MEVLANA TIP BİLİMLERİ
MEVLANA MEDICAL SCIENCES
eISSN:2757-976X

OLGU SUNUMU/CASE REPORT DOI:http:/dx.doi.org/10.56752/Mevmedsci.2024.67
Mev Med Sci, 2024; 4(2): 96-98

A Case of Infantile Hypertrophic Pyloric Stenosis Presenting with Episodes of Apnea

Apne Ataklari ile Başvuran bir İnfantil Hipertrofik Pilor Stenozu

Abdullah Akkus[1], Methiye Dogan[1], Ezgi Yilik[1], Ahmet Osman Kilic[1], Abdullah Yazar[1], Fatih Akin[1]

[1]Necmettin Erbakan University, Faculty of Medicine, Child Health and Diseases, Konya, Türkiye

Makale Tarihleri/Article Dates:
Geliş Tarihi/Received: 20 April 2024
Kabul Tarihi/Accepted: 20 May 2024
Yayın Tarihi/Published Online: 20 August 2024

Sorumlu Yazar/Corresponding Author:
Abdullah Akkus,
Necmettin Erbakan University, Faculty of Medicine, Child Health and Diseases, Konya, Türkiye
e mail: abdullahakkus29@gmail.com

Açıklama/Disclosure: Yazarların hiçbiri, bu makalede bahsedilen herhangi bir ürün, aygıt veya ilaç ile ilgili maddi çıkar ilişkisine sahip değildir. Araştırma, herhangi bir dış organizasyon tarafından desteklenmedi. Yazarlar çalışmanın birincil verilerine tam erişim izni vermek ve derginin talep ettiği takdirde verileri incelemesine izin vermeyi kabul etmektedirler.

ÖZET

İnfantil hipertrofik pilor stenozu (İHPS), pilorik düz kas liflerinin hiperplazisine bağlı olarak pilor kanalının daralmasıdır. Yenidoğan döneminde görülen mide çıkışı darlıklarının en sık sebebidir. Genel olarak, doğum sonrası herhangi bir problemi olmayan 3-6 haftalık infantlarda beslenmeyi takiben fışkırır tarzda ve safrasız kusma ile kendini gösterir. Tanı konulamayan vakalarda dehidratasyon, ciddi beslenme bozukluğu, asit-baz dengesizliği ve apne ile seyredebilmekte olup, ihmal edilmesi halinde ölümle sonuçlanabilir. Yapılan çalışmalarda erkek çocuklarda kızlara göre 4 kat daha sık görülmektedir. Tanı için öykü ve fizik muayene yeterli olsa da ultrasonografi (USG) ve kontrastlı pasaj filmler ile tespit edilmektedir. Tedavi cerrahi olarak yapılan ekstramukozal pyloromyotomidir. Bu yazıda, 1 hafta önce başlayan ara ara nefes almada zorluk-apne, uyku hali ve beslenememe şikâyeti ile başvurup İHPS tanısı alan 1 ay 7 günlük bir erkek olguyu sunmayı amaçladık.

Anahtar Kelimeler: Pilor stenozu,apne,çocuk

ABSTRACT

Infantile hypertrophic pyloric stenosis (IHPS) is narrowing of the pyloric duct due to hyperplasia of pyloric smooth muscle fibers. It is the most common cause of gastric outlet stenosis during neonatal period. It usually presents with projectile nonbilious vomiting in an otherwise healthy infants at 3 to 6 weeks of age. Undiagnosed cases may have a course with dehydration, severely poor feeding, acid-base imbalance and apnea, which may lead to death when neglected. Previous studies have reported that it is 4-fold more common in boys than girls. Although history and physical examination is sufficient to make the diagnosis, it is detected by ultrasonography and contrast-enhanced passage x-rays. Treatment is surgical extramucosal pyloromyotomy. In this manuscript, we aimed to report a 37-day-old male case which was admitted with complaints of intermittent dysnpnea-apnea, somnolence, and difficulty feeding persisting for a week, and then diagnosed with IHPS.

Key words: Pyloric stenosis,apnea,infant

Atıf yapmak için/ Cite this article as: Akkus A, Dogan M, Yilik E, Kilic AO, Yazar A, Akin F. A Case of Infantile Hypertrophic Pyloric Stenosis Presenting with Episodes of Apnea. Mev Med Sci. 2024; 4(2): 96-98

INTRODUCTION

Infantile hypertrophic pyloric stenosis (IHPS) is narrowing of the pyloric duct due to hyperplasia of pyloric smooth muscle fibers. It is the most common cause of gastric outlet stenosis during neonatal period. It usually presents with projectile nonbilious vomiting in an otherwise healthy infants at 3 to 6 weeks of age. Undiagnosed cases may have a course with dehydration, severely poor feeding, acid-base imbalance and apnea, which may lead to death when neglected (1,2). Previous studies have reported that it is 4-fold more common in boys than girls. Although history and physical examination is sufficient to make the diagnosis, it is detected by ultrasonography and contrast-enhanced passage x-rays. Treatment is surgical extramucosal pyloromyotomy. In this manuscript, a case which was admitted with apnea, which is rare, and then diagnosed with IHPS was reported.

CASE REPORT

A 1 month and 7 day-old male patient admitted to our emergency department with complaints of intermittent shortness of breath, somnolence and difficulty feeding which had begun 1 week before. Patient's history revealed that he had had vomiting 5-6 times a day since postnatal 2nd week and that he had been diagnosed with cow's milk allergy and initiated hydrolyzed formula in another center. Patient's personal history shown that he was born at 38 weeks of gestation via vaginal delivery with a birth weight of 3500 g, and that he had not been hospitalized, except neonatal care following birth. His parents were nonconsangineous. There was no history of abortus-infant death. His on-admission physical examination showed a body temperature of 36.5°C, heart rate of 160 bpm, blood pressure of 65/45 mmHg, respiration rate of 66/min, and oxygen saturation of 99%. His weight was 3500 g (3rd-10th p) (as same as the birth weight), height was 51 cm (10th-50th p), and head circumference was 35 cm (10th p). His general condition was poor, he was dehydrated, anterior fontanel was 2x2 cm in size and sunken, and he had sunken globes and reduced skin turgor. Respiratory system examination revealed equal respiratory sounds in both lungs, with no pathological sounds heard. Abdominal examination was unremarkable with no palpable mass. Emergency intervention was made. Patient with severe dehydration was administered physiological saline at a dose of 10 cc/kg 3 times. During follow-up, he was found to have episodes of apnea with accompanying reduced heart rate. Apneas were lasting about 15 seconds and responding to tactile stimulation. Patient was administered oxygen with hood. Blood gas analysis showed pH: 7.50, CO_2: 38 mm/hg, HCO_3: 29.4 mmol/L, and lactate: 12.3 mmol/L; the blood tests showed that sodium: 141 mmol/L, chlorine: 84 mmol/L, acute phase reactants were negative, total bilirubin: 2.2 mg/dl, direct bilirubin: 0.6 mg/dl, white blood cell count: 17.630 mm^3, neutrophil count: 5740 mm^3, lymphocyte count: 9320 mm^3, and hemoglobin: 12.7 g/dl. Urinalysis was normal. Chest x-ray was normal; erect abdominal x-ray revealed increased gastric gas. A magnetic resonance imaging (MRI) was obtained to exclude organic causes of central apnea, which was normal. An abdominal ultasonography was ordered with preliminary diagnosis of pyloric stenosis due to vomiting and hypochloremic metabolic alkalosis. Ultrasonography revealed that "pyloric single-wall thickness was 7 mm, pyloric length 16 mm and double-wall thickness 14 mm, suggesting pyloric stenosis". The patient requiring an emergency surgery was transferred to department of pediatric surgery. The patient whose apneas resolved after surgery was discharged at postoperative day 3 without any additional clinical problem.

DISCUSSION

Although etiology of IHPS has not been clarified, potential major factors are thought to be spasm and compensatory muscular hypertrophy caused by gastric hyperacidity, neurological degeneration or immaturity, and abnormal endocrine signaling (3). Glomerular filtration rate decreases with fluid loss caused by difficulty feeding. HCO_3-compensation ability of the kidney is therefore impaired, as well. To hold potassium, distal renal tubules excrete hydrogen ions, which complicates metabolic alkalosis. With renal compensation, carbon dioxide (CO_2) retention occurs with hypoventilation. Increased partial CO_2 pressure affects pH level in cerebrospinal fluid, stimulating central chemoreceptors and then central apnea develops (4). It is concluded that it will not cause apnea unless accompanied by conditions stimulation chemoreflex like reflux and that apnea occurs due to compensatory respiratory depression and hypoventilation caused by metabolic alkalosis (3). While rate of preoperative apnea was found as 27% in a study, this rate was reported as 5% in another one (4,5). There is no Turkish study regarding frequency of apnea in patients with IHPS. Cases of apnea accompanying IHPS, however, are extremely rare. Thus, as in our patient, IHPS should always be considered in patients with respiratory distress without any palpable mass when distended stomach is found on chest and supine abdominal x-rays, in addition to hypochloremic metabolic alkalosis. As in our patient, apneas do not recur after surgery and the patient can be discharged to home early. Early diagnosis of patients with IHPS, as well as immediate correction of metabolic gap and taking into operation as immediate as possible is crucial for reduction of disease-related mortality and morbidity rates. Despite of a large number of studies on this topic, number of cases of apnea accompanying pyloric stenosis is scent in the literature. We, thus, aimed to report this case to point out pyloric stenosis in patients presenting with episodes of apnea.

Çıkar Çatışması: Çalışmada herhangi bir çıkar çatışması yoktur.

Finansal Çıkar Çatışması: Çalışmada herhangi bir finansal çıkar çatışması yoktur.

Sorumlu Yazar: Abdullah Akkus, Necmettin Erbakan Üniversitesi, Tıp Fakültesi, Çocuk Sağlığı ve Hastalıkları, Konya, Türkiye
e-mail: abdullahakkus29@gmail.com

REFERENCES

1. Uğur C, Ay E, Madenci H. Tekrarlayan Hastane Başvurusuna Rağmen Geç Tanı Alan Nadir Bir Olgu: İnfantil Hipertrofik Pilor Stenozu. Çocuk Dergisi Eylül 2021;21(2):212-4.
2. Altunhan H, Yurtçu M, Tarakçı N, ve ark. (2017). Hipernatremik dehidratasyon ile ilişkili konjenital pilor atrezi. Selçuk Tıp Dergisi 33:(4), 78-9.
3. Pappano Dante MD, MPH. Alkalosis-Induced Respiratory Depression From Infantile Hypertrophic Pyloric Stenosis. Pediatric Emergency Care 2011;27(2):124.
4. van den Bunder FAIM, van Wijk L, van Woensel JBM, et al. Perioperative apnea in infants with hypertrophic pyloric stenosis: A systematic review. Paediatr Anaesth 2020;30(7):749-58.
5. van den Bunder FAIM, van Woensel JBM, Stevens MF, et al. Respiratory problems owing to severe metabolic alkalosis in infants presenting with hypertrophic pyloric stenosis. J Pediatr Surg 2020;55(12):2772-6.