin, aldosterone, and 24-hour urine concentrations of free cortisol, normetanephrine and metanephrine. Additional laboratory findings upon admission included progressive increases in aspartate aminotransferase (AST; 119 → 522 U/L); alanine aminotransferase (ALT; 191→ 368 U/L) and borderline platelet count. Based on available clinical information a diagnosis of hemolysis, elevated liver enzymes, and a low platelet count (HELLP) syndrome was made and emergency termination of pregnancy was carried out at the gestational age of 14 weeks.

The patient`s clinical signs such as hypertension and acute blindness were totally subsided soon after the therapeutic abortion. An additional endocrinological investigation was obtained during the postoperative period while the patient was still hospitalized. Diagnosis of ACTH independent CS was made based on elevated 8 a.m. serum cortisol level unresponsive to low-dose dexamethasone (45→41 µg/dL), elevated urine cortisol, lower ACTH and normal level of serum renin, aldosterone and urine concentrations of metanephrine and normetanephrine. The patient was discharged from the hospital after she recovered from the acute blindness, normalization of the blood pressure and declining liver enzymes.

She was then followed up as an outpatient with the University Endocrine Center. With an exception of myasthenic symptoms, all other symptoms fully recovered within a 2-3 month period. The results of muscular examination of the patient revealed proximal (3 out of 5) and distal (4 out of 5) myasthenia and at the bedside, the patient was not able to get down the bed easily and walk without help. Serial values reveal a gradual decrease in beta- HCG along with serum and

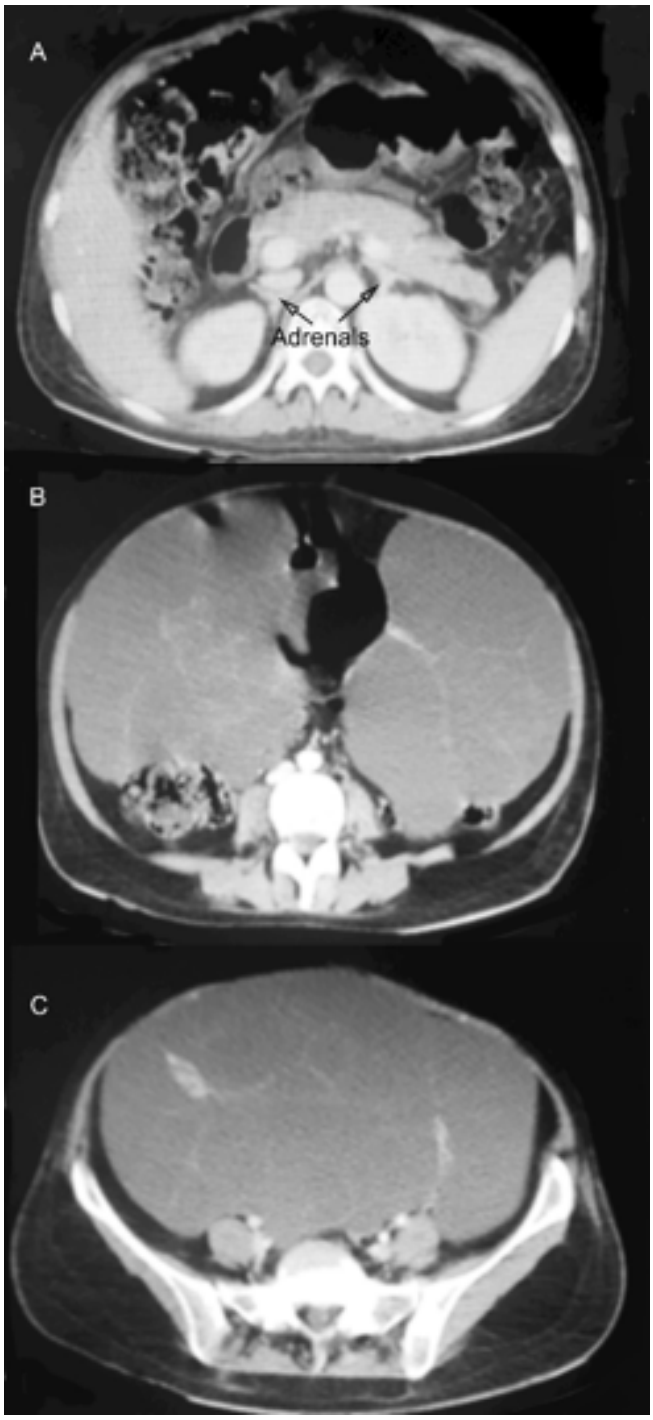

Figure 2. Computer tomographic scan of the adrenal glands showing enlarged adrenals (A), and huge ovaries containing multiple cysts (B and C).

urine cortisol concentrations, which paralleled with gradual increases in ACTH levels (Table 1). The results of CT scan of abdomen and pelvis requested for the patient during the previous hospitalization revealed bilateral hypertrophy of adrenal glands (Figure 2A) in addition to massive bilateral pelvic mass (Figure 2B and 2C). An ultrasound study of the pelvis revealed bilateral polycystic deformation of both ovaries containing multiple parietal nodules. The borderline values ovarian biomarkers, i.e., alpha-1-fetoprotein (AFP)

Table 1. Laboratory findings.

Parameters	Abortion	After abortion							
Date	3/7/18	4/5/18	4/9/18	4/28/18	5/7/18	5/15/18	6/1/18	7/28/18	11/22/18
Beta-HCG (mIU/mL)	838	258	201	100	-	57.6	28.5	16	-
Serum Cortisol (µg/dL)	45	-	-	-	19.1	-	-	6.7	17.7
24 h Urine Free Cortisol (µg/day)	1984	-	346	70	-	-	71.4	-	-
Adrenocorticotropic hormone (pg/ml)	3	-	<5	-	<5	-	-	21.2	16.9

of 4 ng/L, carcinoembryonic antigen (CEA) of 7.1 ng/mL and cancer antigen (CA125) of 151 U/mL were compatible with PCOS. Additionally, serum testosterone concentration was 3.4 nmol/L which later decreased to 0.64 nmol/L without further treatment, and other androgen levels also remained at their borderline levels (dehydroepiandrosterone sulfate DEHAS = 1.27 µmol/L; 17 hydroxyprogesterone (17-OHPG) of 1.86 ng/mL.

Discussion

Cushing`s syndrome rarely occurs during pregnancy[1]. Diagnosis of CS in pregnancy is challenging as the physiological changes during pregnancy may mimic those of CS. Moreover, the placental production of corticotropin releasing hormone (CRH) may result in some symptoms of CS by stimulating the ACTH/cortisol axis. In fact, both free and protein-bound concentrations of cortisol in plasma and urine significantly increase during a normal gestation. Dexamethasone suppression dose generally fails to reduce cortisol levels to the extent that is seen in a non-pregnant individual. Both maternal and fetal complications such as growth retardation, premature birth and stillbirth occur more frequently in the presence of CS[5]. But in our case, postpartum remission of CS after the two consecutive pregnancies suggests a temporal cause-effect relationship between CS and gravidity.

The choice of a treatment strategy for gestational CS is very complex and it varies from one case to another. Among the factors that determine the choice of therapy, maternal age and the severity of disease are very important. The use of metyrapone and/or ketoconazole for treatment of gestational CS have been used with a great success in patients with high operative risk and also for the preparation of patients for surgical treatment[6].

In a study performed by Bevan et al., a woman was diagnosed with CS during the 29th gestational week. Following a course of metyrapone and adrenalectomy in week 31, the patient underwent a successful vaginal delivery in week 36 of pregnancy[7]. Long-term treatment of gestational CS has not been as successful. However, in the case of adrenal hyperplasia, bilateral adrenalectomy offers a certain cure. Abbassy et al. reported that bilateral adrenalectomy effectively treated a 38-year-old patient who was diagnosed with gestational CS[2]. In another report, a 26-year-old patient with a diagnosis of non-ACTH dependent CS during week 19 of pregnancy underwent laparoscopic adrenalectomy with a very good outcome[8]. Although adrenalectomy is now considered the only cure for gestational CS, its timing during pregnancy is still a matter of debate. However, most surgeons prefer to perform adrenalectomy at the second trimester of pregnancy[9]. Our patient had a therapeutic abortion due to severe maternal complications and development of CS during the early gestational age. The patient was reeducated on the subject of contraception and the possibility of adrenalectomy for future pregnancies.

The case report has written in an anonymous characteristic, thus secret and detailed data about the patient has removed. Editor and reviewers can know and see these detailed data. These data are backed up by editor and by reviewers.

Conclusion

In conclusion, we present a patient with reversible pregnancy-induced Cushing Syndrome that led to untimely termination of pregnancy. As expected, serum cortisol levels returned to normal levels during one month, and the clinical manifestations of Cushing completely disappeared in 90 days. It is perceived that the symptoms of Cushing syndrome secondarily developed to an unusual presence of ectopic adrenal LH/hCG receptors that release cortisol in response to hCG stimulation. A proper diagnosis perhaps offers more options such as adrenalectomy to the patients who wish to have children. Adrenalectomy also provides needed tissue samples for immunohistochemistry staining which confirms the expression of LH/hCG receptors by the ectopic adrenal cells.

Conflict of interests

Authors declare no conflict of interests.

DOI:10.33706/jemcr.633840
Journal of Emergency Medicine Case Reports 2020; 11 (1):13-16

Financial Disclosure

The authors declared that this study has received no financial support.

Acknowledgment

Authors would like to acknowledge Dr. Nader Nader Djalal at Department of Anesthesiology, Jacobs School of Medicine and Biomedical Sciences, University at Buffalo, Buffalo, NY 14203, USA for his great help.

References

1. Vilar L, Freitas Mda C, Lima LH, Lyra R, Kater CE. (2007). Cushing's syndrome in pregnancy: an overview. Arq Bras Endocrinol Metabol. 51(8):1293-302.
2. Abbassy M, Kshettry VR, Hamrahian AH, Johnston PC, Dobri GA, Avitsian R, et al. (2015). Surgical management of recurrent Cushing's disease in pregnancy: A case report. Surg Neurol Int. 6(Suppl 25):S640-5.
3. Kita M, Sakalidou M, Saratzis A, Ioannis S, Avramides A. (2007). Cushing's syndrome in pregnancy: report of a case and review of the literature. Hormones-Athens. 6(3):242-46.
4. Niafar M, Nader ND. (2015). Adiponectin as serum biomarker of insulin resistance in patients with polycystic ovarian syndrome. Gynecol Endocrinol. 31(6):473-6.
5. Lindsay JR, Jonklaas J, Oldfield EH, Nieman LK. (2005). Cushing's syndrome during pregnancy: personal experience and review of the literature. J Clin Endocrinol Metab. 90(5):3077-83.
6. Shaw JA, Pearson DW, Krukowski ZH, Fisher PM, Bevan JS. (2002). Cushing's syndrome during pregnancy: curative adrenalectomy at 31 weeks gestation. Eur J Obstet Gynecol Reprod Biol. 105(2):189-91.
7. Bevans JS, Gough MH, Gillmere MDG, Burke CW. (1987). Cushing's syndrome in pregnancy: The timing of definitive treatment. Clinical Endocrinology. 27(2):225-33.
8. Nassi R, Ladu C, Vezzosi C, Mannelli M. (2015). Cushing's syndrome in pregnancy. Gynecol Endocrinol. 31(2):102-4.
9. Sam S, Molitch ME. (2003). Timing and special concerns regarding endocrine surgery during pregnancy. Endocrinol Metab Clin North Am. 32(2):337-54.

Case Report

Journal of Emergency Medicine Case Reports

Chronic Licorice Consumption as a Rare Cause of Hypocalcemia

Eda Kan[1], Tuba Sarıaydın[1], Murat Dogan[2], Ali Kemal Erenler[1]
[1]Department of Emergency Medicine, Hitit University Faculty of Medicine, Corum, Turkey
[2]Department of Internal Medicine, Hitit University Faculty of Medicine, Corum, Turkey

Abstract

Introduction: Licorice ingestion is well-described with its hypokalemic and hypertensive effects in the literature. To our knowledge, up to now, hypocalcemia due to licorice ingestion has not been described. Our aim is to create awareness for hypocalecemia concomittant with hypokalemia in patients with chronic licorice ingestion.

Case Report: A 51-year old male patient presented to our Emergency Department (ED) with complaint of numbness on his whole body. On anamnesis, it was determined that he has been consuming licorice to regulate his hypertension for 3 months. Laboratory analysis revealed hypokalemia and hypocalcemia. After replacement therapy, the patient was discharged from the hospital with full recovery.

Conclusion: Focusing on hypokalemia may lead to misdiagnosis of hypocalcemia in patients with chronic licorice ingestion. Physicians must be aware of other electrolyte disturbances not described in the literature yet.

Key Words: licorice ingestion, hypocalcemia, hypokalemia, emergency medicine

Introduction

Ingestion of licorice is a well-known reason for a syndrome mimicking mineralocorticoid excess, of which the pathophysiology is completely clarified. The characteristics of this syndrome are hypertension, hypokalaemia, alkalosis, low renin activity and hypoaldosteronism[1]. In this report, we present you a case with hypocalcemia accompanying hypokalemia in a patient with licorice consumption for hypertension regulation for 3 months. Our aim is to create awareness to hypocalcemia as a rare finding of chronic licorice consumption.

Case Report

A 51-year-old male patient was admitted to our Emergency Department (ED) due to generalized numbness. On admission he was hypertensive (150/95 mmHg) with a heart rate of 84 beats/min, a saturation of 96% by probe and a temperature of 36,2°C. On his medical history, hypertension for 10 years was determined. It was also determined that the patient has been consuming licorice for its antihypertensive properties for 3 months. Physical examination revealed apparent Chvostek's sign and Trousseau's sign. In laboratory analysis, Calcium (Ca) was 6.7 mg/dL and Potassium (K) was 3.5 mmol/L. A spot urine sample analysis showed urinary sodium of 122 mmol/L and potassium of 90 mmol/L. Other parameters were normal. An ECG was obtained and it did not reveal any abnormalities. In order to exclude a intracranial pathology, a computerized tomography (CT) was obtained and it was also normal. Hypocalcemia and hypokalemia due to chronic licorice ingestion was considered as a predignosis and the patient was consulted with an internal medicine specialist and a nephrologist. According to their suggestions, 40 ml K in 500 cc serum physiologique in 5 hours and 3 ampoules of %10 Ca in 5% dextrose in 2 hours were initiated. After 5 hours of ED observation, the patient was transferred to internal medicine ward. After 3-days follow-up with replacement therapy in the ward, symptoms of the patient and the laboratory values improved. The patient has been discharged with full recovery. The laboratory findings of the patient in the follow-up are summarized in Table 1.

Discussion

Licorice (Glycyrrhiza glabra L.) is one of the most widely used herb for ages. Licorice grows in the sub-tropical and warm temperate regions of the world, particularly in India and

Corresponding Author: Eda Kan **e-mail:** dredakan@gmail.com
Received: 04.07.2019 • **Accepted:** 05.03.2020
DOI:10.33706/jemcr.577197

DOI:10.33706/jemcr.577197
Journal of Emergency Medicine Case Reports 2020; 11 (1):17-18

Table 1: Follow-up of Ca+ and K+ values.

	On admission	Day 1	Day 2	Day 3
Ca+ (mg/dL)	6.7	6	7.4	7.8
K+ (mmol/L)	3.5	2.9	3	3.6

Mediterranean countries. Roots and stolon parts of licorice plants are used in mixed herbal preparations to promote digestion and vitality. Licorice root extract has been studied for its anti-cancer and anti-viral activities and healing of gastriculcers. Pharmacological investigations indicate that licorice root extracts have antioxidant, antibacterial and anti-inflammatory activities. The main ingredient of licorice roots glycyrrhizin or glycyrrhizic acid (GA), a triterpinoid saponin, is used for the control of cough, asthma, bronchitis, pepticulcer, arthritis and allergic reactions[2]. In our case, the patient declared that he used the herb to regulate hypertension.

Although hypokalemia is the most dangerous side effect of licorice consumption, the main adverse effect is hypertension. The USA Food and DrugAdministration (FDA) advises avoiding eating large amounts of black licorice at one time[3]. Our patient was also hypertensive on admission, however, we could not determine whether the hypertension was linked to licorice or the idiopatic hypertension in patient's medical history. It is an interesting example for misinformation since the patient uses a herb that causes hypertension to reduce hypertension. Unconscious consumptions of herbs may result in such life-threatening conditions.

Chronic licorice intoxication is a well-recognized cause of hypokalemia[4]. Additionally, it may cause hypokaliemic rhabdomyolysis, characterised by a sthenic deficit exclusively involving the distal muscles of the upper limbs and secondary to chronic glycyrrhizic acid intoxication, and by the absence of even ictal arterial hypertension[5]. Our patient complained of insensitiveness in whole body and laboratory findings revealed hypokalemia in concordance with the literature. However, in our case we also determined hypocalcemia which could not be explained with another pathology. To our knowledge, so far, hypocalcemic effect of licorice ingestion has not been identified in the literature.

Licorice withdrawal and potassium replacement are the mainstay of therapy, though caution is advised in the use of IV potassium and recommendations are made about the rate of infusion. The potassium-replacement therapy should consist of potassium administered in glucose free solutions, since glucose may cause a further decrease in potassium concentration and can reverse any beneficial effect of administered potassium, with the risk of precipitating arrhythmias, neuromuscular paralysis, and respiratory failure. The use of glucose-free solutions during IV administration of potassium should be regarded as an important therapeutic principle[4]. In our case, in addition to K replacement, due to hypocalcemia, Ca replacement was required. A close follow-up and monitorization may be beneficial in such cases with multiple electrolyte abnormalities.

Conclusion

It is recommended for physicians working in the ED to be aware of the problems in electrolyte and blood pressure homeostasis that can occasionally occur with the excessive intake of licorice-containing products. In chronic licorice intake hypocalcemia accompanying hypokalemia must be kept in mind.

References

1. Janse A, van Iersel M, Hoefnagels WH, Olde Rikker MG. The old lady who liked liquorice: hypertension due to chronic intoxication in a memory-impaired patient. Neth J Med. 2005; 63(4): 149-50.
2. Rohinishree YS, Negi PS. Effect of licorice extract on cell viability, biofilm formation and exotoxin production by Staphylococcus aureus. J Food Sci Technol. 2016; 53(2): 1092-100.
3. Caravaca-Fontan F, Martinez-Saez O, Delgado-Yague M, Yerovi E, Liano F. An Unexpected Cause of Severe Hypokalemia. Case Rep Nephrol. 2015; 2015: 957583.
4. Famularo G, Corsi FM, Giacanelli M. Iatrogenic worsening of hypokalemia and neuromuscular paralysis associated with the use of glucose solutions for potassium replacement in a young woman with licorice intoxication and furosemide abuse. Acad Emerg Med. 1999; 6(9): 960-4.
5. Barrella M, Lauria G, Quatrale R, Paolino E. Hypokaliemic rhabdomyolysis associated with liquoric eingestion: report of an atypical case. Ital J Neurol Sci. 1997; 18(4): 217-20.

Case Report

Journal of Emergency Medicine Case Reports

Adrenal Cystic Lesion Presented With Spontaneous Hemorrhage

Barış Sevinç, Nurullah Damburacı, Cevdet Duran, Ömer Karahan

[1]Uşak University Medical School, Department of General Surgery, Uşak, Turkey
[2]Uşak University Medical School, Department of Internal Medicine, Division of Endocrinology and Metabolism, Uşak, Turkey

Abstract

Introduction: Adrenal hemorrhage is a rare but life-threatening condition. Spontaneous hemorrhage occurs without any trauma. Focal bleeding may cause even a subclinical condition or cardiovascular collapse with massive bleeding. Non-proper diagnosis may cause even death. In literature most of the cases are treated conservatively. Conservative treatment necessitates close follow up and if fails, surgery becomes challenging.

Case: Here, we present a case with spontaneous adrenal hemorrhage that was first taken to conservative treatment and undergone emergency surgery.

Conclusion: Conservative treatment of adrenal hemorrhage necessitates close follow up and if fails, surgery becomes challenging.

Key Words: Adrenal hemorrhage, retroperitoneal hematoma, cyst

Introduction

Spontaneous adrenal hemorrhage is a rare cause of retroperitoneal bleeding. The most common cause of unilateral adrenal bleeding is blunt abdominal trauma (traumatic adrenal rupture), which should be differentiated from the hemorrhage caused by primary or metastatic adrenal tumors[1]. Adrenal hemorrhage can be seen in cases with liver transplant, primary or metastatic adrenal tumors. Unilateral adrenal bleeding rarely caused by uncomplicated pregnancy, neurofibromatosis and long term non-steroidal anti-inflammatory drug use[1, 2]. The cases can be even asymptomatic or may present with hemorrhagic shock, flank pain or fever. In autopsy series, adrenal hemorrhage was detected in 0.3-1.8% of asymptomatic cases; bilateral adrenal hemorrhage was detected in 15% of cases with hemorrhagic shock[2, 3].

We aimed to present a case with adrenal hemorrhage; first followed conservatively and undergone emergency adrenalectomy.

Case

Twenty-five years old female patient admitted to hospital with abdominal pain localized on the left side of the abdomen. She had no fever, no urinary discomfort. At physical examination, nothing but left upper quadrant tenderness was detected. At abdominal ultrasonography, 11x10 cm mass that pushes the left kidney laterally and enlarged mesenteric lymph nodes were identified. Abdominal computed tomography revealed an adrenal hematoma 10 cm in diameter that pushes the stomach and the spleen (Figure 1). The patient was hemodynamically stable and internalized to the surgery department for follow up. At the follow up, hemoglobin level of the patient dropped by 3 gr/dl and control computed tomography detected the enlargement of the hematoma. Therefore, the patient had undergone emergency adrenalectomy. At the operation, an encapsulated adrenal hematoma, 15 cm in diameter was detected and left adrenalectomy was performed. The patient had no postoperative complaints and discharged from the hospital at the 5th postoperative day. Postoperative evaluation of the patient revealed no hormonal disturbance related to the adrenal disease. At the histopathological examination a ruptured endothelial cyst of the adrenal gland was detected.

Discussion

In literature most of the adrenal hemorrhage cases were related to pregnancy. Adrenal hemorrhage can be related to traumatic or non-traumatic reasons. Non-traumatic reasons are coagulopathy, anti-coagulant therapy, and tumor like angiomyelipoma, stress and shock. Tumors that may cause

Corresponding Author: Barış Sevinç **e-mail:** drbarissevinc@gmail.com
Received: 18.01.2019 • **Accepted:** 17.01.2020
DOI:10.33706/jemcr.514519

DOI:10.33706/jemcr.514519
Journal of Emergency Medicine Case Reports 2020; 11 (1):19-20

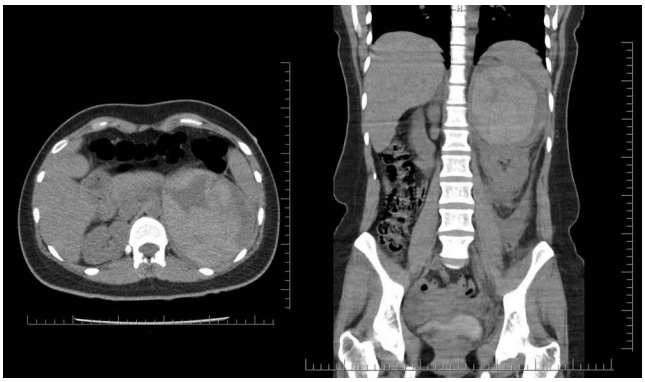

Figure 1

spontaneous hemorrhage are pheochromocytoma, myelipoma, metastasis, carcinoma and rarely adenoma. Spontaneous hemorrhage at a young age is extremely rare. In literature there are only two cases with spontaneous life threatening adrenal hemorrhage[5, 6]. The case we present is the third case in literature.

Adrenal cystic lesions can be subdivided as; pseudocyst, endothelial cyst, epithelial cyst and parasitic cyst. Endothelial cysts consist of the 45% of all adrenal cysts[7]. The first case with intra-cystic hemorrhage was reported by Vega et al. in 2014[8]. However in that case the patient was hemodynamically stable and therefore, operated electively. Besides in our case the hematoma enlarged and the hemoglobin level dropped, therefore, the patient underwent emergency surgery. At the operation a hematoma 15 cm in diameter was detected and adrenalectomy was performed as it was reported at the literature[9, 10].

As an adrenal tumor may be the cause of the hemorrhage, the patient should be evaluated for the hormonal disturbances. In our case, due to emergency surgical need, adrenal hormonal panel could not been evaluated. However, there was no electrolyte imbalance before the surgery.

In adrenal hemorrhage most of the cases are asymptomatic or flank pain, fever or hemorrhagic shock may be the cause of admittance to hospital. Therefore even it is asymptomatic, adrenal hemorrhage should be kept in mind in cases with flank pain.

The cases with adrenal or retroperitoneal bleeding should be closely followed and pheochormacytoma should always be kept in mind. The priority should be to keep the patient hemodynamically stable. Hematocrit level and hematoma diameter should be followed closely. Non-traumatic cases can be evaluated by computed tomography or magnetic resonance imaging. In evaluation of the retroperitoneum magnetic resonance imaging is favorable.

Angiographic embolization is a valuable tool for cases with active bleeding. If the embolization is enough to keep the patient stable, conservative follow can be sustained. Surgical options are always kept in mind in case of hemorrhagic shock. Before the operation medications for pheochromacytoma should be ready to use.

Conclusion

In case of retroperitoneal hematoma, abdominal or flank pain, adrenal hemorrhage should be an option in differential diagnosis.

References

1. Hiroi N, Yanagisawa R, Yoshida-Hiroi M, Endo T, Kawase T, Tsuchida Y ve diğ. Akciğer adenokarsinomundan bilateral adrenal metastaza bağlı retroperitoneal hemoraji. J Endocrinol Invest. 2006; 29 : 551-4.
2. Gavrilova-Ürdün L, Edmister DB, Farrell MA, Watson WJ. Gebelikte spontan adrenal kanama: Başarılı bir konservatif yaklaşımın literatür ve olgu sunumu. Obstet Gynecol Surv Mart. 2005; 60 : 191–5.
3. Kovacs KA, Lam YM, Pater JL. Bilateral kitle adrenal kanama. Varsayımsal risk faktörlerinin vaka kontrollü yöntemle değerlendirilmesi. Tıp (Baltimore) 2001; 80 : 45-53.
4. Pushkar P, Agarwal A (2015) Spontaneous massive adrenal hemorrhage: a management dilemma, *Journal of Endourology Case Reports* 1:1, 52–53, DOI: 10.1089/cren.2015.0003.
5. Paluszkiewicz P, Ambroziak I, Holynska-Dabrowska K, Siezieniewska-Skowronska Z, Paluszkiewicz A. Spontaneous rupture of adrenal haemangioma mimicking abdominal aortic aneurysm rupture. Arch Med Sci. 2010;6(1):122–125
6. Peng X, Luo W, Zhang X, Zhu W. Sudden onset flank pain: a case report of retroperitoneal hemorrhage secondary to a ruptured adrenal hemangioma. J Pain Res. 2018;11:1421-1424.
7. Foster DG. Adrenal cysts. Review of literature and report of case. Arch Surg 92:131–43, 1966.
8. Fernández-Vega, I., Camacho-Urkaray, E. & Guerra-Merino, I. Huge Adrenal Hemorrhagic Endothelial Cyst Secondary to an Adrenal Arteriovenous Malformation and Mimicking a Malignant Lesion Endocr Pathol (2014) 25: 443.
9. Lyu X, Liu L, Yang L, Gao L, Wei Q. Surgical management of adrenal cysts: a single-institution experience. Int Braz J Urol. 2014 Sep-Oct;40(5):656-65
10. Damburacı, N , Güner, M , Sevinç, B , Karahan, Ö . (). Use of a vessel sealing system (LigaSure Impact™) in the sealing and division of the intestine. Ege Tıp Bilimleri Dergisi, 1 (1), 7-12

Case Report

Journal of Emergency Medicine Case Reports

The Contribution of Measuring the Optic Nerve Sheath Diameter by using Ultrasonography to the Diagnosis and Monitoring of Intracranial Hypertension

Özlem Tolu Kendir[1], Hayri Levent Yılmaz[1], Ahmet Kagan Ozkaya[1], Sinem Sarı Gökay[1], Sevcan Bilen[1]

Cukurova University, Pediatrics, Emergency Care Unit, Balcalı Hospital Adana Turkey

Abstract

Background: Intracranial hypertension, which may have a number of different causes, is a medical emergency. Although difficult to identify due to non-specific symptoms, it must be treated appropriately. An early sign of intracranial hypertension is an increase is observed in the optic nerve sheath diameter. . Ultrasonography is a rapid and easy-to-apply method to detect an increase in optic nerve sheath diameter, and may be useful for the emergency diagnosis and treatment, as well as contributing to clinical follow-up.

Case Presentation: In the present study, seven patients with clinical suspicion for rapid intracranial pressure were admitted. Clinical scenarios included traffic accident, metabolic imbalance, infection, and intracranial mass. The pediatric emergency specialist performed optic nerve sheath measurement by using ultrasonography and a dilatation was detected. All measurements were taken by the certified and experienced pediatric emergency specialist blinded to the patient's clinical state and also the specialist was under observation of a lecturer during the measurements. Except for one patient, the control optic sheath diameter measurements were performed in the period, when they had no complaint, and the values were determined to be normal. To our knowledge, this case series is the first one examined in Turkey. The data from cases was collected between 2015 and 2016.

Conclusion: Optic nerve sheath diameter measurement is a non-invasive method that offers ease of diagnosis and follow-up of suspected intracranial hypertension.

Key Words: Optic nerve sheath diameter measurement, intracranial hypertension, ultrasonography

Background

Intracranial hypertension is a condition that may result in mortality or negative neurological consequences, and therefore requires emergent diagnosis and treatment. Computed tomography of the head is preferred as the first option in order to support clinical assessment, however, involves radiation exposure. It may not be possible to perform the lumbar puncture or invasive intracranial monitoring under specific conditions. Fundoscopic examination may also be misleading since the papilla stasis is a late-phase symptom of intracranial hypertension. Ultrasound, which is a non-invasive and rapid method that is useful in during the acute period and follow-up, may be an option for measuring the optic nerve sheath diameter. The present case series demonstrates how measurement of the optic nerve sheath diameter may contribute to diagnosing and following intracranial hypertension.

Cases Presentation

After obtaining the written consents of their parents, seven patients with suspicion of intracranial hypertension (IH), who were admitted to the Pediatric Emergency Department of Medical Faculty of **Çukurova** University due to traffic accident, metabolic imbalance, infection, and intracranial mass formation, are presented. The data of patients can be seen in Tables 1 and 2. Six of the patients had undergone control ultrasonography (USG) when they arrived to the polyclinic. No control USG was applied to one of these patients because the patient was followed-up in another center after the discharge from hospital.

Method

After the first examination, transorbital ultrasonography was performed by placing the patient in supine and neutral position with closed eyes. Sonosite Edge ultrasonography device was used with the high frequency (15-6 MHz) planar probe and gel. The diameter of optic nerve sheath, which is seen as a hypoechoic double-sided line at the 3mm depth of globe that is known to be most sensitive to the intracranial pressure changes, was measured in both longitudinal and transverse cross-sections, and the mean value was calculated[1] (Figure 1-2). All of the measurements were performed

Corresponding Author: Özlem Tolu-Kendir **e-mail:** otolu80@yahoo.com

Received: 05.02.2019 • **Accepted:** 05.03.2020

DOI:10.33706/jemcr.522919

DOI:10.33706/jemcr.522919
Journal of Emergency Medicine Case Reports 2020; 11 (1):21-26

Table 1. Age, gender, and vital findings of patients, Glasgow Coma Scale

Patient	Age	Gender	Body Temperature °C	Heart rate /min	Respiration rate /min	Blood pressure (mmHg)	GCS
1	12 year-old	Girl	36.5	152	8	80-40	5 (PTS<8)
2	28 month-old	Girl	36.5	144	46	85-40	6
3	4.5 year-old	Girl	38.5	132	24	110-60	10
4	7 year-old	Boy	38	135	48	100-60	13
5	17 year-old	Girl	37.8	88	22	110-60	13
6	17 year-old	Boy	38.3	100	24	110-70	14
7	10 year-old	Boy	36.4	118	24	100-60	14

by a pediatric emergency subspecialist having USG training background.

Discussion and Conclusions

Intracranial hypertension requires early rapid diagnosis, however, clinical symptoms and findings may be confused with other conditions. In the present study, three patients were found to have IH because of infectious factors, two patients because of metabolic and autoimmune encephalopathy, one patient because of a non-vehicular traffic accident and one patient because of intracranial mass.

In practice, the diagnosis of IH is made based on the basis of clinical findings. This case series describes the usability of USG, which is a non-invasive monitoring method, in measuring optic nerve sheath diameter (OSND) for these patients.

The ultrasonography is a non-invasive, reliable, affordable, repeatable, and highly available method that has no known adverse effect. In 1990s, emergency specialists, who had USG training from American College of Emergency Physicians (ACEP), started to use the USG in emergency units, and the first guideline on this subject was published in 2001. The documents published by Council of Emergency Medicine Residency Directors between 2009 and 2012 were used as a reference for the relevant procedures[2]. Even though no specific guideline on the use of USG in pediatric emergency has been published yet, the focused use became a very increasingly popular[2].

When diagnosing the intracranial hypertension, Computed Tomography (CT) is used as the primary option in practice. Disadvantages include radiation exposure, and difficulties associated with patient transportation. However,

Table 2. Complaints and clinical conditions of patients, radiologic assessment, and ONSD measurements

Patient	Complaint	BBT	Papilla stasis	ONSD mm (avg.)	Diagnosis	Clinical course	Control ONSD
1	ADTK	Cerebral edema (moderate)-subaracnoid-intraparenchymal hemorrhage	+	0.59	ADTK	Discharged with recovery	0.41
2	Drowsiness	Disseminated cerebral edema –subfalcine herniation	+	0.77 (ODE+)	Encephalopathy/hyperammoniemia	Discharged with recovery	-
3	Drowsiness - Fever-Vomiting	Cerebral edema (moderate-severe), Leptomeningeal contrast	+	0.68 (ODE+)	Meningoencephalitis	Discharged with recovery	0.45
4	Fever-Vomiting	Normal	-	0.55	Meningoencephalitis	Discharged with recovery	0.45
5	Drowsiness - Fever-Vomiting	Cerebral edema (mild)	+	0.64	Ensefalit (otoimmun)	Discharged with recovery	0.44
6	Diarrhea, then drowsiness	Leptomeningeal contrasting, no edema	-	0.54	Meningoensefalit	Discharged with recovery	0.38
7	Headache-vomiting	Cystic lesion in left frontoparietal	+	0.64 (ODE+)	Cyst hydadid	Discharged with recovery	0.38

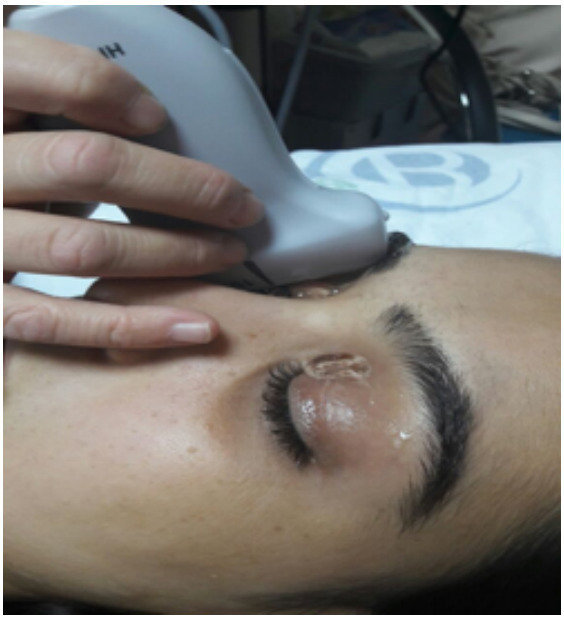

Figure 1. An application from the study

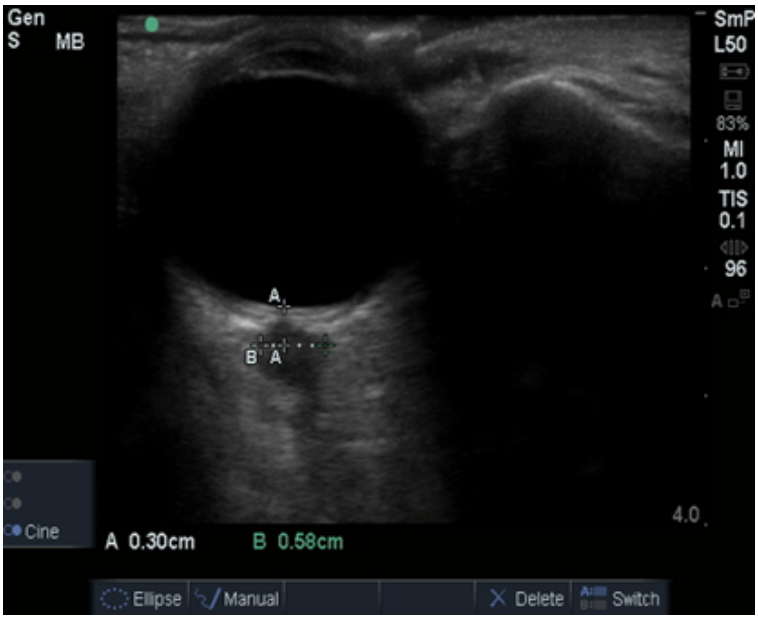

Figure 2. Images of Optic Nerve Sheath Diameter of the patients from this study

magnetic resonance (MR) is not an examination method that can be easily accessed, or that yields fast results. Neither CT nor MR is solely practical or non-risky in the acute setting[3].

Lumbar puncture can be used to directly measure the cerebrospinal fluid pressure, however, it is invasive, and contraindicated under certain conditions[4]. On the other hand, papilla stasis can be examined by using the fundoscopic examination. The examination depends on the individual. Normal findings may be achieved in early periods of IH, and there may be misleading findings under the emergency conditions because of mid-term/delayed identification of IH[5, 6].

In the present study, no invasive method was applied in order to confirm the diagnoses. The CT images of two patients were found to be normal, whereas there was cerebral edema in the others and one of these patients had post-herniation syndrome (Figure 3). All of the patients seen to have edema in CT images were determined to have papilla stasis, whereas it was not observed in other two cases.

Even though it was at lower levels in two patients, who were clinically projected to have IH and in CT images of whom we found no cerebral edema, it was determined that all the patients had dilated ONSD and these values exceeded the cut-off value reported in literature. No papilla stasis was found in ophthalmologic examination of two patients, who were found to have no cerebral edema. It was determined that there was a recovery in ONSD measurement values of both patients in the period, when the general clinical conditions of them gradually recovered. There was no patient which had intracranial hypertension has measurement within normal ONSD range.

The optic nerve is surrounded by three meningeal membranes, and the cerebrospinal fluid (CSF) freely circulates in intracranial and intraorbital subarachnoid space. In case of IH, the increase in intracranial subarachnoid pressure reflects on the intraorbital subarachnoid space[4, 6]. In the previous studies, it was reported that the increase in ONSD could be determined to be unechoic most accurately at 3 mm posterior to the globe. Even though there still is no standard cut-off value, it is asserted that ONSD values of 4.5 mm for children and 5mm for adults are suggestive of increased intracranial pressure[7, 8, 9].

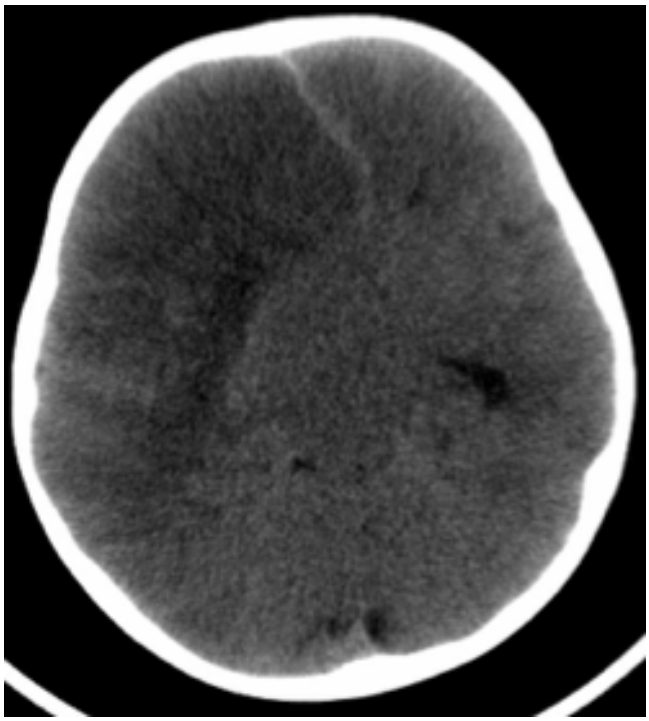

Figure 3. Subfulccial herniation in CT image of second patient

DOI:10.33706/jemcr.522919
Journal of Emergency Medicine Case Reports 2020; 11 (1):21-26

As in two patients in this series, the ONSD dilatation can be showed before the papilla stasis develops and cerebral edema is detected using radiologic method. Thus, the treatment can be started in early period before the clinical symptoms and findings become severe[5]. From another perspective, ONSD was larger in seven patients, for whom we confirmed the diagnosis of cerebral edema by using radiologic and fundoscopic methods. Our two cases with severe cerebral edema had optic disc elevation (ODE) in which the optic disc has process towards the posterior camera. One of them had subfalcine herniation. ODE in literature has been presented as indication of severe cerebral edema like our cases[10, 11].

In literature on examining the advantages of ONSD measurement, the studies using CT, lumbar puncture and/or papilla stasis as the gold standard reported the ONSD measurement as a reliable diagnostic modality. In their study on the patients having non-traumatic radiographic (CT) cerebral edema, Salahuddin et al. (2016) reported the cut-off value to be 0.57cm, sensitivity to be 84%, specificity to be 71%, and AUC to be 0.785. In their study on 16 patients aged between 12 and 18 years and suspected to have pseudo-tumor cerebri, Irazuzta et al. (2016) reported the mean ONSD to be 0.51cm±1.2 mm for those having cerebrospinal liquid pressure (CFP) of >20 cm-H_2O, and 0.38±0.02 cm for those with CFP <20 cm-H_2O ($p<0.01$) [12, 13]. In their study on 76 patients, Marchese et al. (2017) taking the fundoscopic examination as reference detected papilla stasis in 20 (26%) of IH cases, in which they observed ONSD enlargement (cut-off:0.45) (sensitivity: 90% and specificity: 55%). In their study carried out on 174 patients, Padayachy et al. (2016) took the intracranial pressure measurement as a reference and found the cut-off value to be 0.55 when the intracranial pressure at the moment of lumber puncture is below > 20 mmHg (sensitivity:92.2% - specificity: 74%)[14, 15].

In the case series presented here, it was determined that, ODE was observed and the level of increase in ONSD was higher when the cerebral edema was detected by using fundoscopic and radiologic methods, and that the cerebral edema can be detected in much earlier phase. It was determined that, in the period when the clinical symptoms of IH regressed, the ONSD measurements approached the upper limit of normal.

Based on the findings obtained in this study, it can be thought that the ONSD measurement might support the early diagnosis, that it is useful in follow-up period, and that this method is a highly available, affordable and useful, and may protect the patients from exposure to radiation. In order to make clearer inferences, more comprehensive studies on higher number of patients are needed.

Acknowledgements: N/A.

Abbreviations

IH: Intracranial Hypertension
USG: Ultrasonography
MR: Magnetic Resonance
CT: Computer Tomography
LP: Lumber Puncture
ONSD: Optic Nerve Sheath Diameter Measurement by using Ultrasonography
GKS: Glasgow Coma Scale
CFP: Cerebrospinal Fluid Pressure
PTS: Penetrant Trauma Score
ODE: Optic Disc Elevation

Declarations

- **Ethics approval and consent to participate:** This article does not contain any studies with human or animal subjects performed by any of the authors.
- **Consent for publication:** Authors have permission from the patient's parents as written and also there is no any information in the main text related with patients.
- **Availability of data and material:** All required data can be found in the case report
- **Competing interests:** The authors declare that they have no competing interests
- **Funding:** There is no any funding for this article.
- **Authors' contributions:** Each author have participated sufficiently in the work to take public responsibility for appropriate portions of the content.
- **Acknowledgements:** There is no any contributor out of Author list.

References

1. Del Saz-Saucedo P, Redondo-González O, Mateu-Mateu Á, Huertas-Arroyo R, García-Ruiz R, Botia-Paniagua E. Sonographic assessment of the optic nerve sheath diameter in the diagnosis of idiopathic intracranial hypertension. J Neurol Sci. 2016; 361:122-127
2. Marin JR, Lewiss RE. Point-of-Care ultrasonography by pediatric emergency medicine physicians. Pediatrics. 2015; 135(4): 1113-22.
3. Marchese RF, Mistry RD, Scarfone RJ, Chen AE. Identification of optic disc elevation and the crescent sign using point-of-care oculer ultrasound in children. Pediatr Emerg Care. 2015; 31(4): 304-7.
4. Amin D, McCormick T, Mailhot T. Elevated intracranial pressure diagnosis with emergency department bedside ocular ultrasound. Case Rep Emerg Med. 2015; 385970: 3p.

5. Zaidi SJH, Yamamoto LG. Optic nerve sheath diameter measurements by CT scan in ventriculoperitoneal shunt obstruction. Hawaii J Med Public Health. 2014; 73(8): 251-5.
6. Qayyum H, Ramlakhan S. Can oculer ultrasound predict intracranial hypertension? A pilot diagnostic accuracy evaluation in a UK emergency department. Eur J of Emerg Med. 2013; 20(2):91-97
7. Maude RR, Hossain A, Hassan MU, Osbourne S, Sayeed KLA, et al. Transorbital sonographic evaluation of normal optic nerve sheath diameter in healthy volunteers in Bangladesh. PLoS One. 2013; 8(12): e81013.
8. McLario DJ, Sivitz AB. Point of care ultrasound in pediatric clinical care. *JAMA Pediatr*. 2015;169(6):594-600.
9. Yakov MB, Desjardins MP, Fischer JWJ. Emergency point-of-care ultrasound detection of papilledema in the pediatric emergency department. Pediatr Emerg Care. 2015; 31(11): 766-70.
10. Stone MB. Ultrasound diagnosis of papilledema and increased intracranial pressure in pseudotumor cerebri. Am J Emerg Med. 2009; 27(3): 376.e1–376.e2.
11. Lochner P, Nardone R, Tezzon F, Coppo L, Brigo F. Optic nerve sonography to monitor treatment efficacy in idiopathic intracranial hypertension: A Case Report. J Neuroimaging. 2013; 23: 533-534.
12. Salahuddin N, Mohammed A, Alharbi N, Ansari H, Zaza KJ, Marashly Q et al. The incidence of increased ICP in ICU patients with non-traumatic coma as diagnosed by ONSD and CT: a prospective cohort study. BMC Anesthesiology. 2016; 16(1):106.
13. Irazuzta JE, Brown ME, Akhtar J. Bedside optic nerve sheath diameter assessment in the identification of increased ıntracranial pressure in suspected idiopathic intracranial hypertension. Pediatr Neurol. 2016; 54: 35-38.
14. Marchese RF, Mistry R, Binenbaum G, Liu GT, Scarfone RJ, Woodford AL et al. Identification of optic nerve swelling using point-of-care ocular ultrasound in children. Pediatr Emerg Care. 2018; 34(8): 531-536.
15. Padayachly LC, Padayachly V, Galal U, Gray R, Fieggen AG. The relationship between transorbital ultrasound measurement of the optic nerve sheath diameter (ONSD) and invasively measured ICP in children. Childs Nerv Syst. 2016; 32:1769–1778.