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INSTRUCTIONS FOR AUTHORS

AIM AND SCOPE

The Journal will not consider manuscripts any that have been published elsewhere, or manuscripts that are being considered for another publication, or are in press. Studies previously announced in the congresses are accepted if this condition is stated. If any part of a manuscript by the same author(s) contains any information that was previously published, a reprint or a copy of the previous article should be submitted to the Editorial Office with an explanation by the authors

A technical review is performed to confirm that all of the required documentation has been submitted and to conduct a preliminary evaluation of the manuscript and supplementary files to assess suitability for the Journal. The manuscript will be returned to the Author in the event of any deficiency.

Pediatric Practice and Research Journal operates a blind review process. Contributions deemed suitable are then typically sent to a minimum of two independent expert reviewers in the field of study to assess the scientific quality of the paper.

The Editor/Editors are responsible for the final decision regarding acceptance or rejection of articles. The Editor's decision is final. If necessary, author(s) may be invited to submit a revised version of the manuscript. This invitation does not imply that the manuscript will be accepted for publication. Revised manuscripts must be sent to the Editorial Office within 4 (four) weeks, otherwise they will be considered as a new application. The corresponding author will be notified of the decision to accept or reject the manuscript for publication.

Statements and suggestions published in manuscripts are the authors' responsibility and do not reflect the opinions of the Editor, Associate Editors and the Editorial Board members.

The manuscript will not be returned to the authors whether the article is accepted or not. Copyright fee is not paid for the articles published in the journal. A copy of the journal will be sent to the corresponding author.

Language of the Journal

The official languages of the Journal are Turkish and English. The manuscripts that are written in Turkish have abstracts in English, which makes the abstracts available to a broader audience.

Authorship Criteria

After accepted for publication, all the authors will be asked to sign "Coyright Transfer Form" which states the following: " This work is not under active consideration for publication, has not been accepted for publication, nor has it been published, in full or in part (except in abstract form). I confirm that the study has been approved by the ethics committee. " All authors should agree to the conditions outlined in the form.

Pediatric Practice and Research Journal has agreed to use the standards of the International Committee of Medical Journal Editors. The author(s) should meet the criteria for authorship according to the "Uniform Requirements for Manuscripts Submitted to Biomedical Journals: Writing and Editing for Biomedical Publication. It is available at www.icmje.org.

Ethical Responsibility

The protocol of clinical research articles must be approved by the Ethics Committee.

In all studies conducted on humans, the "Material and Method" section was approved by the relevant committee or the Helsinki Declaration of Principles (https://www.wma.net/what-we-do/medical-ethics/declaration-of-helsinki/).

It should be stated in the text that all persons included in the study signed the am Informed Consent Form ".

The articles submitted to the Pediatric Practice and Research Journal will be deemed to have been conducted in accordance with the Helsinki Declaration of Principles, and have received ethical and legal permissions and will not be held responsible.

If the "Animal" item was used in the study, the authors stated that in the Material and Method section of the article, they protect the animal rights in their studies in accordance with the principles of Guide for the Care and Use of Laboratory Animals (www.nap.edu/catalog/5140.html) and that they have received approval from the ethics committees of their institutions. must specify.

In case reports, Informed Consent a should be obtained from patients regardless of the identity of the patient.

If the article includes the institution (directly or indirectly) providing financial support for the commercial connection or work, the authors; the commercial product used, the drug, the company has no commercial relationship with, or if there is any relationship (consultant, other agreements, etc.), the editor must inform the presentation page.

If Ethics Committee Approval is required in the article; the received document should be sent with the article.

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The manuscript should be submitted to the Academic Plagiarism Prevention Program by the authors.

It is the authors' responsibility to ensure that the article complies with the ethical rules.

Policy of Screening for Plagiarism

The manuscripts are scanned by the Journal using the iThenticate program for determination of plagiarism and non-ethical situations. Pediatric Practice and Research Journal will immediately reject manuscripts leading to plagiarism.

TYPES OF MANUSCRIPT

Manuscripts should be submitted online via www.pprjournal.com

Original Articles should not exceed 3000 words and should be arranged under the headings of Abstract (not more than 300 words), Introduction, Materials and Methods, Results, Discussion, Conclusion and References.

Case Reports should not exceed 1000 words and 10 references, and should be arranged as follows: Abstract, Introduction, Case Report, Discussion and References. It may be accompanied by only one figure or table.

Letter to the Editor should not exceed 500 words. Short relevant comments on medical and scientific issues, particularly controversies, having no more than five references and one table or figure are encouraged. Where letters refer to an earlier published paper, authors will be offered right of reply.

Reviews are not accepted unless written on the invitation of the Editorial Board.

PREPARATION OF MANUSCRIPTS

All articles submitted to the Journal must comply with the following instructions:

a) Submissions should be doubled-spaced and typed in Arial 10 points.

b) All pages should be numbered consecutively in the top right-hand corner, beginning with the title page.

c) The title page should not include the names and institutions of the authors.

d) The manuscript should be presented in the following order: Title page, Abstract (English, Turkish), Keywords (English, Turkish), Introduction, Materials and Methods, Results, Discussion, Conclusion, Acknowledgements (if present), References, Figure Legends, Tables (each table, complete with title and foot-notes, on a separate page) and Appendices (if present) presented each on a separate page.

Title

The title should be short, easy to understand and must define the contents of the article.

Abstract

Abstract should be in both English and Turkish and should consist "Aim, Materials and Methods, Results and Conclusion". The purpose of the study, the setting for the study, the subjects, the treatment or intervention, principal outcomes measured, the type of statistical analysis and the outcome of the study should be stated in this section (up to 300 words). Abstract should not include reference. No abstract is required for the letters to the Editor.

Keywords

Not more than five keywords in order of importance for indexing purposes should be supplied below the abstract and should be selected from Index Medicus Medical Subject Headings (MeSH), available at www.nlm.nih.gov/meshhome.html.

Text

Authors should use subheadings to divide sections regarding the type of the manuscript as described above. Statistical methods used should be specified in the Materials and Methods section.

References

In the text, references should be cited using Arabic numerals in parenthesis in the order in which they appear. If cited only in tables or figure legends, they should be numbered according to the first identification of the table or figure in the text. Names of the journals should be abbreviated in the style used in Index Medicus. The names of all authors should be cited when there are six or fewer; when seven or more, the first three should be followed by et al. The issue and volume numbers of the referenced journal should be added.

References should be listed in the following form:

Journal article

Teke Z, Kabay B, Aytekin FO et al. Pyrrolidine dithiocarbamate prevents 60 minutes of warm mesenteric ischemia/reperfusion injury in rats. Am J Surg 2007;194(6):255-62.

Supplement

Solca M. Acute pain management: Unmet needs and new advances in pain management. Eur J Anaesthesiol 2002; 19(Suppl 25): 3-10.

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Online article not yet published in an issue

Butterly SJ, Pillans P, Horn B, Miles R, Sturtevant J. Off-label use of rituximab in a tertiary Queensland hospital. Intern Med J doi: 10.1111/j.1445-5994.2009.01988.x

Book

Sample1: Murray PR, Rosenthal KS, Kobayashi GS, Pfaller MA. Medical microbiology. 4th ed. St. Louis: Mosby; 2002.

Sample 2: Sümbüloğlu K, Akdağ B. Regresyon Yöntemleri ve Korelasyon Analizi. Hatiboğlu Yayınevi: Ankara; 2007.

Chapter in a book

Meltzer PS, Kallioniemi A, Trent JM. Chromosome alterations in human solid tumors. I n: Vogelstein B, Kinzler KW, editors. The genetic basis of human cancer. New York: McGraw-Hill; 2002. p. 93113.

Journal article on the Internet

Abood S. Quality improvement initiative in nursing homes: The ANA acts in an advisory role. Am J Nurs [serial on the Internet] 2002 [cited 12 Aug 2002]; 102. Available from: www.nursingworld.org/AJN/2002/june/wawatch.htm

Website

Cancer-pain.org [homepage on the Internet]. New York: Association of Cancer Online Resources [updated 16 May 2002; cited 9 Jul 2002]. Available from: www.cancer-pain.org

An organization as an author

The Intensive Care Society of Australia and New Zealand. Mechanical ventilation strategy in ARDS: Guidelines. Int Care J Aust 1996;164:282-4.

Acknowledgements

The source of financial grants and the contribution of colleagues or institutions should be acknowledged.

Tables

Tables should be complementary, but not duplicate information contained in the text. Tables should be numbered consecutively in Arabic numbers, with a descriptive, self-explanatory title above the table. All abbreviations should be explained in a footnote. Footnotes should be designated by symbols in the following order: *,†, ‡, §, ¶.

Figures

All illustrations (including line drawings and photographs) are classified as figures. Figures must be added to the system as separate .jpg or .gif files (approximately 500x400 pixels, 8 cm in width and at least 300 dpi resolution). Figures should be numbered consecutively in Arabic numbers and should be cited in parenthesis in consecutive order in the text.

Figure Legends

Legends should be self-explanatory and positioned on a separate page. The legend should incorporate definitions of any symbols used and all abbreviations and units of measurements should be explained. A letter should be provided stating copyright authorization if figures have been reproduced from another source.

Measurements and Abbreviations

All measurements must be given in metric system (Système International d'Unités, SI). Example: mg/kg, µg/kg, mL, mL/kg, mL/kg/h, mL/kg/min, L/min, mmHg, etc. Statistics and measurements should always be given in numerals, except where the number begins a sentence. When a number does not refer to a unit of measurement, it is spelt out, except where the number is greater than nine.

Abbreviations that are used should be defined in parenthesis where the full word is first mentioned. Some common abbreviations can be used, such as iv, im, po, and sc.

Drugs should be referred to by their generic names, rather than brand names.

Editorial Correspondence

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YAZARLARA BİLGİ

AMAÇ ve KAPSAM

Pediatric Practice and Research Dergisi, dört ayda bir yayımlanır ve üç sayı ile bir cilt tamamlanır. Dergi; pediatri ile ilgili tüm nitelikli klinik ve deneysel araştırmaları, olgu sunumlarını ve editöre mektupları yayımlar.

Pediatric Practice and Research Dergisi, bilimsel yayınlara açık erişim sağlar. Dergi basımından hemen sonra, makalelerin tam metinlerine ücretsiz ulaşılabilir.

Dergide yayımlanmak üzere gönderilen yazıların daha önce başka bir yerde yayımlanmamış veya yayımlanmak üzere gönderilmemiş olması gerekir. Daha önce kongrelerde sunulmuş çalışmalar, bu durum belirtilmek koşuluyla kabul edilir. Makale, yazar(lar) ın daha önce yayımlanmış bir yazısındaki konuların bir kısmını içeriyorsa bu durum belirtilmeli ve yeni yazı ile birlikte önceki makalenin bir kopyası da Yayın Bürosu'na gönderilmelidir.

Gerekli tüm belgelerin sunulduğunu teyit etmek ve dergiye uygunluğunu değerlendirmek için makale ve ek dosyaların ön değerlendirmesini yapmak üzere teknik bir inceleme yapılır. Herhangi bir eksiklik olması halinde makale yazara iade edilecektir. Pediatric Practice and Research Dergisi kör bir inceleme süreci yürütmektedir. Uygun görülen yazılar daha sonra makalenin bilimsel kalitesini değerlendirmek için çalışma alanında en az iki bağımsız uzmana gönderilir. Editör / Editörler makalelerin kabulü veya reddi ile ilgili nihai karardan sorumludur.

Editörün kararı kesindir. Gerekli olduğu durumlarda, yazar(lar) dan düzeltme istenebilir. Yazardan düzeltme istenmesi, yazının yayımlanacağı anlamına gelmez. Bu düzeltmelerin en geç 21 gün içinde tamamlanıp dergiye gönderilmesi gereklidir. Aksi halde yeni başvuru olarak değerlendirilir. Sorumlu yazara yazının kabul veya reddedildiğine dair bilgi verilir.

Dergide yayımlanan yazıların etik, bilimsel ve hukuki sorumluluğu yazar(lar)a ait olup Editör, Editör Yardımcısı ve Yayın Kurulu'nun görüşlerini yansıtmaz.

Dergide yayımlanması kabul edilse de edilmese de, yazı materyali yazarlara geri verilmez. Dergide yayımlanan yazılar için telif hakkı ödenmez. Bir adet dergi, sorumlu yazara gönderilir.

Derginin Yazı Dili

Derginin yazı dili Türkçe ve İngilizcedir. Dili Türkçe olan yazılar, İngilizce özetleri ile yer alır. Yazının hazırlanması sırasında, Türkçe kelimeler için Türk Dil Kurumundan (www.tdk.gov.tr), teknik terimler için Türk Tıp Terminolojisinden (www.tipterimleri.com) yararlanılabilir.

Yazarlık Kriterleri

Dergide yayınlanması uygun bulunan tüm yazıların araştırma ve yayın etiğine uygun hazırlandığı, varsa sağlanan fonun kaynağının tanımlandığı, başka yerde yayımlanmadığı veya yayımlanmak üzere gönderilmediği, çalışmaya katılan tüm yazarlar tarafından yazının son halinin onaylandığı, yayımlanacak yazı ile ilgili telif haklarının dergiye devredildiği, tüm yazarların imzaları ile "Yayın Hakkı Devir Formu"nda belirtilmesi gerekir.

Pediatric Practice and Research Dergisi, Uluslararası Tıp Dergileri Editörleri Kurulu'nun (International Committee of Medical Journal Editors) "Biyomedikal Dergilere Gönderilen Makalelerin Uyması Gereken Standartlar: Biyomedikal Yayınların Yazımı ve Baskıya Hazırlanması (Uniform Requirements for Manuscripts Submitted to Biomedical Journals: Writing and Editing for Biomedical Publication)" standartlarını kullanmayı kabul etmektedir. Bu konudaki bilgiye www.icmje.org adresinden ulaşılabilir.

Etik Sorumluluk

Etik Sorumluluk / Kurallar: Klinik araştırma makalelerinin protokolü Etik Komitesi tarafından onaylanmış olmalıdır.

İnsanlar üzerinde yapılan tüm çalışmalarda "Gereç ve Yöntem" bölümünde çalışmanın ilgili komite tarafından onaylandığı veya çalışmanın Helsinki İlkeler Deklarasyonu'na (https://www.wma.net/what-we-do/medical-ethics/declaration-of-helsinki/) uyularak gerçekleştirildiğine dair bir cümle yer almalıdır.

Çalışmaya dahil edilen tüm kişilerin Bilgilendirilmiş Onam Formu'nu imzaladığı metin içinde belirtilmelidir.

Pediatric Practice and Research Dergisi'ne gönderilen makalelerdeki çalışmaların Helsinki İlkeler Deklarasyonu'na uygun olarak yapıldığı, kurumsal etik ve yasal izinlerin alındığı varsayılacak ve bu konuda sorumluluk kabul edilmeyecektir.

Çalışmada "Hayvan" öğesi kullanılmış ise yazarlar, makalenin Gereç ve Yöntem bölümünde hayvan haklarını Guide for the Care and Use of Laboratory Animals (www.nap.edu/catalog/5140.html) prensipleri doğrultusunda koruduklarını, çalışmalarında ve kurumlarının etik kurullarından onay aldıklarını belirtmek zorundadır.

Olgu sunumlarında hastanın kimliğinin ortaya çıkmasına bakılmaksızın hastalardan "Bilgilendirilmiş rıza" alınmalıdır.

Makalede ticari bağlantı veya çalışma için maddi destek veren kurum (doğrudan veya dolaylı) mevcut ise yazarlar; kullanılan ticari ürün, ilaç, firma ile ticari hiçbir ilişkisinin olmadığını veya varsa nasıl bir ilişkisinin olduğunu (konsültan, diğer anlaşmalar vs.), editöre sunum sayfasında bildirmek zorundadır.

Makalede Etik Kurul Onayı alınması gerekli ise; alınan belge makale ile birlikte gönderilmelidir.

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Makale yazarlar tarafından akademik intihal önleme programından geçirilmelidir.

Makalenin etik kurallara uygunluğu yazarların sorumluluğundadır.

İntihal Taraması Politikası

Makaleler, intihal ve etik olmayan durumların belirlenmesi için iThenticate programı kullanılarak Journal tarafından taranır. Pediatric Practice and Research Dergisi intihallere yol açan makaleleri derhal reddedecektir.

YAZI TÜRLERİ

Yazılar, elektronik ortamda www.pprjournal.com adresine gönderilir.

Orijinal makaleler, 3000 sözcük sayısını aşmamalı, "Öz (en fazla 300 kelime), Giriş, Gereç ve Yöntem, Bulgular, Tartışma, Sonuç, Kaynaklar" bölümlerinden oluşmalıdır.

Olgu Sunumu, "Öz, Giriş, Olgu Sunumu, Tartışma, Kaynaklar" şeklinde düzenlenmelidir. En fazla 1000 sözcük ile sınırlıdır. Sadece bir tablo veya şekil ile desteklenebilir.

Editöre Mektup, yayımlanan metinlerle veya mesleki konularla ilgili olarak 500 sözcüğü aşmayan ve beş kaynak ile bir tablo veya şekil içerecek şekilde yazılabilir. Ayrıca daha önce dergide yayınlanmış metinlerle ilişkili mektuplara cevap hakkı verilir.

Yayın Kurulu'nun daveti üzerine yazılanlar dışında derleme kabul edilmez.

MAKALENİN HAZIRLANMASI

Dergide yayınlanması istenilen yazı için aşağıdaki kurallara uyulmalıdır.

a) Yazı; iki satır aralıklı olarak, Arial 10 punto ile yazılmalıdır.
b) Sayfalar başlık sayfasından başlamak üzere, sağ üst köşesinde numaralandırılmalıdır.
c) Online makale sistemine yüklenen word dosyasının başlık sayfasında (makalenin adını içeren başlık sayfası), yazarlara ait isim ve kurum bilgileri yer almamalıdır.
d) Makale, şu bölümleri içermelidir: Her biri ayrı sayfada yazılmak üzere; Türkçe ve İngilizce Başlık Sayfası, Öz, Abstract, Anahtar Sözcükler, Keywords, Giriş, Gereç ve Yöntem, Bulgular, Tartışma, Sonuç, Açıklamalar (varsa), Kaynaklar, Şekil Alt Yazıları, Tablolar (başlıkları ve açıklamalarıyla beraber), Ekler (varsa).

Yazının Başlığı

Kısa, kolay anlaşılır ve yazının içeriğini tanımlar özellikte olmalıdır.

Özetler

Türkçe (Öz) ve İngilizce (Abstract) olarak yazılmalı, Amaç, Gereç ve Yöntem, Bulgular ve Sonuç (Aim, Materials and Methods, Results, Conclusion) olmak üzere dört bölümden oluşmalı, en fazla 300 sözcük içermelidir. Araştırmanın amacı, yapılan işlemler, gözlemsel ve analitik yöntemler, temel bulgular ve ana sonuçlar belirtilmelidir. Özette kaynak kullanılmamalıdır. Editöre mektup için özet gerekmemektedir.

Anahtar Sözcükler

Türkçe Öz ve İngilizce Abstract bölümünün sonunda, Anahtar Sözcükler ve Keywords başlığı altında, bilimsel yazının ana başlıklarını yakalayan, Index Medicus Medical Subject Headings (MeSH)'e uygun olarak yazılmış en fazla beş anahtar sözcük olmalıdır. Anahtar sözcüklerin, Türkiye Bilim Terimleri'nden (www.bilimterimleri.com) seçilmesine özen gösterilmelidir.

Metin

Yazı metni, yazının türüne göre yukarıda tanımlanan bölümlerden oluşmalıdır. Uygulanan istatistiksel yöntem, Gereç ve Yöntem bölümünde belirtilmelidir.

Kaynaklar

Pediatric Practice and Research Dergisi, Türkçe kaynaklardan yararlanmaya özel önem verdiğini belirtir ve yazarların bu konuda duyarlı olmasını bekler.

Kaynaklar metinde yer aldıkları sırayla, cümle içinde atıfta bulunulan ad veya özelliği belirten kelimenin hemen bittiği yerde ya da cümle bitiminde noktadan önce parantez içinde Arabik rakamlarla numaralandırılmalıdır. Metinde, tablolarda ve şekil alt yazılarında kaynaklar, parantez içinde Arabik numaralarla nitelendirilir. Sadece tablo veya şekil alt yazılarında kullanılan kaynaklar, tablo ya da şeklin metindeki ilk yer aldığı sıraya uygun olarak numaralandırılmalıdır. Dergi başlıkları, Index Medicus'ta kullanılan tarza uygun olarak kısaltılmalıdır. Kısaltılmış yazar ve dergi adlarından sonra nokta olmamalıdır. Yazar sayısı altı veya daha az olan kaynaklarda tüm yazarların adı yazılmalı, yedi veya daha fazla olan kaynaklarda ise üç yazar adından sonra et al. veya ve ark. yazılmalıdır. Kaynak gösterilen derginin sayı ve cilt numarası mutlaka yazılmalıdır.

Kaynaklar, yazının alındığı dilde ve aşağıdaki örneklerde görüldüğü şekilde düzenlenmelidir.

Dergilerdeki yazılar

Teke Z, Kabay B, Aytekin FO et al. Pyrrolidine dithiocarbamate prevents 60 minutes of warm mesenteric ischemia/reperfusion injury in rats. Am J Surg 2007;194(6):255-62.

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Ek sayı (Supplement)
Solca M. Acute pain management: Unmet needs and new advances in pain management. Eur J Anaesthesiol 2002;19(Suppl 25):3-10.

Henüz yayınlanmamış online makale
Butterly SJ, Pillans P, Horn B, Miles R, Sturtevant J. Off-label use of rituximab in a tertiary Queensland hospital. Intern Med J doi: 10.1111/j.1445-5994.2009.01988.x

Kitap
Örnek 1: Murray PR, Rosenthal KS, Kobayashi GS, Pfaller MA. Medical microbiology. 4th ed. St. Louis: Mosby; 2002.
Örnek 2: Sümbüloğlu K, Akdağ B. Regresyon Yöntemleri ve Korelasyon Analizi. Hatiboğlu Yayınevi: Ankara; 2007.

Kitap bölümü
Meltzer PS, Kallioniemi A, Trent JM. Chromosome alterations in human solid tumors. I n: Vogelstein B, Kinzler KW, editors. The genetic basis of human cancer. New York: McGraw-Hill; 2002. p. 93113.

İnternet makalesi
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Yazar olarak bir kuruluş
The Intensive Care Society of Australia and New Zealand. Mechanical ventilation strategy in ARDS: Guidelines. Int Care J Aust 1996;164:282-4.

Açıklamalar
Varsa finansal kaynaklar, katkı sağlayan kurum, kuruluş ve kişiler bu bölümde belirtilmelidir.

Tablolar
Tablolar metni tamamlayıcı olmalı, metin içerisinde tekrarlanan bilgiler içermemelidir. Metinde yer alma sıralarına göre Arabik sayılarla numaralandırılıp tablonun üstüne kısa ve açıklayıcı bir başlık yazılmalıdır. Tabloda yer alan kısaltmalar, tablonun hemen altında açıklanmalıdır. Dipnotlarda sırasıyla şu semboller kullanılabilir: *, †, ‡, §, ¶.

Şekiller
Şekil, resim, grafik ve fotoğrafların tümü "Şekil" olarak adlandırılmalı ve ayrı birer .jpg veya .gif dosyası olarak (yaklaşık 500x400 piksel, 8 cm eninde ve en az 300 dpi çözünürlükte) sisteme eklenmelidir. Şekiller metin içinde kullanım sıralarına göre Arabik rakamla numaralandırılmalı ve metinde parantez içinde gösterilmelidir.

Şekil Alt Yazıları
Şekil alt yazıları, her biri ayrı bir sayfadan başlayarak, şekillere karşılık gelen Arabik rakamlarla çift aralıklı olarak yazılmalıdır. Şeklin belirli bölümlerini işaret eden sembol, ok veya harfler kullanıldığında bunlar alt yazıda açıklanmalıdır. Başka yerde yayınlanmış olan şekiller kullanıldığında, yazarın bu konuda izin almış olması ve bunu belgelemesi gerekir.

Ölçümler ve Kısaltmalar
Tüm ölçümler metrik sisteme (Uluslararası Birimler Sistemi, SI) göre yazılmalıdır. Örnek: mg/kg, µg/kg, mL, mL/kg, mL/kg/h, mL/kg/min, L/min, mmHg, vb. Ölçümler ve istatistiksel veriler, cümle başında olmadıkları sürece rakamla belirtilmelidir. Herhangi bir birimi ifade etmeyen ve dokuzdan küçük sayılar yazı ile yazılmalıdır.

Metin içindeki kısaltmalar, ilk kullanıldıkları yerde parantez içinde açıklanmalıdır. Bazı sık kullanılan kısaltmalar; iv, im, po ve sc şeklinde yazılabilir.

İlaçların yazımında jenerik isimleri kullanılmalıdır.

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ORIGINAL ARTICLE
ORİJİNAL ARAŞTIRMA

Evaluation of Children Presenting to the Emergency Department with Iron Intoxication

Acil Servise Demir Zehirlenmesi İle Başvuran Çocuk Vakalarının Değerlendirilmesi

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ABSTRACT

Purpose: Intoxications is a common and preventable cause of childhood morbidity and mortality. Acute iron intoxication usually occurs in children under 5-year-old due to accidental ingestion, and composes 2% of all intoxication cases in children and adolescents. In this study, demographic, epidemiological, clinical features, treatments, and complications of the patients admitted to our emergency department with acute iron intoxication were evaluated.

Material and Method: Patients admitted to a Pediatric Emergency Service of the Faculty of Medicine diagnosed with iron intoxication between 2018 and 2020 were retrospectively investigated. The patients' demographic characteristics, information about intoxication, and laboratory results were recorded, and statistical analyzes were performed.

Results: Of the 12 patients included in the study, 66.7% were female, and 33.3% were male. The mean age was 81.3±83.52 months. When separated by age group, 66.6% of the patients were younger than 5-year-old. Those who came to the hospital via an ambulance were only 25%. All patients were transported to the hospital within an average of 40±15 minutes. While there was no life-threatening risk in 7 patients, the condition of 5 patients was severe. Only 2 of the patients took it to suicide. All cases received iron orally. One patient presented abdominal pain, and 2 patients presented nausea and vomiting. Activated charcoal was administered to 4 of the patients. In the laboratory follow-ups of the patients, all mean results, excluding iron, were normal. In addition, the blood gases of the patients at the time of admission were compensated metabolic acidosis.

Conclusion: As a pediatric emergency, iron intoxication in children remains important as one of the preventable morbidity and mortality causes. We believe that iron preparations are packaged in a single-dose form and do not have an attractive taste and appearance for children, have protective caps in medicine boxes, and when physicians prescribe iron drugs to adult patients, warning them about toxicity in children will reduce mortality and morbidity.

Keywords: Children, iron intoxication, emergency

ÖZ

Amaç: Bu çalışma ile oral demir alımı sonrası intoksikasyon nedeniyle başvuran hastaların; demografik, epidemiyolojik, klinik özellikleri, tedavileri ve komplikasyonlarının geriye dönük olarak değerlendirilmesi neticesinde ülkemiz çocukluk çağı demir zehirlenmeleri verilerine katkı sağlamak amaçlandı.

Gereç ve Yöntem: 2018-2020 yılları arasında demir intoksikasyonu tanısı ile bir Tıp Fakültesi Çocuk Acil Polikliniği'ne başvurmuş hastalar retrospektif olarak tarandı. Hastaların demografik özellikleri, intoksikasyona ait bilgiler ve labaratuvar tetkik sonuçları kayıt altına alınarak istatistiksel analizleri gerçekleştirildi.

Bulgular: Çalışmaya dahil edilen 12 hastanın %66,7'si kadın %33,3'ü erkekti. Ortalama yaş 81,3± 83,52 aydı. Yaş gruplarına göre ayrıldıklarında olguların %66,6'sı 5 yaşından küçük çocuklar olarak gözlendi. Hastaneye bir ambulans yardımı ile gelenler sadece %25'ini oluşturmaktaydı. Ortalama 40±15 dk içerisinde tüm hastaların hastaneye nakli gerçekleştirilmiştir. 7 hastanın hayati bir riski bulunmazken, 5 hastanın durumu ciddi idi. Hastalardan sadece 2 tanesi suisid amaçlı zehirlenmişti. Tüm vakalar demiri oral yoldan almıştır. 1 hastada karın ağrısı, 2 hasta da bulantı, kusma oluşmuştur. Hastalardan 4'üne tedavide aktif kömür uygulandı. Hastaların laboratuvar takiplerinde demir dışındaki diğer tüm değerlerin ortalama sonuçları normal bulundu. Ayrıca hastaların başvuru esnasındaki kan gazları kompanse metabolik sendrom şeklindedir.

Sonuç: Pediatrik acil olarak; çocuklarda demir intoksikasyonu, önlenebilir morbidite ve mortalitenin nedenlerinden birisi olarak halen önemini korumaktadır. Demir preparatlarının tek doz şeklinde paketlenmesinin yanı sıra cocukların cezbedici tat ve görünümünde olmaması, koruyucu kapakların ilaç kutularında mutlaka kullanılması, hekimlerin demir ilaçlarını reçete ettiği yetişkin hastalarını çocuklardaki toksisite açısından uyararak gerekli bilgileri vermesi mortalite ve morbiditeyi azaltacağı kanaatindeyiz.

Anahtar Kelimeler: Demir, zehirlenme, acil servis

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INTRODUCTION

Intoxications, a common and preventable cause of childhood morbidity and mortality, are among the most important reasons for admission to pediatric emergency services and hospitalizations in our country, including in developed countries worldwide(1).

Acute iron intoxication usually occurs in children under 5-year-old due to accidental ingestion, and it is less common in adolescents and adults. The American Association of Poison Control Centers reported in 2004 that 2% of all intoxication cases in children and adolescents are iron poisoning (2). The main reasons are the widespread availability of iron-containing dietary supplements, the tablets' sugar-like appearance, and parents' lack of potential toxicity awareness (1-4). A study showed that only one-third of parents keep their iron-containing drugs in areas out of reach of children (5).

The excess amount of iron taken orally affects the gastrointestinal system barrier via its direct caustic effect. As a result, massive iron absorption occurs. When the serum iron level exceeds the binding capacity, free radicals emerge, and lipid peroxidation and cell destruction occur. Although toxicity mainly affects the liver, kidney, heart, lungs, and hematological system are also adversely affected (4-6). The toxicity severity depends on the amount of iron taken. The toxicity risk is generally low, below 20 mg/kg. Decontamination is recommended, with patient monitoring for 6 hours. There is a moderate toxicity risk in iron intake of 20-40 mg/kg. Chelation therapy should be considered in addition to decontamination. Doses above 60 mg/kg are regarded as high risk, and chelation therapy should be started in addition to decontamination (6).

In this study, we aimed to contribute to the data on childhood iron intoxication with the retrospective evaluation of demographic, epidemiological, clinical features, treatments, and complications of the patients who applied due to intoxication after oral iron intake in our emergency department.

MATERIAL AND METHOD

The study was carried out with the permission of Selçuk University Ethics Committee (Date: 29.03.2022, Decision No: 2022/151). All procedures were carried out in accordance with the ethical rules and the principles of the Declaration of Helsinki.

This study is retrospective. Patients between 1 month and 18 years who were diagnosed with iron intoxication and followed up in the Pediatric Emergency Service of Selcuk University Faculty of Medicine between January 1, 2018, and June 30, 2020, were examined and included. Patients with missing data, incomplete diagnostic codes, and intoxication with non-ferrous and/or other drugs were excluded from the study. Ethics committee approval was obtained.

In addition to demographic characteristics of the patients such as age, gender, presence of chronic diseases, many intoxication-related factors such as the year, season, admission time to the hospital, the way and duration of admission, the presence of additional comorbidities, intoxication types (suicide, accident, substance, etc.), consciousness status were included in the examination. In addition to this, laboratory examinations and the treatments administered are included.

Statistical Analysis

All data were evaluated using the SPSS 21.0 statistical package program. The normal distribution of the variables was studied using visual (histogram and probability graphs) and analytical methods (Kolmogorov-Smirnov/ Shapiro-Wilk tests). Descriptive analyzes were given using mean and standard deviation values for normally distributed variables and median values for non-normally distributed variables. The results were evaluated at the 95% confidence interval, and the significance level was $p<0.05$. Number, percentage, mean and standard deviation were used to assess the data.

RESULTS

Of 12 patients who met the criteria of our study between 2018 and 2020 and applied to the pediatric emergency department due to acute iron intoxication, 66.7% were female, and 33.3% were male. The mean age was 81.3±83.52 (12-215) months. When separated by age group, 66.6% of the patients were children younger than 5-year-old. Sociodemographic characteristics of iron intoxication patients are shown in **Table 1**.

It was observed that the patients mainly applied in the spring (41.6%) of the year. Intoxication occurred mainly between 16:00-23:59 hours. Those who came to the hospital via an ambulance were only 25%. All patients were transferred to the hospital within an average of 40±15 minutes (**Table 1**).

While the reason for the admission of three patients was the presence of the symptom, the other 9 patients were admitted as forensic cases. As a result, 7 (58.3%) patients were discharged after patient monitoring in the emergency department. 4 patients were hospitalized, and 1 patient was referred (**Table 1**).

Headache, dizziness, fainting, convulsions, palpitations, arrhythmias, respiratory distress, cough, fever, color change in the mouth, bruising, bleeding, paresthesia, and arrest were not observed. In addition, CK-MB and troponin markers were evaluated in a normal range. All patients were conscious at the time of admission. Again, no changes in consciousness were observed in all of them. According to the previous medical records, only 1 of the patients had a history of hematological disease.

There was no life-threatening risk in 7 patients, but, 5 patients were severe. Only 2 of the all patients were poisoned to suicide. All cases received iron orally. One patient presented abdominal pain, and 2 patients presented nausea and vomiting. Activated charcoal was administered to 4 patients.

When we examined the laboratory results of our study, the average results of all values except iron were normal. In addition, when the blood gas was analyzed, the pH was 7.40±0.04. According to these results, the patients' blood gases were compensated metabolic acidosis at the admission. The laboratory findings of the patients applying to the pediatric emergency department after iron intoxication are shown in **Table 2**.

DISCUSSION

Acute iron intoxication is one of the most important toxicities in childhood. Patients may be hypovolemic due to fluid loss and GI bleeding due to irritation. As a result of iron absorption, systemic toxicity findings may occur with the formation of free radicals. Free radicals causing disruption in oxidative phosphorylation, mitochondrial dysfunction, and cell death can be observed in severe poisonings (7).

In patients with digestive system symptoms, gastric lavage with ample hydration and gastrointestinal decontamination methods should be applied. In addition to iron intake, there are symptoms and signs such as lethargy, hypovolemia, persistent vomiting, shock, diarrhea, metabolic acidosis, or a serum iron level of 500 µg/dl and above. Chelation therapy with desferrioxamine should be applied (6-8). Since all patients in our study had a serum iron level of less than 500 µg/dl and had mild signs of poisoning, the patients were usually discharged after being kept under monitoring for 48 hours.

In many studies, it has been reported that poisoning is observed mainly between 1-5-year-old. A survey conducted on 2482041 poisoning cases in the USA in 2007 reported that 51.23% of them were children younger than five-year-old (9). In a study conducted in Greece, poisoning cases under five-year-old were 93% of the patients (10). The survey conducted by Sümer et al. on 233 patients who applied to the emergency department reported that 73.8% were children under 5-year-old (11). In addition to the low consciousness level in this age group, children are curious and try to recognize the world, especially with their sense of taste, which is why poisoning is more common before 5-year-old. The result of our study is similar to studies in Turkey and other countries, with 66.6% of poisonings occurring in children under 5-year-old.

In drug intoxication studies conducted in adults, the cause of intoxication is suicide in over 80%, while this rate drops below 50% in children (9,11,12). As a matter of fact, in our study, iron intake to suicide was very low (16.6%).

Table 1. Sociodemographic Characteristics of Iron Intoxication Patients

Parameters	(N)	(%)
Gender		
Female	8	66.7
Male	4	33.3
Categorical age (month)		
Between 0-60 months	8	66.6
Between 121-180 months	2	16.7
180 months and older	2	16.7
Admission season		
Spring	5	41.6
Summer	2	16.7
Fall	2	16.7
Winter	3	25
Time of cases		
08:00-15:59	2	16.7
16:00-23:59	8	66.6
00:00-07:59	2	16.7
First applied hospital		
Yes	9	75
No	3	25
Admission type		
Without ambulance	9	75
With ambulance	3	25
Activated charcoal application		
Yes, administered	4	33.3
No, not administered	8	66.7
Application result		
Discharge after patient monitoring in emergency service	7	58.3
Hospitalized	4	33.3
Referring	1	8.4

Table 2: Laboratory Findings of the Patients Presenting to the Pediatric Emergency Department after Iron Intoxication

Parameters	Mean±standart deviation (min. – max.)
Iron (µg.dl^{-1})	223.25±167.33 (37-411)
Alanine transaminase (U/L)	18.08±8.36 (9-34)
Aspartatetransaminase (U/L)	33.75±10.38 (16-47)
Hemoglobin (g/dL)	12.78±1.62 (10.90-15.80)
Hemotocrit (%)	38.13±4.70 (32.40-46.80)
Leukocyte (mm^3)	10.34±2.52 (6.30-14.40)
Lymphocyte (K/uL)	3.92±1.73 (2.30-8.28)
Thrombocyte (K/uL)	310.83±84.33 (177-477)
Glucose (mg/dL)	95.40±16.66 (78-128)
Urea (mg/dL)	27.84±7.58 (13.0-40.0)
Creatinin (mg/dL)	0.37±0.16 (0.22-0.69)
Uric acid (mg/dL)	4.18±1.05 (3.10-6.40)
Aptt (sec)	30.96±6.86 (26.00-41.40)
PT-INR (INR)	1.01±0.04 (0.96-1.06)
Sodium (mEq/L)	137.00±1.65 (134-140)
Potassium (mmol/L)	4.11±0.34 (3.46-4.83)
Calcium (mg/dL)	9.94±0.43 (9.30-10.70)
Magnesium (mg/dL)	2.22±0.14 (2.02-2.47)
Sedimentation (mm/saat)	5.20±5.21 (2.0-14.0)
C-reactive protein (mg/L)	1.85±1.73 (0.11-5.12)

Aptt: Activated partial thromboplastin time, PT-INR: Protrombin time- International Normalized Ratio

66.3% of our patients are girls. While all of the cases over 5-year-old were girls, if they were under 5-year-old, this ratio was equal for boys and girls. According to the literature, it was seen that accidental intoxication cases were more common in boys, and self-destructive intoxications were more common in girls. In addition, our results are compatible with the literature as we mainly observe accidental intoxication cases under the age of 5 years (12-14).

In their study, Akın et al. demonstrated that 96.1% of poisonings occur at home, and 96.1% occur orally (15). On the other hand, Ödek et al. found that 91.3% of poisonings occurred orally, and 89.1% occurred at home (12). We also found that all patients in our study had iron poisoning either orally or at home. This situation was considered that iron drugs at home and taken orally caused most of the poisonings.

The effects of iron poisoning are examined in four stages in the clinic. The first stage is gastrointestinal irritations such as nausea-vomiting. It occurs approximately 6 hours after ingesting the drug. Hypotension and metabolic acidosis develop with increased capillary permeability in the second stage. In the third stage, due to the cytotoxic effects of iron, organ failures such as kidney and liver failure are observed. Patients in the fourth stage, the last stage, have a higher risk of gastrointestinal bleeding. (16) In the first 6 hours of patient monitoring, if the patient's iron level is below 20 mg/kg and the patient is asymptomatic, there is usually no need for treatment. (17) The patients in our study had normal blood values and showed mild symptoms. Thus, the average blood gas results of the patients were compensated metabolic acidosis.

After admission to the pediatric emergency department, diagnosis and treatment of poisoning cases are usually carried out (15-42%) in pediatric emergency services. In our study, 33.3% of the patients were hospitalized and continued treatment in the pediatric emergency department (12-14). As the patients in our study did not have high-risk iron intoxication and the poisoning symptoms were mild, an intensive care unit was not required.

Activated charcoal administration is recommended for patients with an early diagnosis of iron intoxication. Activated charcoal was administered to 33.3% of the cases in our study. It was found that oral activated charcoal was administered at 42.7% to 48% in the study of Akgül et al., while this rate was 72.8% in the study of Ödek et al. (12,13). The relatively lower rate in our study was attributed to the fact that the admission time to the emergency department after poisoning was longer than 60 minutes on average.

Limitations

Our study has a relatively small sample size, and larger sample size will be much more meaningful in terms of results. One of the most important reasons for the small sample size may be excluded patients with multiple drug intakes and iron-containing nutritional supplements from our study. In addition, the retrospective nature of our research and the inability to adequately determine the time between iron intake and transportation to the emergency room are other significant limitations.

CONCLUSION

As a pediatric emergency, iron intoxication in children still maintains its importance as one of the preventable morbidity and mortality causes. We believe that iron preparations are packaged in a single-dose form and do not have an attractive taste and appearance for children, have protective caps in medicine boxes, and when physicians prescribe iron drugs to adult patients, warning them about toxicity in children will reduce mortality and morbidity.

ETHICAL DECLARATIONS

Ethics Committee Approval: The study was carried out with the permission of Selçuk University Ethics Committee (Date: 29.03.2022, Decision No: 2022/151).

Informed Consent: Because the study was designed retrospectively, no written informed consent form was obtained from patients.

Referee Evaluation Process: Externally peer-reviewed.

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ORIGINAL ARTICLE
ORİJİNAL ARAŞTIRMA

The Effect of Obesity on Emergence Agitation in Children Undergoing General Anesthesia

Çocuklarda Obezitenin Postoperatif Derlenme Ajitasyonuna Etkisi

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ABSTRACT

Aim: Although the factors affecting emergence agitation (EA) have been examined in the literature, the literature evaluating the effect of obesity on postoperative EA in children is limited. In this study, we aimed to evaluate whether obesity has an effect on postoperative EA.

Material and Method: The medical records of patients aged 2-14 years, with ASA I-II physical status and undergoing elective surgery were reviewed (November 2018 and November 2022). Patients who underwent emergency surgery or who used an anesthetic agent other than sevoflurane for the maintenance of anesthesia were excluded from the study. Patients will be divided into 3 groups according to their body mass index; Group 1; Normal (5th to 85% percentile), Group 2; Overweight (85-95% percentile), Group 3; Obese (95th percentile and above). Emergence agitation was assessed by anesthesiologists using the Watcha scale.

Results: 567 patients were included in the study and EA occurred in 115 (20.3%) of them. Of the patients, 428 were considered normal, 64 were overweight and 75 were obese. When demographic data were compared, there was no statistical difference between the groups. EA incidence was statistically higher in Group II (28.1%) and Group III (29.3%)compared to Group I (17.5%) (p=0.006).

Conclusion: We are of the opinion that both overweight and obesity increase the incidence of EA in children undergoing general anesthesia, but prospective further studies are also required.

Keywords: Emergence agitation, general anesthesia, pediatric

ÖZ

Amaç: Literatürde derlenme ajitasyonuna etki eden faktörler incelenmiş olmakla beraber çocuklarda obezitenin postoperatif derlenme ajitasyonuna etkisinin değerlendirildiği literatür bilgisi kısıtlıdır. Bu çalışmada obezitenin postoperatif derlenme ajitasyonuna etkisinin olup olmadığını değerlendirmeyi amaçladık.

Gereç ve Yöntem: 2-14 yaş arasında, ASA I-II fiziksel statusa sahip, Kasım 2018 ve Kasım 2022 tarihleri arasında elektif cerrahi geçiren hastaların kayıtları incelendi. Acil cerrahi geçiren veya anestezi idamesinde sevofluran dışında bir anestezik ajan kullanılan hastalar çalışma dışında bırakıldı. Hastalar vücut kitle indekslerine göre 3 gruba ayrıldı: Grup 1; Normal (%5 ila %85 persentil), Grup 2; Fazla kilolu (%85-95 persentil), Grup 3; Obez (95. persentil ve üzeri). Derlenme ajitasyonu bir anestezist tarafından Watcha skalası ile değerlendirildi.

Bulgular: Çalışmaya 567 hasta dahil oldu ve bunların 115'inde (%20.3) derlenme ajitasyonu meydana geldi. Hastaların 428'i normal, 64'ü fazla kilolu ve 75'i obez olarak değerlendirildi. Demografik veriler karşılaştırıldığında gruplar arasında istatistiksel oalrak herhangi bir fark yoktu. Derlenme ajitasyonu insidansı Grup II (%28,1) ve Grup III'te (%29,3) Grup I'e (%17.5) göre istatistiksel olarak daha yüksekti (p=0,006).

Sonuç: Genel anestezi uygulanan çocuklarda hem fazla kilolu olmanın hem de obezitenin derlenme ajitasyonu insidansını arttırdığı görülmüştür ancak prospektif çalışmalara da ihtiyaç vardır.

Anahtar Kelimeler: Derlenme ajitasyonu, genel anestezi, pediatrik

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INTRODUCTION

Childhood obesity has become a global health problem today (1,2). According to World Health Organization (WHO) data, it is estimated that 378 million children worldwide are overweight or obese (3). While obesity directly affects health services, it is indirectly associated with bad social and economic outcomes (4). The incidence of comorbidities that increase the perioperative risk, such as hypertension, type 2 diabetes mellitus, asthma, obstructive sleep apnea, fatty liver and gastroesophageal reflux disease, is higher in obese children compared to non-obese children (5-9). Obesity not only causes physical problems, but also causes an increase in the incidence of depression and is associated with a decrease in quality of life (10,11). Both adult and pediatric obese patients are becoming an important part of the surgical population and their postoperative management is difficult compared to non-obese patients (12).

Emergence agitation (EA) was first reported in the early 1960s and genellikle çocuklarda genel anesteziden recovery sırasında görülür (13). Emergence agitation defined as "a disturbance in a child's awareness of and attention to his or her environment with disorientation and perceptual alterations, including hypersensitivity to stimuli and hyperactive motor behavior in the immediate post anesthesia period " (14). Emergence agitation presents challenges for healthcare professionals and an unpleasant experience for parents. The incidence of EA varies greatly in the literature [reaching as high as 80% (15)]. However, EA is thought to be very common and closely related to the use of sevoflurane and desflurane in pediatric anesthesia (16).

Although many studies have examined the factors affecting EA, studies evaluating the effect of obesity on postoperative EA in children are limited. In this study, we aimed to evaluate whether obesity has an effect on postoperative EA.

MATERIAL AND METHOD

This study was approved by the Institutional Ethics Committee and was conducted in accordance with the Declaration of Helsinki and EQUATOR guidelines. The requirement for written informed consent from patients and their parents was waived in this study because this is a retrospective observational study and the data were analyzed anonymously.

Medical records of the Selcuk University Medical Faculty Hospital between November 2018 and November 2022 were retrospectively reviewed. Pediatric patients (2-14 years old) with American Society of Anesthesiologists physical status (ASA PS) 1 or 2 who had undergone elective surgery under sevofluran general anesthesia were enrolled in this study.

The exclusion criteria were as follows: Under 2 years of age, over 14 years of age, American Society of Anesthesiologists Physical Status Classification System (ASA PS) of 3 or more, born prematurely, children who were intubated before induction of anesthesia or whose extubation was not planned after surgery, emergency cases, general anesthesia with an anesthetic agent other than sevoflurane.

Patients will be divided into 3 groups according to their body mass index; Group I; Normal (5th to 85% percentile), Group II; Overweight (85-95% percentile), Group III; Obese (95th percentile and above).

Anesthesia induction was performed in all patients as standard of institution: Face mask inhalation induction with sevoflurane 8% in oxygen and then intravenous (i.v.) catheter was placed. Intravenous rocuronium 0.6 mg/kg was given to facilitate tracheal intubation. Anesthesia was maintained using sevoflurane in an oxygen/air mixture and remifentanil (0.25 mc/kg/min). Paracetamol (10 mg/kg) were given i.v. to attenuate postoperative pain. Sevoflurane and remifentanil infusion was stopped near the end of the surgery. Sugammadex (2 mg/kg) was used for neuromuscular recovery in all patients. Following tracheal extubation, patients were cared for in the postanesthesia care unit (PACU).

Data obtained from medical records included data for age, sex, height, weight, body mass index (BMI), ASA PS, general anesthetic agents, anesthesia time, airway device, PACU stay time, incidence of occurrence agitation and pain. Emergence agitation was assessed by anesthesiologists using the Watcha scale as follows: 1, asleep or calm; 2, crying, but can be consoled; 3, crying, cannot be consoled; and 4, agitated and thrashing around (17). According to the Watcha scale, children who are 3 or 4 were accepted as emergence agitation. In the PACU, the criteria for discharge (which included consciousness, normal vital signs, no pain, and no nausea or vomiting) were the same for all patients. All patients discharged from the PACU according to the customary guidelines practiced in the institution. For measuring pain, FLACC scale (18) was used in PACU.

Statistical Analysis

Statistical analyses were performed with SPSS 21.0 software (SPSS Institute, Chicago, IL, USA). The Kolmogorov-Smirnov test was used to determine whether continuous variables followed a normal distribution. Parametric data were tested with Student's t test or the Mann–Whitney test and presented as means with standard deviation (SD) or medians with interquartile range (IQR). Categorical data were analyzed with the two-tailed Pearson's Chi-square test and are given as numbers and percentages. A P-value <0.05 was considered statistically significant.

RESULTS

In the medical records of Selcuk University Medical Faculty Hospital, it was determined that 1107 pediatric patients were operated between November 2018 and November 2022. The demographic and clinical characteristics of the patients are summarized in **Table 1**. Of these, only 567 met the inclusion criteria of the current study. Of all patients, 64% were male and 82.7% had ASA PS I. The median (IQR) age of the patients included in the study was 5.5 (3-9). The median (IQR) value of anesthesia time and PACU stay time was 60 (45-90) and 23 (18-25) min, respectively. Emergence agitation occurred in 115 children (20.3%).

Table 1. Demographic and clinical characteristics of the patients (n=567).

Categories	Values
Age, years	5.5 (3-9)
Gender, female/male	363 (64%)/204 (36%)
ASA PS, I/II	469 (82.7%)/98 (17.3%)
Anesthesia time, min	60 (45-90)
Airway device, ETT/SG	263 (46.4%)/304 (53.6%)
PACU stay time, min	23 (18-25)
FLACC score	2 (0-5)
Emergence agitation, n (%)	115 (20.3%)

ASA PS: American Society of Anesthesiologists Physical Status Classification System, ETT: endotracheal tube, SG: supraglottic, PACU: postanesthesia care unit. Data are presented median (IQR) or n (%).

As a result of the data obtained from the medical records, the patients were divided into 3 groups according to their body mass index: Group I; Normal (5th to 85% percentile) (n=428) , Group II; Overweight (85-95% percentile) (n=64) , Group III; Obese (95th percentile and above) (n=75). 11.2% of the patients included in the present study were overweight and 13.2% were obese.

Comparison of demographic and clinical characteristics of the groups is summarized in **Table 2**. There were no significant differences between the 3 groups regarding age, gender, ASA PS, anesthesia time, airway device used, PACU stay time and FLACC score (p=0.057, p=0.578, p=0.356, p=0.202, p= 0.756, p=0.107, and p=0.629, respectively). Emergence agitation incidence was statistically higher in Group II (28.1%) and Group III (29.3%) compared to Group I (17.5%) (p=0.006).

DISCUSSION

In this retrospective study, which included 567 patients aged 2-14 years, we found that obesity increased the incidence of agitation in pediatric patients undergoing general anesthesia.

Childhood obesity is associated with an increased risk of many comorbidities, particularly pulmonary, metabolic, orthopedic disorders and cardiovascular (19,20). Medical care of obese children can present challenges for all clinicians, including anesthetists. Although the care of adult patients in the perioperative period has been extensively studied, trials on this subject in obese children are limited. Various studies have reported that adverse events such as difficult mask ventilation, difficult airway, difficult laryngoscopy, laryngospasm, and oxygen desaturation are more common in obese children (21,22). There are still unclear areas about obesity in pediatric patients. Considering the adult literature, it is obvious that there is an inadequacy in the literature for pediatric patients.

In a study conducted with pediatric patients undergoing procedural sedation, it was reported that obesity caused an increase in the frequency of respiratory adverse events and a delay in recovery (23). In a study evaluating the effect of obesity degree on PACU discharge times in a pediatric patient group, it was reported that although PACU stay time was prolonged in both moderately and severely obese children who were operated on with general anesthesia compared to non-obese children, however, stated the degree of obesity had no effect. (24). In the present study, it was determined that obesity did not have any effect on PACU stay time.

Although the peroperative effects of obesity in pediatric patients have been evaluated in different studies, trials evaluating its effect on EA are limited. EA is a well-defined psychological and physical phenomenon that can be exhibited by children recovering from general anesthesia. EA is often described as: 1-Disorder of receptivity (abnormal reception of auditory or visual cues – child is inconsolable even by familiar voices or toys), 2-Disorder of perceptivity (heightened perception of stimuli and hyperactive motor behavior) (25). Although

Table 2. Comparison of demographic and clinical characteristics of the groups.

	Group In=428	Group IIn=64	Group IIIn=75	p
Age, years	6 (3-10)	4.5 (2-9)	6 (3-8)	0.057
Gender, female/male	277 (64.7%)/151(35.3%)	42 (65.6%)/22 (34.4%)	44 (58.7)/31 (41.3)	0.578
ASA PS, I/II	350 (81.8%)/78 (18.2%)	57 (89.1%)/7 (10.9%)	62 (82.7%)/13 (17.3%)	0.356
Anesthesia time, min	60 (45-90)	60 (45-85)	50 (35-78)	0.202
Airway device, ETT/SG	202 (47.2%)/226 (52.8%)	29 (45.3%)/35 (54.7%)	32 (42.7%)/43 (57.3)	0.756
PACU stay time, min	23 (18-25)	35 (28-38)	30 (25-35)	0.107
FLACC score	2 (0-5)	1.5 (0-5)	2 (0-4)	0.629
Emergence agitation, n(%)	75 (17.5%)	18 (28.1%)	22 (29.3%)	0.006

ASA PS: American Society of Anesthesiologists Physical Status Classification System, ETT: endotracheal tube, SG: supraglottic, PACU: postanesthesia care unit. Data are presented median (IQR) or n (%).

EA is usually short-lived, if it is prolonged, it causes a bad experience for both health professionals and parents in the postoperative period (26,27).

The effect of severe obesity on EA after pediatric ambulatory surgery was evaluated in a prospective, cross-sectional study by Reynolds et al (28). In this study severe obesity described as a risk factors for Emergence Agitation in pediatric ambulatory surgery. Unlike our study, this study included children between the ages of 4 and 17.In addition, the association of EA with PACU stay time was also evaluated in this study. Unlike our results, it was determined that PACU stay time was longer in patients with EA. Also, EA was documented in 66 (6.1%) patients. In our study, the incidence of EA was 20.3%.

While studies evaluating the effect of obesity on EA in pediatric patients are limited, this situation has been discussed more extensively in studies conducted in adult obese patients. Studies in adult patients have also shown that obesity is a risk factor for EA (29,30).

CONCLUSION

We are of the opinion that both overweight and obesity increase the incidence of EA in children undergoing general anesthesia, but prospective further studies are also required.

ETHICAL DECLARATIONS

Ethics Committee Approval: This study was conducted by ethics committee approval obtained from Selçuk University Faculty of Medicine (Approval number: 2022/532).

Informed Consent: Because the study was designed retrospectively, no written informed consent form was obtained from patients.

Referee Evaluation Process: Externally peer-reviewed.

Conflict of Interest Statement: The authors have no conflicts of interest to declare.

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Author Contributions: All of the authors declare that they have all participated in the design, execution, and analysis of the paper, and that they have approved the final version.

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ORIGINAL ARTICLE
ORİJİNAL ARAŞTIRMA

Our Approach to Labial Fusion During Childhood: Eight Years of Experience in a Single Center

Çocukluk Döneminde Labial Füzyona Yaklaşımımız: Tek Merkezde Sekiz Yıllık Deneyim

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ABSTRACT

Aim: In this study; we aimed to determine the clinical information and treatment results of labial fusion patients who applied directly to or consulted the Pediatric Surgery Outpatient Clinic of our hospital.

Material and Method: Female patients in the prepubertal age group with labial fusion who applied to the Pediatric Surgery Outpatient Clinic of our hospital were retrospectively reviewed in terms of age, complaints, recommended treatment, recurrence rates, time to recurrence, and complications.

Results: This study involved 438 patients with labial fusion aged between 32 days and 10 years in our hospital. Labial fusion was detected in 359 (82%) of the patients, a nearly complete opening only in the labia minora, and additional problems related to labial fusion in 180 (41%) patients. The patients were treated with topical estrogen and creams containing the raw material of the Centella Asiatica plant and manual fusion separation. Recurrence of labial fusion was seen in 18(4%) of these patients. It was found that the patient's age at the time of detection and additional problems related to labial fusion were effective on the recurrence of the disease.

Conclusion: Older age and additional problems may increase the recurrence rate of labial fusion. We think that we can get results faster, with less risk of recurrence, by detecting the pathology early, applying the necessary precautions, and using the creams containing the raw material of Centella Asiatica as well as the raw material of the Centella Asiatica plant in combination, if necessary.

Keywords: Girl, labial fusion, estrogen, Centella Asiatica

ÖZ

Amaç: Bu çalışmada; hastanemiz Çocuk Cerrahisi Polikliniği'ne doğrudan başvuran ya da konsülte edilen labial füzyon hastalarının klinik bilgileri ve tedavi sonuçlarını tespit etmeyi amaçladık.

Gereç ve Yöntem: Hastanemiz Çocuk Cerrahisi Polikliniği'ne başvuran labial füzyonlu puberte öncesi yaş grubunda olan kız hastalar, yaşları, şikayetleri, önerilen tedavi şekli, tekrarlama süre ve oranları, oluşan komplikasyonlar açısından geriye dönük olarak hastanemiz veritabanı kullanılarak gözden geçirildi.

Bulgular: Bu çalışmaya, hastanemizde yaşları 32 gün ile 10 yıl arasında olan labial füzyonlu 438 olgu dahil edildi. Hastaların 359(%82)' unda labial füzyon, sadece labia minörlerde tama yakın bir açıklık ve 180(%41)' inde labial füzyona bağlı ek sorunlar tespit edildi. Hastalar topikal östrojen ve Centella Asiatica bitkisinin hammaddesini içeren kremler ve manuel füzyon ayrılmasıyla tedavi edildi. Bu hastaların 18(4%)' inde labial füzyonda tekrarlama görüldü. Tespit esnasındaki hasta yaşının büyük ve labial füzyona bağlı ek sorunların hastalığın tekrarı üzerinde etkili olduğu bulundu.

Sonuç: Labial füzyonun tekrarlama oranını, yaşın büyük olması ve ek problemler arttırabilmektedir. Patolojinin erken saptanıp, gerekli önlemler uygulanması ve tedaviye topikal östrojenli yanısıra Centella Asiatica bitkisinin hammaddesini içeren kremlerin gerekirse kombine olarak kullanılmasıyla da daha hızlı, yüzgüldüren ve tekrarlama riskinin daha az olduğu sonuçlar alabileceğimizi düşünmekteyiz.

Anahtar Sözcükler: Kız çocuk, labial füzyon, östrojen, Centella Asiatica

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INTRODUCTION

Labial fusion is a benign pathology that happens moderately or entirely in adhesion between the labia minora in girls. It is mostly encountered between 0-2 years of age with a rate of 1.8%-3.3% in prepubertal girls (1). The etiology of labial fusion is still unknown. Although hypoestrogenism in prepubertal girls induces labial adhesion, complaints such as diaper rash, urinary tract infection (UTI), allergic dermatitis, diaper rash, low frequency of body washing, fungal infections, poor hygiene, vaginal stream, and diaper changes can be considered in the etiology (2). Labial adhesion can bring about problems such as UTIs, challenges in urination, and even hydronephrosis (2,3).

In this research article; we aimed to retrospectively assess the features and treatment methods of the patients with labial adhesions admitted to or consulted with the Pediatric Surgery Outpatient Clinic.

MATERIAL AND METHOD

After getting approval from the Ethics Committee with the date 26/07/2022 and the decision number 2022/7-16, patients who underwent intervention for labial fusion between July. 2014-July.2022 were involved in this research. The parameters of the patients were obtained from the Kardelen computer database of our hospital. The files of patients were scanned through the hospital data processing system. The patients' age at the time of diagnosis, complaints at presentation, use of estrogen therapy, frequency and duration of recurrence of adhesion, and complications were assessed.

After the families were adequately informed about the treatment methods, the use of topical cream with estrogen was suggested first in the patients who did not have any additional problems. Labial adhesion was opened manually under topical local anesthesia in patients who did not endorse the exercise of estrogen cream, and whose labial fusion continued after topical treatment.

A two-week warm sitting bath was recommended for these patients, and they were educated about the use of topical estrogen cream, lower care suggestions, and the importance of protection from irritation. Creams containing the raw material of the Centella Asiatica plant were prescribed for these patients as restorative and concealer. After two weeks we called these patients for clinical examination. At the control clinical examination who had thick, harsh adhesions which were found in the physical examination, that were not suitable for manual separation, the adhesion was separated surgically with bipolar electrocautery under sedation after getting the approval of the family. The patients were discharged on the same day as outpatients. During the follow-up period, topical estrogen, creams containing the raw material of Centella Asiatica, and warm baths were advised for 14 days postoperatively.

SPSS 15.0 statistical package program (SPSS Inc, Chicago Ill) was used for statistical assessments. Mann-Whitney U test was used in the analysis of continuous variables. Chi-square and Fisher's exact chi-square test were used in the comparison of categorical variables. A "p" value of <0.05 was evaluated statistically considerably in all analyses.

RESULTS

In our hospital, 438 patients with labial fusion who met the study criteria were reviewed. Nine patients were not included in the study due to missing information in hospital records. The ages of the patients included in the study ranged from 32 days to 10 years, with a mean age of 3.1 and a median age of 2.02 ± 2.42.

Although 346 (79%) of the patients whose records accessed from the hospital database were asymptomatic, the number of those applied directly to the Pediatric Surgery Clinic after the parents noticed the problem was 302 (69%).

The patients were classified into three groups according to their age: under 1 year (n=196, 44.7%), 1-5 years old (n=225, 51.4%) and 5 years old (n=17, 3.9%). It was investigated in which age group the risk of recurrence was higher.

Labial fusion repetition was found to be significantly lower in the under-1 age group (n=25, 12.8%) compared to other age groups (n=68, 30.2% and n=5, 29.4%, respectively) ($p=0.012$). It was found that labial fusion could recur for up to 5 years. Our follow-up period after intervention in labial fusion ranged from 15 days to 3 years (median 2.8 months).

Twenty-six (5.9%) of the patients had an attempt to open a fusion in another center before admission and applied to our polyclinic with the complaint of recurrence of the fusion. Anal fissure to labial fusion in 35 (8%) of the patients, perineal diaper dermatitis in 30 (6.9%) patients, constipation in 35 (8%), diarrhea in 3 (0.7%) ,and parasitosis was detected in 7 (1.6%) patients. Forty-eight (11%) of the patients had UTIs at the time of admission (**Table 1**). The most common microorganism grown in urine culture in UTI was Escherichia coli (n=41, 85.4%), followed by Klebsiella pneumonia (n=5, 10.4%) and Proteus (n= 2, 4.2%).

Table 1. Additional problems related to labial fusion

Additional problems related to labial fusion	Frequency n (%)
UTI	35 (8%)
Difficulty urinating	61 (14%)
Previously opened synechia	18 (4%)
Purulent discharge from the genital area	18 (4%)
Constipation	26 (6%)
Anal fissur	22(5%)
Total	180 (41%)

In 359 (82%) of the patients, the labial fusion completely covered the vagina and urethra, leaving only a small opening close to the clitoris. Since this situation caused a

like appearance of the male genitalia in girls, it caused families to apply to the hospital with the thought that their child did not have a uterus. Estrogen-containing topical cream was used for 14 days before the intervention in 35 (8%) of the patients who applied with labial fusion, and the full opening was not achieved in any of these patients in the control.

Labial fusions were opened in all the remaining patients, and in the post-treatment controls of those who received estrogen therapy (**Figure 1, 2**). After the fusion opened, the patients used topical cream with estrogen for 2 weeks. No complication development related to the procedure was encountered. While complete recovery was achieved in 342 (78.1%) of the patients after this treatment, the remaining patients underwent repeated intervention due to recurrence of fusion 1-6 times after the recovery period.

The age of the patients with an additional problem related to labial fusion is 83 days-8.5 years (median 1.8 years), the age of the patients with isolated labial fusion is between 40 days-8.5 years (median 1 year) was changing. It was found that patients with an additional problem related to labial fusion were older ($p<0.001$). It was observed that the fusion recurred in 97 (53.9%) of the patients with an additional problem related to labial fusion, and in 23 (8.9%) of those who did not have any additional problems. Problems accompanying labial fusion were found to have a statistically significant effect on the risk of recurrence of the fusion ($p<0.001$).

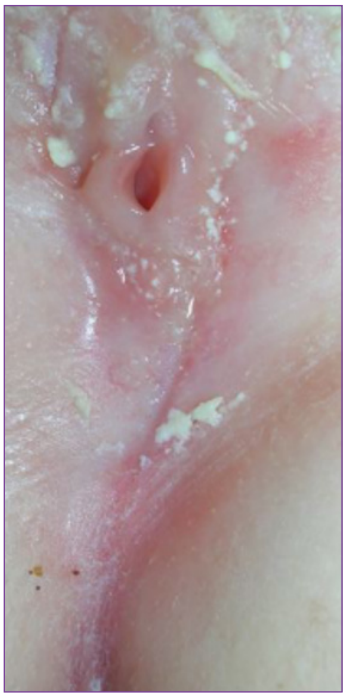

Figure 1. Labial fusion before manuel separation

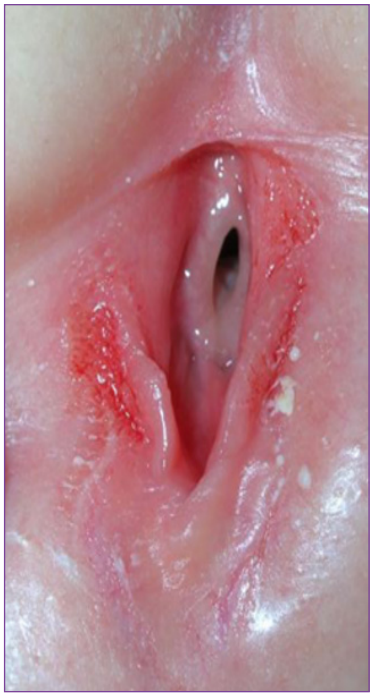

Figure 2. Labial fusion after manuel separation

DISCUSSION

The prevalence of labial adhesion is 1.8%, with the highest incidence (3.3%) in 13 to 23 months (4). Local trauma and damage, especially in the genital area, cause fibrous exudate that causes tissue deterioration. Fibrous exudate is thought to predispose to labial fusion. Labial fusion can occur when the labia minora stick together forming an inflammatory tissue membrane. The extent of labial fusion varies from nearly complete fusion to milder cases where 30-50 percent of the labia minor length is adhered. It usually causes obvious clinical complaints in girls between the ages of three months and three years. The reason why it is not seen very often before 3 months is the effect of estrogen in the mother, whose effect can still continue in the baby (5).

Labial adhesions are usually seen in infants and young girls before puberty. When girls increase their estrogen levels with puberty, labial adhesions are much less common and labia minora can sometimes open spontaneously (6).

Most girls with labial adhesions do not have any obvious symptoms. In some girls, pain, difficulty in urination, clinical signs of urinary tract infection, and labial adhesions can be seen. Even a girl with labial adhesions complains of dripping or leaking urine. This is due to the fact that the urine is trapped behind the labial fusion region and then occurs (7). Most of the patients are asymptomatic and may be noticed by the parents while they are being cared for or during the physical examination by the doctor.

Parents can bring their children to pediatric or pediatric surgery clinics with dermatitis, dysuria, UTI, and obstruction. Although 734 (82.5%) of the patients whose records accessed from the hospital database were asymptomatic, the number of those applied directly to the Pediatric Surgery Clinic after their parents noticed the problem was 643 (72.3%) (8) In this study, 346 (79%) of the patients included were asymptomatic. The number of those who applied directly to the Pediatric Surgery Clinic after the parents noticed the problem was 302 (69%). These findings were also found to be consistent with the literature.

Preliminary treatment with topical estrogens is considered safe, even in long-term treatment applications. According to previous studies on this subject, successful results have been achieved in 50-80% of treated patients.(9-10). In our study, 340 (78.6%) of the cases entirely recovered after the first course of treatment.

The success rate with local estrogen cream treatment varies between 47-100%. There are also those who use 0.05% betamethasone cream as an alternative to estrogen in topical treatment or in combination. In a study, the success rate in labial fusion after betamethasone cream application was 68%, and the recurrence rate was 23%.

In a study by Eroğlu et al., estrogen cream, betamethasone cream, and were administered to 131 patients in three groups for four weeks. In this study, only topically with estrogen 15.5%; The success rate was 15.6% with betamethasone alone, 28.5% with combined treatment, and no statistically significant difference could be found between the results(12). In a study by Soyer, patients were divided into three groups, only topical estrogen was applied to one group for two weeks, manual separation was applied to one group, and manual separation and prophylactic estrogen therapy were applied together to another group. In the estrogen-only group, the success rate was 66.6% in the third month; 55.5% in the ninth month; The success rate in the third and ninth months was 85.7% in the group that only underwent manual separation. The success rate was found to be 100% in the group where manual separation and topical estrogen were applied together. In a study by Muram, topical estrogen therapy was successful in approximately half of 289 patients treated for labial adhesions, and the rest were opened by manual separation. manual separation and prophylactic estrogen therapy were applied together in one group (13).

The mode of action of the components containing the raw material Centella Asiatica plant in the treatment of skin diseases is essentially anti-inflammation, anti-oxidation and reduction of damage to mitochondria by oxidative stress, which is also compatible with the pathogenesis of these diseases. Studies at the cell level suggested that Centella Asiatica standard extract (ECa 233) might affect filopodia formation and increase wound healing by activating FAK, Akt and MAPK signaling pathways (14). In particular, creams containing Centella Asiatica plant raw materials were thought to be beneficial to use together with estrogen due to their repairing and concealing effect in irritation after the manual opening of the labial fusion.

CONCLUSION

As the age increases in patients with labial fusion, additional problems related to this pathology are seen more frequently. The coexistence of these two conditions also increases the fusion recurrence rate. Labial fusion is a pathology that is frequently seen in prepubertal girls and can cause serious concern in families. While the labial fusion regresses with the prevention of irritations due to diaper use and hygiene recommendations in the early period, raw materials containing Centella Asiatica plant and estrogen-containing creams may be used in advanced cases and manual fusion may be required when necessary. With the early detection of labial fusion and the implementation of necessary precautions, treatment can be better. We think that it is easy, and the risk of recurrence is less.

ETHICAL DECLARATIONS

Ethics Committee Approval: This study was conducted by ethics committee approval obtained from Karamanoğlu Mehmetbey Unıversity School of Medicine (Date: 26.07.2022, Meeting Number: 07, Decision Number: 16)

Informed Consent: All patients signed the free and informed consent form.

Referee Evaluation Process: Externally peer-reviewed.

Conflict of Interest Statement: The authors have no conflicts of interest to declare.

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Author Contributions: All of the authors declare that they have all participated in the design, execution, and analysis of the paper, and that they have approved the final version.

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ORIGINAL ARTICLE
ORİJİNAL ARAŞTIRMA

Evaluation of the Opinions of the Pediatric Palliative Care Patients' Families Regarding the COVID-19 Pandemic: A Cross-Sectional Study

Pediatrik Palyatif Bakım Hasta Ailelerinin COVID-19 Pandemisine İlişkin Görüşlerinin Değerlendirilmesi: Kesitsel Bir Çalışma

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Abstract

Aim: While many countries around the world have faced similar challenges in pediatric palliative care as COVID-19, there have also been challenges that vary from country to country. In this study, it was aimed to evaluate the difficulties and opinions of families receiving pediatric palliative care in our country during the pandemic process.

Materials and Method: The study is a cross-sectional survey applied to families followed in the pediatric palliative care service between 01.12.2018 and 01.12.2020. In the study, caring parents were asked to share their experiences and opinions in the last year.

Results: Of the 175 families followed, 112 were included in the study. In the first year of the pandemic, it was determined that 42% of the families had no change in their lives, 16.1% had psychological problems, 35.7% had social (quarantine and restrictions) problems and 6.3% had economic problems. When the family order and endurance of those who stated that there were significant changes in their lives in the first year of the pandemic were examined, it was found that these individuals were bored with their families and had difficulty staying at home (2.482; p=0.013).

Conclusion: The COVID-19 pandemic process in pediatric palliative care has been milder and has a better prognosis than expected. Psychosocial difficulties are the most prominent areas of distress.

Keywords: COVID-19, pandemic, pediatric palliative care

Öz

Amaç: Dünya çapında birçok ülke pediatrik palyatif bakımda COVID-19 ile benzer zorluklar yaşarken, ülkeden ülkeye değişen zorluklar da olmuştur. Bu çalışmada pandemi sürecinde ülkemizdeki pediatrik palyatif bakım alan ailelerin yaşadıkları güçlüklerin ve görüşlerinin değerlendirilmesi amaçlanmıştır.

Gereç ve Yöntem: Araştırma, 01.12.2018 ile 01.12.2020 tarihleri arasında pediyatrik palyatif bakım servisinde takip edilen ailelere uygulanan kesitsel bir anket çalışmasıdır. Araştırmada bakım veren ebeveynlerden son bir yıldaki deneyimlerini ve görüşlerini paylaşmaları istenmiştir.

Bulgular: Takip edilen 175 aileden 112'si çalışmaya dahil edildi. Pandeminin ilk yılında ailelerin %42'sinin hayatında bir değişiklik olmadığı, %16,1'inin psikolojik, %35,7'sinin sosyal (karantina ve kısıtlamalar) ve %6,3'ünün ekonomik sorunları olduğu belirlendi. Pandeminin ilk yılında hayatlarında önemli değişiklikler olduğunu belirtenlerin aile düzeni ve tahammülleri incelendiğinde bu bireylerin ailelerinden sıkıldıkları ve evde kalmakta zorlandıkları saptandı (2.482; p= 0.013).

Sonuç: Pediatrik palyatif bakımda COVID-19 pandemi süreci beklenenden daha hafif ve daha iyi prognozlu seyretmiştir. Psikososyal zorluklar, en belirgin sıkıntı alanlarıdır.

Anahtar kelimeler: COVID-19, pandemi, pediatrik palyatif bakım

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INTRODUCTION

Coronavirus disease-2019 (COVID-19) is a disease that emerged in the city of Hubei Province-Wuhan in China in December 2019 and affected the whole world (1-3). The World Health Organization (WHO) has announced that the number of confirmed cases to date is 121,969,223 and deaths as 2,694,094 (4). For Turkey, T.R. The Ministry of Health reported a total number of 2,992,694 cases and 29,959 deaths so far (5). The COVID-19 pandemic mostly affects those with advanced age and chronic diseases (6). Despite this, these terrifying figures have caused various restrictions, significant changes in the way of life, especially social isolation, worldwide. Thus, behavioral and social changes occured (7). The comorbidities of children with life-limiting or threatening diseases such as primary disease and/or chronic lung disease constitute a risk factor for COVID-19 (8,9). Although the difficulties experienced in the process related to COVID-19 in pediatric palliative care (PPC) in many countries of the world show similarities, the differences in the health system and practices have led to varying difficulties from country to country (10). This study aimed to evaluate the difficulties patients and their families who received pediatric palliative care during the pandemic process and the effect of the pandemic.

MATERIAL AND METHOD

Organization of PPC Unit

Izmir Dr. Behcet Uz Pediatric Diseases and Surgery Training and Research Hospital is a tertiary hospital, and the PPC center started to serve in November 2018. It is the only center in the Aegean Region. Our PPC center has 12 beds and is an example of teamwork consisting of three doctors, eight nurses, four staff, one psychologist, one dietician, one social worker, one physiotherapist, one religious worker, one secretary. To our PPC center, children whose treatment is possible but unsuccessful (cancer, complex cyanotic congenital heart disease), with potentially progressive conditions (cystic fibrosis, severe immunodeficiency), without therapeutic options (trisomy 13, trisomy 18, osteogenesis imperfecta), children with non-progressive but irreversible disease (cerebral palsy) are admitted.

Study Design

The study was a cross-sectional questionnaire study planned to be applied to the parents of all patients followed up the PPC center between December 01, 2018 and December 01, 2020. The families were reached by phone, and after they were informed, they were asked to answer the questionnaire by asking the prepared questions. Parents whose children died, parents who could not speak Turkish, parents whose telephone records were not available, and parents who refused to participate in the study excluded. Only one of the parents (mother or father) was included in the study. Sociodemographic data, whether they had COVID-19 infection, social media use, family life, what the pandemic changed in families' lives, the experiences of the sick child and parents, their future hopes and expectations from the palliative care team were investigated. A total of 39 questions were asked, and 21 subgroup options were opened one by one for multiple-choice questions (**Figure 1**).

Ethics

The study was carried out with the permission of Izmir Dr Behçet Uz Children's Hospital Clinical Researches Ethics Committee (Date: 25.03.2021, Meeting Number: 540, Decision Number: 2021/06-05). This study was performed as per the Declaration of Helsinki. Verbal consent was obtained from the parents who agreed to participate in the study by making the necessary explanations during the phone call to the parents.

Statistical Analysis

SPSS (Statistical Package for Social Sciences) package program version 15.0 was used for statistical analysis to evaluate the data obtained in the study. In addition to descriptive statistical methods (e.g., mean, standard deviation, frequency, percentage.), Chi-square test or Fisher's exact test for categorical variables, Student T-test or Mann Whitney U test for continuous variables were used to cpmpare two groups. Results were evaluated a 95% confidence interval and $p < 0.05$ significance level.

RESULTS

A total of 175 caregiver parents whose children were followed up by the PPC center were enrolled in the study. However, only 112 of these caregiver parents were included in the study. Twenty-six parents were excluded from the study because of the death of their children, 24 parents because they did not have a telephone record or could not be reached, five parents could not speak the language, six parents were nursing home caregivers, and two parents did not want to participate in the study. Only one of the parents (mother or father) was included in the study.

The mean age of the parents in the study group was 35.6 ± 6.8 year (24-54), and 79 (70.5%) were female. The rate of caregiver parents who had COVID-19 infection was 7.1% (n=8), and the rate of COVID-19 in children who received the care was 1.7% (n=2). The rate of those who had COVID-19 in their first degree relatives and/or friends were 33.9% (n=38), while those who lost their first degree relatives and/or friends due to COVID-19 were 7.1% (n=8).

General information					
Name:					
Gender:					
Age:					
Have you had COVID-19?					
Do you have a relative with COVID?					
Have you lost a loved one due to COVID-19?					
Do you have any health problems?					
Has your use of social media and internet increased?					
Has your palliative care relationship with social media increased?					
Have the apps you use changed?					
Has your mode of transport changed?					
What has been the most important thing that has changed in your life?					
Part 1. Family routine and endurance					
1. We feel calm and peaceful as a family	1	2	3	4	5
2. Sometimes we get bored as a family or have trouble staying at home	1	2	3	4	5
3. We try to find positive aspects, we are hopeful	1	2	3	4	5
4. We enjoy family time we didn't have before	1	2	3	4	5
5. We are already used to change and uncertainty	1	2	3	4	5
6. Tired of not having the help I received before (eg personal assistant, babysitter, grandma)	1	2	3	4	5
7. I miss contact with my friends and relatives	1	2	3	4	5
8. I have many practical and economic concerns	1	2	3	4	5
Part 2. Changes in child and caregiver life					
1. My family feels calm and peaceful	1	2	3	4	5
2. Sometimes my family gets bored or has trouble staying at home	1	2	3	4	5
3. My child's routine remains the same despite limitations	1	2	3	4	5
4. As a family, we set our new routine to find mental balance	1	2	3	4	5
5. I dedicate myself to unresolved problems and jobs that I can't find time for	1	2	3	4	5
6. I rediscover outdoor environments such as a garden or terrace	1	2	3	4	5
7. I am resting, having a comfortable time	1	2	3	4	5
8. I don't have free time; I always have something to do	1	2	3	4	5
9. I let my son break some rules	1	2	3	4	5
Part 3. Top concern for caregiver					
Health of family and friends					
Possible COVID-19 infection in your child					
Possible COVID-19 infection of the caregiver or other cohabiting family members long term future					
Part 4. Wishes and plans for the future after COVID-19					
Specific outdoor activity					
"Going to the beach"					
"Going to the Mountains"					
"Go shopping"					
"Relaxing on the grass"					
"Going on vacation"					
"Going out to dinner"					
"Go to cinema"					
Social relations					
"Seeing grandma"					
"Seeing the rest of the family"					
"Getting together with friends and relatives"					
"Rediscovering the taste of sociality"					
Health problems					
"Going swimming to do physiotherapy"					
"Restarting Physiotherapy"					
"Going to hospital checkups"					
Sports					
"Riding a horse"					
"To swim"					
"To do sport"					
Others					
"Embrace Everyone"					
"Moving to a new home"					
"Going to the hairdresser"					
"Wearing heels"					
Part 5. Perception of support from the PPB team					
Is there anything palliative care can do for you during this period? Yes No					
Comments: _____					

Figure 1. Survey Form

In the first year of the pandemic, it was determined that 58% of the caregivers, who were asked whether there was a significant change in their lives, had a significant change in their lives, and 42% had no change in their lives. Among parents who have had significant changes in their lives due to the pandemic, it was determined that 16% had psychological issues, 35.7% had social issues (quarantine and restrictions), and 6.3% had economic problems. 35.7% (n=40) did not use public transportation, and 17.9% (n=20) accessed palliative care through applications such as WhatsApp. It was determined that the use of social media by 50.9% (n=57) of caregiving parents increased, and the social media applications used by 33% (n=37) changed. The relationship between parents, who stated that there were significant changes in their life in the first year of the pandemic, and the situation of having COVID-19 infection was examined. It was found that 78.9% of those whose first-degree relatives and/or friends had COVID-19 infection had significant changes in their lives (9.068; p=0.003). When the family routine and endurance of those who stated that there were significant changes in their lives in the first year of the pandemic were examined, these individuals were bored with their families and had difficulty staying at home (2.482; p=0.013). When the life of the child in need of care and the caregiver parent was examined during the pandemic, no difference was found between those who stated that there was a significant change in their lives and those who stated that there was no change in their lives (**Table 1**). Examining what caregivers were most concerned about in the first year of the pandemic, it was determined that they were concerned about the long-term future and the health of their first-degree relatives and friends. Sources of concern are presented in **Table 2**.

Table 1. Comparison of the scores of the study group on items related to "Changes in the life of the child and caregiver"

	(Mean-SD) Median (Q1-Q3)	Significant changes in life		p-value
		No (Mean-SD) Median (Q1-Q3)	Yes (Mean-SD) Median (Q1-Q3)	
My family feels calm and peaceful	3.5 (1.3) 3.0 (3.0-5.0)	3.7 (1.3) 4.0 (3.0-5.0)	3.3 (1.3) 3.0 (2.5-5.0)	1.618; 0.106
Sometimes my family is bored or has trouble staying home	3.0 (1.4) 3.0 (1.0-4.0)	3.0 (1.4) 3.0 (2.0-4.0)	2.9 (1.5) 3.0 (1.0-4.0)	0.182; 0.856
My child's routine remained the same despite the limitations	3.5 (1.4) 4.0 (3.0-5.0)	3.5 (1.4) 4.0 (2.0-5.0)	3.5 (1.3) 4.0 (3.0-5.0)	0.237; 0.812
As a family, we set our new routine to find mental balance	2.8 (1.3) 3.0 (2.0-4.0)	2.6 (1.3) 2.0 (1.0-3.0)	3.0 (1.3) 3.0 (2.0-4.0)	1.724; 0.085
I devoted myself to unsolved problems and work where I could not find time	3.2 (1.4) 3.0 (2.0-4.0)	3.1 (1.3) 3.0 (2.0-4.0)	3.2 (1.5) 3.0 (2.0-5.0)	0.485; 0.628
I rediscover outdoor environments such as gardens or terraces	2.7 (1.5) 3.0 (1.0-4.0)	2.7 (1.3) 3.0 (1.0-4.0)	2.7 (1.7) 3.0 (1.0-4.0)	0.006; 0.995
I am resting, having a comfortable time	3.1 (1.5) 3.0 (2.0-5.0)	3.1 (1.4) 3.0 (2.0-4.0)	3.0 (1.6) 3.0 (1.0-5.0)	0.416; 0.677
I do not have free time; I always have a job to do	3.6 (1.4) 4.0 (3.0-5.0)	3.8 (1.3) 4.0 (3.0-5.0)	3.5 (1.5) 4.0 (2.0-5.0)	1.100; 0.271
I let my boy break some rules	2.4 (1.3) 2.0 (1.0-3.0)	2.5 (1.3) 2.0 (1.0-3.0)	2.4 (1.3) 2.0 (1.0-3.0)	0.420; 0.974

Table 2 . Situations that are "a source of concern" due to COVID-19

Situations of concern due to COVID-19		Significant changes in life		p-value
		No	Yes	
Worrying about the health of family and friends	Yes	1 (6.3)	15 (93.8)	8.140; 0.004
	No	46 (47.9)	50 (52.1)	
Worrying about possible COVID-19 infection in their child	Yes	31 (42.5)	42 (57.5)	0.000; 1.000
	No	16 (41.0)	23 (59.0)	
Worrying about the caregiver and caregiver child's COVID-19 infection	Yes	20 (45.5)	24 (54.5)	0.165; 0.685
	No	27 (39.7)	41 (60.3)	
Worry about the long-term future	Yes	5 (20.8)	19 (79.2)	4.550; 0.033
	No	42 (47.7)	46 (52.3)	
Total		47 (42.0)	65 (58.0)	112 (100.0)

What they wanted to do most when the pandemic process was over included going to the hospital (79.5%), meeting with friends and relatives (69.6%),and relaxing on the grass (65.2%). Considering the expectations of the groups with and without changes in their lives from the future, going on vacation (14.491; p = 0.000), seeing family elders (10.277; p = 0.001), seeing the rest of the family (6.913; p = 0.009), getting together with friends and distant relatives (6.735; p = 0.009), swimming (4.188; p = 0.041), and moving to a new house (4.675; p = 0.031) were determined as the most important requests and expectations (**Table 3**). It was determined that 20.5% (n=23) of the participants needed the support of the palliative care team, 47.8% (n=11) were related to psychological support, and 26% were related to drugs and materials.

Table 3. Comparison of "Planning for the future" in the study group

Wishes and plans for the future after COVID-19		n (%)	Significant changes in life		p-value
			No	Yes	
Going to the beach	Yes	22 (19.6)	6 (27.3) (12.8)	16 (72.7) (24.6)	1.734; 0.188
	No	90 (80.4)	41 (45.6) (87.2)	49 (54.4) (75.4)	
Going to the mountains	Yes	55 (49.1)	18 (32.7) (38.3)	37 (67.3) (56.9)	3.078; 0.079
	No	57 (50.9)	29 (50.9) (61.7)	28 (49.1) (43.1)	
Going shopping	Yes	60 (53.6)	27 (45.0) (57.4)	33 (55.0) (50.8)	0.257; 0.612
	No	52 (46.4)	20 (38.5) (42.6)	32 (61.5) (49.2)	
Relaxing in the grass	Yes	73 (65.2)	29 (39.7) (61.7)	44 (60.3) (67.7)	0.208; 0.649
	No	39 (34.8)	18 (46.2) (38.3)	21 (53.8) (32.3)	
Going on vacation	Yes	42 (37.5)	8 (19.0) (17.0)	34 (81.0) (52.3)	14.491; 0.000
	No	70 (62.5)	39 (55.7) (83.0)	31 (44.3) (47.7)	
Going out to dinner	Yes	31 (27.7)	10 (32.3) (21.3)	21 (67.7) (32.3)	1.153; 0.283
	No	81 (72.3)	37 (45.7) (78.7)	44 (54.3) (67.7)	
Going to cinema	Yes	29 (25.9)	8 (27.6) (17.0)	21 (72.4) (32.3)	2.573; 0.109
	No	83 (74.1)	39 (47.0) (83.0)	44 (53.0) (67.7)	
Seeing grandmother	Yes	28 (25.0)	4 (14.3) (8.5)	24 (85.7) (36.9)	10.277; 0.001
	No	84 (75.0)	43 (51.2) (91.5)	41 (48.8) (63.1)	
Seeing the rest of the family	Yes	65 (58.0)	20 (30.8) (42.6)	45 (69.2) (69.2)	6.913; 0.009
	No	47 (42.0)	27 (57.4) (57.4)	20 (42.6) (30.8)	
Getting together with friends and relatives	Yes	78 (69.6)	26 (33.3) (55.3)	52 (66.7) (80.0)	6.735; 0.009
	No	34 (30.4)	21 (61.8) (44.7)	13 (39.4) (20.0)	
Rediscovering the taste of sociability	Yes	53 (47.3)	18 (34.0) (38.3)	35 (66.0) (53.8)	2.058; 0.151
	No	59 (52.7)	29 (49.2) (61.7)	30 (50.8) (46.2)	
Going swimming to do physiotherapy	Yes	16 (14.3)	5 (31.3) (10.6)	11 (68.8) (53.8)	0.441; 0.506
	No	96 (85.7)	42 (43.2) (89.4)	54 (56.8) (46.2)	
Restarting physiotherapy	Yes	28 (25.0)	9 (32.1) (19.1)	19 (67.9) (29.2)	0.990; 0.320
	No	84 (75.0)	38 (45.2) (80.9)	46 (54.8) (70.8)	
Going to hospital checks	Yes	89 (79.5)	39 (43.8) (83.0)	50 (56.2) (76.9)	0.298; 0.585
	No	23 (20.5)	8 (34.8) (17.0)	15 (65.2) (23.1)	
Riding a horse	Yes	20 (17.9)	5 (25.0) (10.6)	15 (75.0) (23.1)	2.092; 0.148
	No	92 (82.1)	42 (45.7) (89.4)	50 (54.3) (76.9)	
Swimming	Yes	37 (33.0)	10 (27.0) (21.3)	27 (73.0) (41.5)	4.188; 0.041
	No	75 (67.0)	37 (49.3) (78.7)	38 (50.7) (58.5)	
Doing sports	Yes	59 (52.7)	21 (35.6) (44.7)	38 (64.4) (58.5)	1.562; 0.211
	No	53 (47.3)	26 (49.1) (55.3)	27 (50.9) (41.5)	
Embracing everyone	Yes	52 (46.4)	17 (32.7) (36.2)	35 (67.3) (53.8)	2.753; 0.097
	No	60 (53.6)	30 (50.0) (63.8)	30 (50.0) (46.2)	
Moving to a new home	Yes	27 (24.1)	6 (22.2) (12.8)	21 (77.8) (32.3)	4.675; 0.031
	No	85 (75.9)	41 (48.2) (87.2)	44 (51.8) (67.7)	
Going to the hairdresser/barbershop	Yes	28 (25.0)	10 (35.7) (21.3)	18 (64.3) (27.7)	0.306; 0.580
	No	84 (75.0)	37 (44.0) (78.7)	47 (56.0) (72.3)	
Wearing heels¶	Yes	13 (16.5)	3 (23.1) (9.4)	10 (76.9) (21.3)	1.191; 0.275
	No	66 (83.5)	29 (43.9) (90.6)	37 (56.1) (78.7)	

¶: Calculated over the number of women.

DISCUSSION

The results of this study revealed that parents who made significant changes in their lives had first-degree relatives and/or friends who were diagnosed with COVID-19 infection. It was determined that these families were bored with the changes and had difficulty staying at home, but this situation was not compelling for the caring parent and child.

According to the joint WHO and China report, COVID-19 in adults is 3-10%, mostly through domestic transmission, especially in large families (11). In Turkey, according to the T.C. Ministry of Health data, the rate of those who had COVID-19 infection was reported to be 9.8% (12). COVID-19 has been found with a rate of 2.1-2.7% in children (13,14). For COVID-19 infection, where epidemiological studies still continue, it is observed that the presence of older age, male gender, immunodeficiency, heart disease, and respiratory diseases constitute a high risk and cluster in some families (11,12). Mortality rates vary according to age and region, but it has been reported as 21.9% globally and 2.57% in our country (11,12). In our study group, the rate of those who had COVID-19 infection was lower, and no mortality was observed. This situation may have caused these families, who already experienced social difficulties and isolation due to their children's illness, to come out of the COVID-19 pandemic process with less harm than expected.

Having a child with a life-limiting/threatening disease negatively affects the lives, feelings, thoughts, and behaviors of family members. Parents need to change their duties and responsibilities, financial resources, vital activities, and behaviors (15). In addition to the child's disease, many family problems such as psychological, social, financial, and education accompany the process. Studies show that these families are more physically and emotionally vulnerable and lead a lower quality of life (16). Evidence-based data show that some families choose to be at home and together in this challenging process, while some families choose a more isolated life and spend time in the hospital (17). Our study reveals that children with life-limiting/threatening diseases and their caregivers are already accustomed to the challenges. Besides, those with a first-degree relative/friend who had COVID-19 during the pandemic are more careful, have grasped the seriousness of the situation, and have made significant changes in their lives as a whole family. This may be due to being a caring parent, having an ill child, close contact with COVID-19, and exposure concerns.

Care for children with life-limiting/ threatening diseases and their families has become more difficult with infection control measures and restrictions (18). In our country, the area where families' quality of life is affected the most in pediatric palliative care is the social field, and the importance of providing social support on quality of life has been demonstrated (19). Social distance and visitor restrictions applied to ensure infection control during the pandemic process are for the protection of families and healthcare workers. However, stricter rules and restrictions for these children who already have social difficulties and their families increased the time they stayed at home, prevented them from receiving care support, and caused an increase in social difficulties (20,21). Our study revealed that the family as a whole had difficulty staying at home due to new rules, restrictions and quarantine practices. When only the life of the child-caregiving parent couple was questioned, it was observed that staying at home and living with restrictions did not bring additional difficulties. This situation made us think that the restrictions cause distress and difficulties in other familiy members, and the caregiver and the caregiving parent are already used to this lifestyle. The low COVID-19 infection rates for these children and families with chronic respiratory disease may be due to this lifestyle they are used to.

Health threats against oneself and their loved ones cause psychosocial stress in individuals (19). During the pandemic process, psychosocial changes occurred due to interruptions in routines, separation from family members and friends, lack of daily needs, salary cut, social isolation, and school closure, and the reactions to these new normals are variable (15,19). In infectious diseases, the feeling of malaise, overestimation of the possibility of infection, excessive and inappropriate adoption of precautionary measures may be seen, as well as rejecting the risks of infection, not performing recommended health behaviors such as hand hygiene and social distance (15,16,19). The lifetime prevalence of depression was reported as 10.8% and anxiety as 4.66% (22-25). During the COVID-19 pandemic, depression was reported in 19% of adults and anxiety in 14% (26). It has been shown that social isolation and loneliness are associated with bad moods (19). During the COVID-19 process, individuals had to be more at home and live in isolation. In the literature, people who have not had an epidemic before, those who are worried about not being able to find enough surgical masks, those who are not able to work from home, and those who are worried about not being able to find enough materials are stated as people who are prone to anxiety and depression and who are recommended more psychological support (26). It has been reported that those who have experienced epidemic diseases before are more resistant, experienced in obeying the precautions, and less stressed (25). Contrary to what was expected in our study, the most important concern of families has been identified as the health of family and friends and the long-term future. The reason why there is little concern about the health of the child-caregiving parent may be that they are already living mostly at home and partly in isolation.

This situation may be related to the fact that there is no significant change in the routine life of the caregiver and the child receiving care.

Individuals increase their adaptation capacity by functionally re-meaning a traumatic event and change their perspective on events. Although some aspects of post-traumatic strengthening emerge immediately, most effects extend over a long period of time and develop over time. The speed of this development varies according to demographic factors, the effect of the traumatic event and the stress it creates, the resources available, the strength of social solidarity, the use of functional coping skills, and personality traits (27). Despite having a child with pediatric palliative care patients and the added COVID-19 pandemic, parents and families have hopes, dreams, and expectations. The most apparent expectation in our study was visiting grandmother, seeing the rest of the family, getting together with relatives and friends were found to be in the social area. Going on vacation, swimming, and moving to a new home is an indication of the need for change. In our study, psychological support was found to be the most expectation from palliative care, and the difficulties experienced less were the medication and supplies. In pediatric palliative care, it can be said that the COVID-19 pandemic most prominently causes psychosocial distress.

Study Limitations

Since the study was an inquiry for the past year, the variability of the situations that families feel anxious and hoped for due to difficulty in remembering and rapid changes in the process are the limitations of the study. Again, since the phone records did not belong to a single parent (mother or father), the differences in viewpoints in the inquiries made are a limitation of the study in terms of standardization.

CONCLUSION

The COVID-19 pandemic process in pediatric palliative care is a milder and better prognosis than expected. Families whose first-degree relatives suffer from the disease make significant changes in their lives. Psychosocial difficulties are the most obvious areas of distress. Due to the nature of the pandemic, there may be different challenges in each country.

ETHICAL DECLARATIONS

Ethics Committee Approval: The study was carried out with the permission of Izmir Dr Behçet Uz Children's Hospital Clinical Researches Ethics Committee (Date: 25.03.2021, Meeting Number: 540, Decision Number: 2021/06-05).

Informed Consent: All patients signed the free and informed consent form.

Referee Evaluation Process: Externally peer-reviewed.

Conflict of Interest Statement: The authors have no conflicts of interest to declare.

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Author Contributions: All of the authors declare that they have all participated in the design, execution, and analysis of the paper, and that they have approved the final version.

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CASE REPORT
OLGU SUNUMU

Çocuklarda Nöbet ile Prezente Olan Nadir Görülen Nöraksisin Kalsifiye Psödoneoplazmı (CAPNON)

Rare Calcified Pseudoneoplasm of Neuraxis Presenting with Seizure in Children (CAPNON)

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Öz

Nöraksisin kalsifiye psödoneoplazmaları (CAPNON), santral sinir sisteminin nonneoplastik ve nadir görülen lezyonlarıdır. Etiyolojisi belirsizdir ve cerrahi olarak çıkarılması genellikle küratiftir. Radyografik özellikleri, bilgisayarlı beyin tomografide belirgin kalsifikasyonlar ve manyetik rezonans (MR) görüntülemede değişken kontrastlanma gösteren, T1 ve T2 ağırlıklı görüntülerde hipointensite görülmesiyle tanımlanmıştır. Erişkinlerde de sık rastlanmayan CAPNON, çocuklarda ise çok nadir görülmektedir. Bildirilen CAPNON tümörlerinin büyük çoğunluğu intrakraniyal olmakla birlikte, spinal kord da bulunabilir. Asemptomatik olabildiği gibi semptom gösterdiğinde de baş ağrıları, nöbet ve fokal nörolojik defisitler ile presente olabilir. Literatürde çok nadir bildirilen, değişken bilincin eşlik ettiği fokal nöbet epizodları ile başvuran ve CAPNON tanısı alan opere olmadan takip edilen 12 yaşında çocuk olguyu sunuyoruz.

Anahtar Kelimeler: Nöraksisin kalsifiye psödoneoplazmaları (CAPNON), çocuk, beyin tümör,

Abstract

Calcified pseudoneoplasms of neuraxis (CAPNON) are nonneoplastic and rare lesions of the central nervous system. Its etiology is unclear and its surgical removal is usually curative. Its radiographic features were defined by prominent calcifications seen on computed tomography (CT) and hypointensity on T1- and T2-weighted images with variable enhancement on magnetic resonance (MR) imaging. CANON, which is not common in adults, is very rare in children. The majority of reported CAPNON tumors are intracranial, although the spinal cord may also be found. It may be asymptomatic or present with headaches, seizures and focal neurological deficits when symptomatic. We present a case of a 12-year-old boy who presented with focal seizure episodes accompanied by variable consciousness and was diagnosed as CAPNON, which was rarely reported in the literature, and was followed up without surgery.

Keywords: Calcified pseudoneoplasms of neuraxis (CAPNON), child, brain tumor, seizure

GİRİŞ

Nöraksisin kalsifiye psödoneoplazmaları (CAPNON), merkezi sinir sistemi (MSS) içinde herhangi bir yerde ortaya çıkabilen nadir ve tipik olarak iyi huylu lezyonlardır. Bu iyi huylu ve yavaş büyüyen tümör, MSS'nin intraaksiyal, ekstraaksiyal herhangi bir yerinde bulunabilir ve tipik olarak metastaz yapmaz. CAPNON tümörlerinin büyük çoğunluğu intrakraniyal olmakla birlikte, spinal kordda da bulunabilir. Tümörün etyolojisi ve risk faktörleri bilinmemektedir. CAPNON tümörü olan hastaların klinik sunumu heterojendir. En sık görülen semptomları baş ağrıları, nöbet ve fokal nörolojik defisitlerdir. Literatürde nöbet ile başvuran olguların biri jeneralize tonik-klonik, diğeri tanımlanamayan bilinç değişikliği semptomlarıyla başvurmuştu (1). Literatürde CAPNON'un postmortem muayenede veya asemptomatik hastalarda bulunduğu da bildirilmiştir (2). Semptomlar çoğunlukla lezyonun yeri ve boyutuna bağlıdır (3). Standart tedavi, tam cerrahi rezeksiyon içerir ve küratif olduğu bilinmektedir (4).

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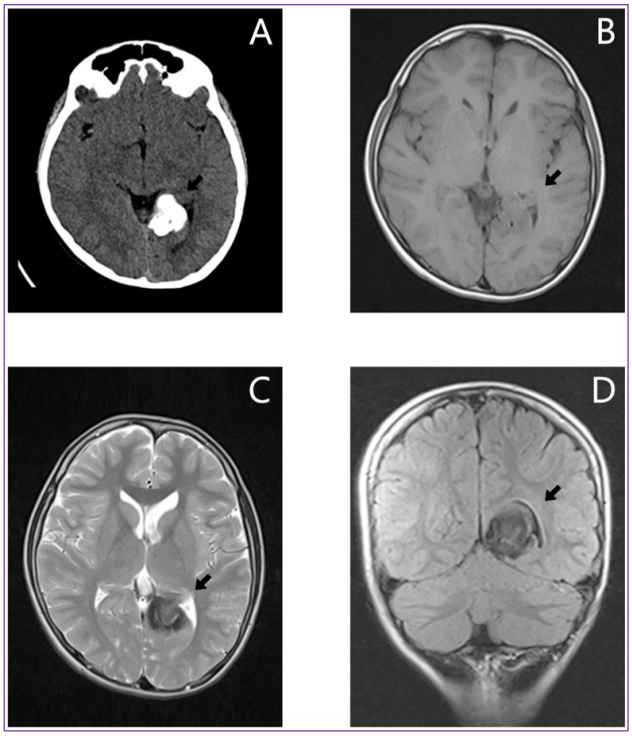
OLGU

On iki yaşında erkek olgu 6 aydır aralıklarla devam eden baş dönmesi, baş ağrısı ve 2 haftada bir olan, yaklaşık 30 sn süren nöbet şikâyeti ile başvurdu. Nöbetlerin genellikle sabah saatlerinde başlayıp, heyecanlandığında tetikleniyormuş. Ailesi nöbet esnasında bilincin kaybolduğunu, ileri geri sallanma ve ardışık hareketlerin de olduğunu ifade etti. Anne baba arasında akrabalık olmayan, öz ve soy geçmişinde özellik bulunmayan olgunun, yapılan sistemik ve nörolojik muayene bulguları ve psikomotor gelişimi ve okul başarısı normal bulundu. Yapılan rutin biyokimyasal testlerinin (hemogram, sedimentasyon akut faz reaktanları, karaciğer ve böbrek fonksiyon testleri) hepsi normaldi.

Olgunun beyin manyetik rezonans görüntülemesinde (MRG), sol oksipitotemporal bölgede T2 ağırlıklı (T2A) görüntülemelerde hipointens, T1 ağırlıklı (T1A) görüntülemelerde zayıf hipointensite gösteren lezyon tespit edildi. Bilgisayarlı beyin tomografisinde (BT); Sol oksipitotemporal bölgede lateral ventrikül oksipital hornu komşuluğu medialinde yaklaşık 3x2,5 cm boyutunda düzensiz konturlu yoğun amorf kalsifikasyon görüldü (**Şekil 1**). Radyolojik bulguları tipik olarak CAPNON ile uyumlu idi.



Şekil 1. Nöraksisin kalsifiye psödoneoplazmı olan olgumuzun tipik nöroradyolojik bulguları. A, Kontrastsız bilgisayarlı beyin tomografi aksiyal görüntüleme sol oksipitotemporal bölgede düzensiz konturlu yoğun amorf kalsifikasyon (ok işareti). B, Beyin manyetik rezonans (MR) T1 ağırlıklı (T1A) aksiyal görüntülemede hafif hipointens alan (ok işareti). C, Beyin manyetik rezonans (MR) T2A aksiyal görüntülemede hipointens alan (ok işareti). D, Beyin manyetik rezonans (MR) T2A fluid attenuated inversion recovery coronal görüntülemede lezyon etrafında ödem olmayan hipointens alan (ok işareti).

Olgunun uyku, uyanıklık ve hiperventilasyonla uyarılmış elektroensefalografisinde (EEG) epileptik aktivite görülmedi. Ancak klinik olarak geçici bilinç kaybının, otomatizmaların eşlik ettiği fokal nöbet ile uyumlu olması ve tümörün derin serebral yerleşimli olması nedeniyle EEG'de epileptik aktivite bulgusu veremeyebileceği düşünülerek valproik asit (VPA) tedavisi (20 mg/kg/gün) başlandı. CAPNON tümörü sebebiyle beyin ve sinir cerrahisi tarafından değerlendirilmesinde; lezyonun derin serebral yerleşimli olması ve olası cerrahi risklerin yüksek olması nedeni ile operasyonsuz takip önerildi. Olgunun VPA tedavisi sonrası fayda gördü, nöbet sayısı azalmıştı. Kliniği stabil devam eden ve klinik nöbet aktivitesi VPA tedavisi ile fayda gören olgu beyin ve sinir cerrahisi ve çocuk nörolojisi tarafından düzenli aralıklarla takibe alındı.

TARTIŞMA

Nöraksisin kalsifiye psödoneoplazmaları literatürde nadir görülen lezyonlardır. Pubmed veritabanı tarandığında literatürde şimdiye kadar CAPNON tümörü olan 5 çocuk vaka bildirilmiştir (1,5-8). CAPNON tümörleri infratentorial ve supratentorial yerleşimli olabilir. Yetişkinlerde tümörün, supratentorial alanda daha çok frontal lobta, infratentorial alanda ise daha çok serebellumda görüldüğü bildirilmiştir (3). Bildirilen çocuk vakalarda ekstradural spinal lezyonu olan 22 aylık ve 12 yaşında iki çocuk, intraaksiyal temporal lob lezyonu olan 6 yaşındaki ve 16 yaşında iki çocuk, frontal bölgede lezyonu olan bir çocuk rapor edilmiştir (1,5-8). Bizim olgumuzda 12 yaşında sol oksipitotemporal bölgede lezyon saptanmıştır. Spinal kord tutulumlarında lokal boyun veya sırt ağrısı ya da yayılan ağrı görülebilir. Buna karşılık, intrakraniyal CAPNON'da en yaygın semptom nöbetler olup baş ağrısı, baş dönmesi, fokal nörolojik semptomlar izlenebilir (3). Bildirilen çocuk vakalarda semptomlar sırt ağrısı, nöbet ve bilinç değişikliği idi. Asemptomatik olan bir vaka mevcuttu. Vakalarda görülen nöbet tipi generalize tonik-klonik şeklindeydi. Nöbet ile prezente olan olgunun lezyonu temporal bölge yerleşimli idi. Bizim vakamızda ilk başvuru şikâyeti baş dönmesi ve bilinç değişikliğinin eşlik ettiği fokal nöbet olup muayenesinde fokal nörolojik defisit saptanmadı. CAPNON tümörler Beyin BT'de hiperdens lezyonlar olarak görülürken, MRG'de, hem T1A hem de T2A görüntülerde belirgin şekilde hipointens sinyal yoğunluğu gösterir (7). Bizim olgumuzun kontrastsız beyin BT'de sol oksipitotemporal alanda yoğun kalsifikasyonları ve beyin MRG T2A görüntülerde hipointens T1A görüntülerde zayıf hipointens idi. Semptomatik CAPNON'lar için tercih edilen tedavi cerrahi rezeksiyondur. Literatürde sunulan tüm pediatrik hastaların postoperatif kliniğinin iyi olduğu bildirilmiştir (8). Bizim olgumuzda lezyonun oksipitotemporal bölgede derin yerleşimli olması nedeniyle cerrahi için riskli bulunmuştur. Pediatrik vaka-

larda küratif cerrahi tedavinin yanı sıra, cerrahi yüksek riskli olgularda, tümörün çok yavaş büyüyen özellikte olduğunuda göz önüne alırsak klinik ve görüntüleme ile takip edilebileceğini vurgulamak isteriz. Olgumuz literatürde bildirilen son pediatrik CAPNON vakası olup cerrahi olmadan yaklaşık 2 yıldır stabil takip edilen ilk vakadır.

SONUÇ

CAPNON vakaları literatürde erişkinde görülmekle birlikte nadir olarak pediatrik yaş grubunda karşımıza çıkmaktadır. Olguların neredeyse hepsi opere edilmesi nedeniyle biz semptom vermeyen, stabil seyreden olası cerrahi riskleri yüksek olan CAPNON vakalarını operasyonsuz takip edilebileceğini vurgulamak istedik.

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Hakem Değerlendirme Süreci: Harici çift kör hakem değerlendirmesi.

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Yazar Katkıları: Yazarların tümü; makalenin tasarımına, yürütülmesine, analizine katıldığını ve son sürümünü onayladıklarını beyan etmişlerdir.

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CASE REPORT
OLGU SUNUMU

Congenital Insensitivity to Pain with Anhidrosis Syndrome: A case report in Diyala province / Iraq

Konjenital Ağrı Duyarsızlığı ile Anhidrosis Sendromu: Diyala vilayeti / Irak'tan Bir Olgu Sunumu

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Abstract

Congenital insensitivity to pain with anhidrosis syndrome (CIPA); is a rare autosomal recessive disorder presenting with pain insensitivity, sweating inability, and intellectual disability. The incapability to sense pain and temperature often leads to recurrent severe and inadvertent self-inflicted harm; these can result in severe complications, as patients settle slowly from skin and bone harm. We present a case of a four-year-old boy with a diagnosis of CIPA, after repeated visits to the hospital emergency department for repeated chest and both ankle joint infections, which prompted further investigations. A four-year-old boy was admitted to Albatool teaching hospital for maternity and children in Baqubah, Diyala, Iraq because of recurrent chest and both ankle joints infection. He is the second child of consanguineous parents. His six-year-old sister is normal. The mother noticed early after birth that her child was suffering from high fever, he was not responding to pricking and injections, and he never sweats with intolerance to warm weather. Examination revealed mental developmental delay, absent upper and lower canine teeth, napkin and face dermatitis which was intractable to therapy, and deep pus discharging ulcers of both heels. Radiology of feet shows signs of osteomyelitis. There is a history of the same disease in two male cousins who died at age of three and five years respectively, the overall clinical context warranted a clinical suspicion of CIPA. Early diagnosis of this extremely rare disease is very important for the treatment and prevention of complications. This case report shows that a clinician should suspect to investigate for CIPA when managing kids with multiple inadvertent self-inflicted harms, anhidrosis, and pain insensitivity.

Keywords: CIPA, self-mutilation, osteomyelitis

Öz

Konjenital Ağrı Duyarsızlığı ile Anhidrosis Sendromu (CIPA); ağrıya duyarsızlık, terleme yetersizliği ve zeka geriliği ile seyreden, nadir görülen otozomal resesif geçişli bir hastalıktır. Acıyı ve sıcaklığı hissedememek, sıklıkla tekrarlayan şiddetli ve istemeden kendi kendine zarar vermeye yol açar; Hastalar deri ve kemik hasarından yavaş yavaş yerleştiğinden bunlar ciddi komplikasyonlara neden olabilir. Bu yazıda, tekrarlayan göğüs ve her iki ayak bileği eklemi enfeksiyonu nedeniyle hastanenin acil servisine tekrarlayan ziyaretleri sonrasında CIPA tanısı konan dört yaşında bir erkek çocuğu vakayı sunuyoruz. Irak'ın Diyala eyaletine bağlı Bakuba kentindeki Albatool Doğum ve Çocuk Eğitim Hastanesi'ne 4 yaşında bir erkek çocuk, tekrarlayan göğüs ve her iki ayak bileği eklemi enfeksiyonu nedeniyle yatırıldı. Akraba anne babanın ikinci çocuğudur. Altı yaşındaki kız kardeşi normal. Anne, doğumdan hemen sonra çocuğunun yüksek ateşi olduğunu, iğnelere ve iğnelere tepki vermediğini ve asla sıcak havaya karşı tahammülsüzlükten terlemediğini fark etti. Muayenede zihinsel gelişim geriliği, üst ve alt köpek dişlerinin yokluğu, tedaviye dirençli peçete ve yüz dermatiti ve her iki topuğun derin irinli ülserleri saptandı. Ayak radyolojisi osteomiyelit belirtileri gösteriyor. Sırasıyla üç ve beş yaşında ölen iki erkek kuzende aynı hastalık öyküsü vardır, genel klinik durum klinik CIPA şüphesini garanti etmiştir. Son derece nadir görülen bu hastalığın erken teşhisi, komplikasyonların tedavisi ve önlenmesi için çok önemlidir. Sonuç: Bu vaka raporu, bir klinisyenin, birden fazla istemeden kendi kendine zarar veren, anhidroz ve ağrı duyarsızlığı olan çocukları yönetirken CIPA araştırmasından şüphelenmesi gerektiğini göstermektedir.

Anahtar Kelimeler: CIPA, kendini yaralama, osteomiyelit

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INTRODUCTION

CIPA also known as hereditary sensory and autonomic neuropathy Type IV (HSAN-IV) is a very rare autosomal recessive disorder presenting with pain insensitivity, sweating inability, and intellectual disability. The cause of CIPA development is mutations in the neurotrophic tyrosine kinase receptor type 1 gene (NTRK1)).(1-7) This gene encodes the high-affinity receptor of nerve growth factor which is found on the 1q21-q22 chromosome. The incidence of the disease is 1 in 125 million newborns (2) so it is a very rare disease; there are only a few hundred cases reported in the world, 60 of them in the united states. The NTRK1 pathway is important for the preservation of autonomic sympathetic postganglionic neurons as it is in charge of skin innervation through sensory axons (8-13). Dearborn is the first one who described the pathology in 1932 as Congenital pure analgesia. Swanson 1963 described the First reference, in literature, to CIPA, and Fruchtman et al 2013 described clinical presentation including morbidity of the condition.CIPA is tremendously risky, and mostly the patient doesn't live over the age of 25. (5,8,10,14-18,19). The presentation of the disorder is with episodes of overheating due to warm ambient temperatures because these patients have absent or decreased sweating. Hypotonia improves with growth.(1,3,19-24) Pain perception is considerably reduced and autonomic nervous system functions are lost but the pressure and touch sensations are preserved which also causes poor healing of wounds and fractures with a tendency for chronic osteomyelitis and Charcot joints development. (17,24-26). Anhidrosis causes thick skin with the appearance of callosities, lichenification of the palms, and chronic dystrophic changes in the nails. There tearing is present, but corneal ulceration occurs from hypoesthesia. Almost all patients have behavioral and cognitive deficits (7-9,26). Only one case of CIPA was reported in Ninawa / Iraq (27).

Diagnostic Tests

The diagnosis of CIPA is based on the clinical presentation, pharmacological test (intradermic reaction to 1:10, 000 histamine) and neuropathological exam in electron microscopy and detection of mutations on the NTRK1 gene represents the last diagnostic step (12). A comprehensive workup is needed to reach the diagnosis. Though the controversial role of nerve biopsy, it has been stated that CIPA is accompanied by the loss of unmyelinated and reduction of small, myelinated fibers in the sural nerve, which could describe these manifestations. (13).Genetic counseling should discourage consanguineous marital relations, especially if there is a positive history of CIPA syndrome in the family (13).

Treatment

There is no definite treatment for the disease but surgical restoration is the only intervention for joint deformity in CIPA (13). Conservative treatment looked to be the best choice to preclude further complications caused by multiple surgeries. (Preventive measures such as local foot care and custom-fitted shoes can help minimize the risk of injury and avoid the need for radical surgeries. (14). Early diagnosis of CIPA could make parents more aware of risk factors, so, accidents could be avoided with continual alertness to the child's activities. Dentistry clinics play a big role in the prevention of injuries due to self-mutilation by the child(15), through the extraction of primary teeth which was recommended in the 1960s for the avoidance of self-mutilation. wearing of dental guards along with strict vigilance by the parents are better options available which t are more appropriate to the quality of life of the CIPA child. The aim of reporting this syndrome is to make physicians familiar with this condition and avoid unnecessary surgeries and even amputations, use conservative treatments, and make the diagnosis of this syndrome easier without extra laboratory requests.

CASE

A four-year-old boy was admitted to Albatool teaching hospital for maternity and children in Baquba, Diyala, Iraq because of recurrent chest infection and osteomyelitis of both ankle joints, He is the second child of consanguinous parents. His 6-year-old sister is normal. His mother noticed early after birth that her child was suffering from high fever, he was not responding to pricking and injections, absence of pain during intravenous line placement and he never sweat with intolerance to warm weather. Teething had started at seven months of age but because of recurrent finger biting and trauma to gum, his canine teeth had fallen at 2.5 years of age. At 10 months of age, he was admitted to the hospital after a febrile convulsion and stayed there for two weeks with a persistent fever. A thorough investigations were done including (CBC, CRP, ESR, urine exam and culture, blood culture, CSF examination and culture, echocardiography, bone marrow aspiration and culture, abdominal ultrasound, and brain CT scan) which were normal. Then three months later , he was readmitted because of a respiratory infection, and episodes of hyperthermia and unexplained fever recurred. At 2 years ofd age he had pneumonia and cellulitis of the right foot. In infancy, He was hypotonic, self-mutilation, and had neurodevelopmental delay which was misdiagnosed as a metabolic disease. At age of three years, the patient was diagnosed with right foot osteomyelitis. The child had a tongue laceration which was difficult to heal with delayed wounds healing in other parts of the body. The result of uric acid, serum glucose, liver, renal, and thyroid function tests, serum lactate, ammonia, creatinine phosphokinase level, and chromatography of amino acids all were normal. Nerve conduction velocity (NCV) was normal and brain MRI showed mild brain atrophy, but intradermal reaction to 1:10,000 histamine was positive. Although aggressive treatment started, his

foot condition shows no improvement so amputation was advised by an orthopedic surgeon. The parents refused this option, later the patient had a left foot infection which necessitates admission to the hospital again. A re-evaluation of the child is done. Examination revealed mental developmental delay with mild mental retardation, absent upper and lower canine teeth, napkin and face dermatitis which was intractable to therapy (**Figure 1**), deep pus discharging ulcers of both heel (**Figure 2**), swab send for culture which revealed staph aureus infection and resistant to a lot of drugs except meropenem), a deep scar in the right ear and skin changes of both legs (**Figure 3**), neurological examination was normal. The patient felt touch and pressure sensations but not pricking and heat. There was no hepatosplenomegaly or lymphadenopathy. ophthalmologic examination was normal. Immunological investigations were normal. Radiology of feet shows signs of osteomyelitis (Figure 4). There is a history of the same disease in two male cousins who died at age of three and five years respectively, one of them underwent below knee amputation. The diagnosis of CIPA was done and it must be confirmed by electron microscopic study of nerve biopsy and genetic tests which is not available in our center.

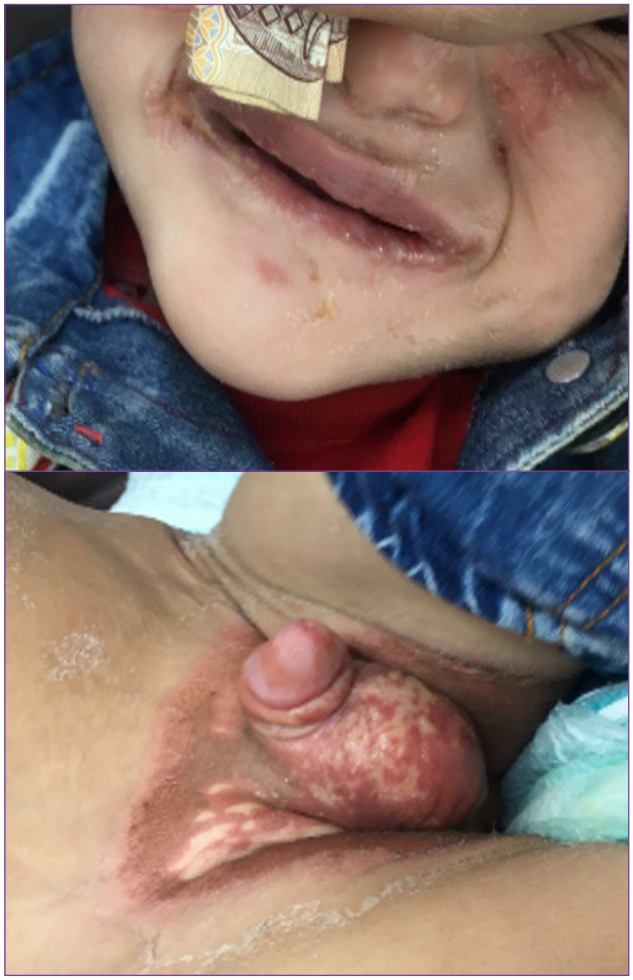

Figure 1. Dermatitis of the face and the napkin area not responding to treatment

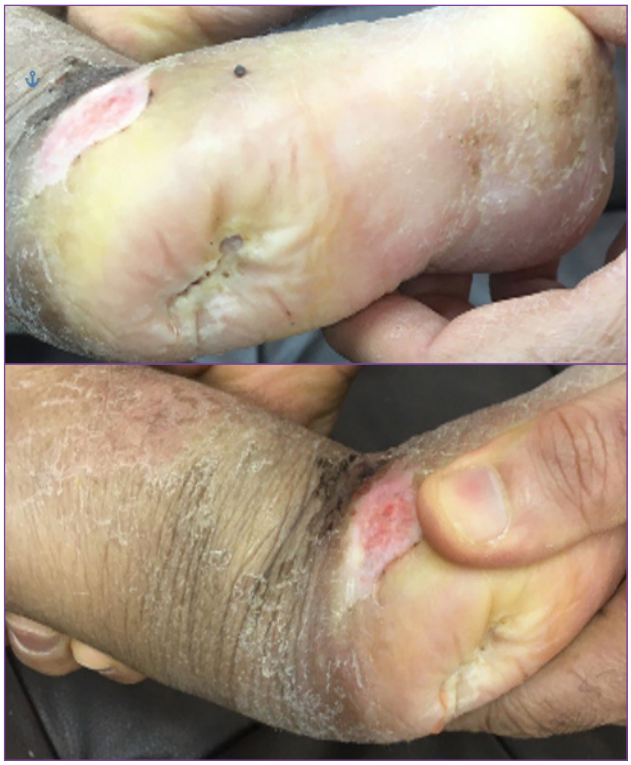

Figure 2. Deep pus discharging ulcers of both heels (osteomyelitis).

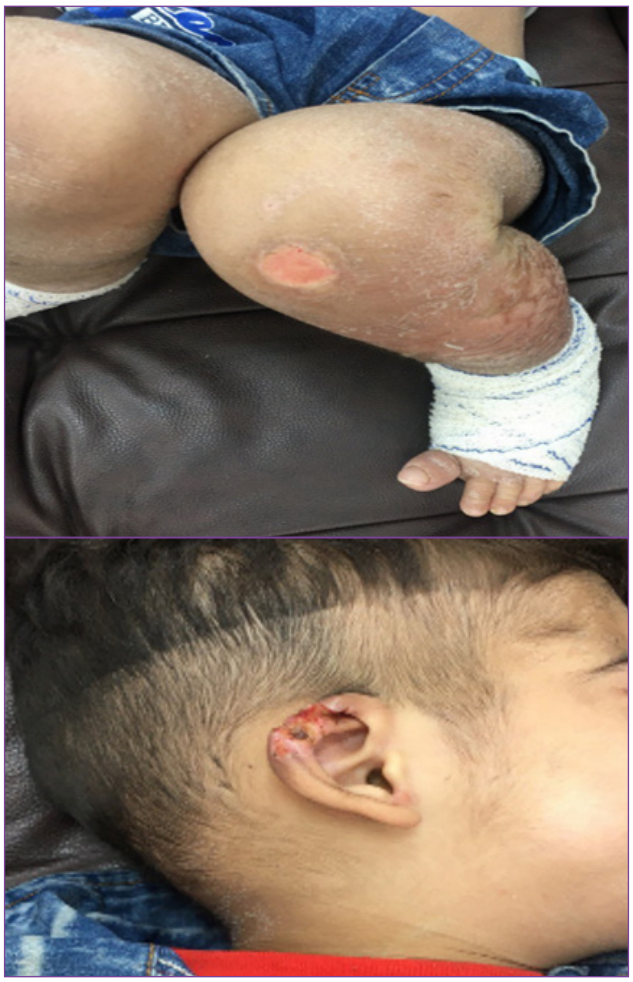

Figure 3. Skin and ear infection

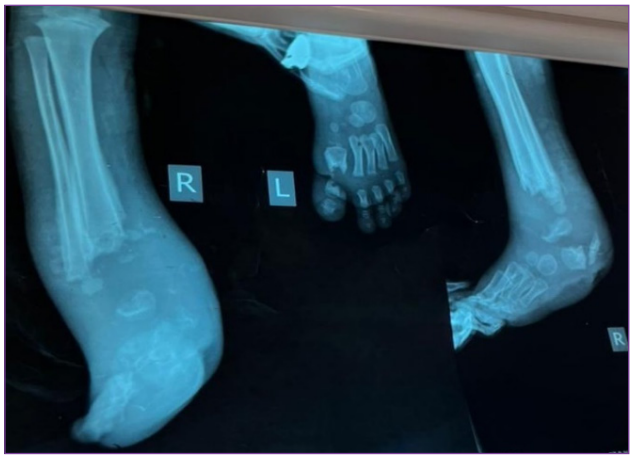

Figure 4. X-Ray of right and left ankle joints: shows nearly destructed joints by inhomogeneous bone sclerosis with the formation of sequestra (osteomyelitis).

DISCUSSION

Congenital insensitivity to pain with anhidrosis is an autosomal recessive disease caused by a mutation in the gene neurotrophic receptor tyrosinase 1 (NTRK1), which is located on chromosome 1q21-22. (16) It has been identified that there are at least 37 mutations among different ethnic groups(25). The major presenting feature of the patient was fever due to anhidrosis which is also common in other reports (22-24). Absence of pain sensation seen in all the patients leading to painless ulcers of mouth structures and extremities with unawareness of injuries produced by trauma or by self-mutilation so fingers and toes infection, scarring of lips and tongue are commonly detected (17,22-25). Self-harm behavior and mild mental retardation present in our patient which had been treated as developmental delay as reported in other literatures (22-24) .There is skin changes like skin dryness which presents as keratoderma in palmo-plantaris in the advanced state as reported by Daneshjou et al (9) The child had eczema, which was difficult to treat which is reported in other literitures (20,22). Chronic bone infections (osteomyelitis) as our patient suffered ,due to delayed healing as seen in other reports (19,22-26). The first sign of this syndrome is fever secondary to anhidrosis, which has recurrent presentation since neonatal period or from the first months of life.(18,22,23,24-27).The febrile seizures occur frequently as seen by our patient. sural nerve biopsy shows myelination defect and loss of small myelinated fibers and it was not performed on our patient due to unavailability of the test in our hospital and family compliance regarding performing the test outside the institution. Unfortunately in our country, we do not have the facility to confirm the diagnosis by genetic tests so we have to start treatment by clinical findings to make the patient's quality of life better. A heat stroke is very dangerous especially in our country so early diagnosis is important to prevent this complication .

CONCLUSION

Early diagnosis of this extremely rare disease is very important for the treatment and prevention of complications. Depending on clinical presentation and high suspicion is important in avoiding unnecessary investigations,early and proper treatment will prevent unnecessary amputation.

ETHICAL DECLARATIONS

Informed Consent: Written informed consent was obtained from all participants who participated in this study.

Referee Evaluation Process: Externally peer-reviewed.

Conflict of Interest Statement: The authors have no conflicts of interest to declare.

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Author Contributions: All of the authors declare that have all participated in the design, execution, and analysis of the paper, and that they have approved the final version.

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CASE REPORT
OLGU SUNUMU

Göz Bulgularının Eşlik Ettiği Trizomi 13 Vakası Sunumu

Trisomy 13 Presenting Mostly with Ocular Findings: Case Report

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Öz

Trizomi 13; ilk defa 1960'ta tanımlanmış olup, mikrosefali, mikroftalmi, yarık damak dudak, mental retardasyon, polidaktili, konjenital kalp anomalileri, üriner sistem anomalileri ve santral sinir sistemi gelişim anomalileri sıklıkla eşlik etmektedir. Klasik trizomi, 47, XX/XY + 13, şeklinde olup daha nadir olarak translokasyon ve mozaisizm gibi kromozomal düzensizlikler de görülmektedir. Vaka: Tokat Gaziosmanpaşa Üniversitesi Hastanesi' nde takip edilen 30 yasındaki annenin G2P2Y2 olarak 32 haftalıkken sezaryen ile 1405 gr olarak dünyaya gelen kız bebeğinin APGAR skoru 1. dakikada 5 ve 5. dakikada 6 idi. Fizik muayenesinde; kilosu 1405 gram (3 persentil altı), boyu 41 cm (3 persentil altı), baş çevresi 26 cm (3 persentil altı) idi. Dismorfik görünümü ve ön fontonel boyutu 3x4 cm olan hastanın mikrosefaliyle birlikte, skalpte sol paryetal alanda aplazia kutis konjenita, sağ gözde mikroftalmi, sol gözde anoftalmi, yarık damak ve dudak mevcuttu. Üç ekstremitede polidaktili olduğu görüldü. Abdomen ultrasonografi (USG), kranial USG ve beyin bilgisayarlı tomografide bir patoloji görülmedi. Ekokardiyografisi normal olarak değerlendirildi. Sağ gözde konjenital katarakt ve mikroftalmi mevcutken; sol göz anoftalmikti. Tartışma Trizomi 13'ün yaklaşık insidansı 10.000 canlı doğumda birdir. Vakamızda; FISH analizi trizomi 13 ile uyumlu bulunan hastadan moleküler karyotiplendirme ise yine 47, XX, + 13 olarak bulunmuştur. Trizomi 13 sendromunun en sık görülen bulguları motor ve mental gerilik, mikrosefali, mikroftalmi, holoprozensefali, hipotelorizm, yarık damak ve/veya yarık dudak, kardiovasküler, genitoüriner ve oküler malformasyonlardır. Mikroftalmi, yarık damak ve yarık dudak, polidaktili olması bu sendrom için karakteristiktir. Vakamızda yarık damak-dudak, polidaktili, sol göz anoftalmi, sağ göz mikroftalmi ve konjenital katarakt, mikrosefali, mikrognati eşlik etmektedir. Bununla birlikte Trizomi-13 vakalarında görülme sıklığı yüksek olan kardiyak anomali, santral sinir sistemi gelişim anomalisi, üriner sistem anomalisi görülmemiştir.

Anahtar Kelimeler: Trizomi 13, aplasia kutis, prematüre

Abstract

Trisomy 13 was first described in 1960, and microcephaly, microphthalmia, cleft palate lip, mental retardation, polydactyly, congenital heart anomalies, urinary system anomalies and central nervous system development anomalies are frequently accompanied. Mostly classical trisomy (47 is XX/XY + 13) is present, but translocation and mosaicism are also seen. Case: A female infant was born at the 32 weeks of gestatiton via cesarean section to a 30-year-old mother who was followed up in Tokat Gaziosmanpaşa University Hospital. The APGAR score of the, was 5 at the 1st minute and 6 at the 5th minute. She was small for her gestational age; 1405 gr in weight, 41 cm in height and 26 cm in her head circumference. The patient had a dysmorphic appearance with anterior fontonel size of 3x4 cm. She had microcephaly, aplasia cutis congenita in the left parietal area of the scalp, congenital cataract and microphthalmia in the right eye, anophthalmia in left eye, cleft palate and lip. Polydactyly was observed in three extremities. Abdominal and cranial ultrasonography, brain computed tomography did not reveal any pathology. Echocardiography was evaluated as normal. Discussion: The approximate incidence of trisomy 13 is one in 10,000 live births. In our case; The molecular karyotyping of the patient whose FISH analysis was found to be compatible with trisomy 13 was again found to be 47, XX, + 13. The most common findings of trisomy 13 syndrome are motor and mental retardation, microcephaly, microphthalmia, holoprosencephaly, hypotelorism, cleft palate and/or cleft lip, cardiovascular, genitourinary and ocular malformations. Microphthalmia, cleft palate and cleft lip, polydactyly are characteristic for this syndrome. In our case, cleft palate-lip, polydactyly, anophthalmia in left eye, microphthalmia and congenital cataract in right eye, microcephaly, micrognathia were accompanied. However, cardiac anomaly, central nervous system developmental anomaly, and urinary system anomaly, which have a high incidence in Trisomy-13 cases, were not observed.

Keywords: Trisomy 13, aplasia kutis, premature

GİRİŞ

Trizomi 13; ilk defa 1960'ta tanımlanmış olup, mikrosefali, mikroftalmi, yarık damak dudak, mental retardasyon, polidaktili, konjenital kalp anomalileri, üriner sistem anomalileri ve santral sinir sistemi gelişim anomalileri sıklıkla eşlik etmektedir (1). Trizomi 13'ün klasik triadı ise: yarık damak/dudak, mikroftalmi, polidaktili olarak tanımlanmıştır (2,3).

Klasik trizomi, 47, XX/XY + 13, şeklinde olup daha nadir olarak translokasyon ve mozaisizm gibi kromozomal düzensizlikler de görülmektedir (4,5). Trizomi 13 ya da Patau Sendromu, Trizomi 21 (Down Sendromu) ve Trizomi 18'den (Edward sendromu) sonra en yaygın görülen otozomal trizomi sendromudur (6).

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Annenin ileri yaşta olması, görülme sıklığını artırmaktadır. Trizomi 13 görülen gebeliklerde spontan düşük sıklığı, canlı doğumuna göre yüz kat daha fazladır. Trizomi 13, tüm spontan düşüklerin yaklaşık %1'inden sorumludur (7).

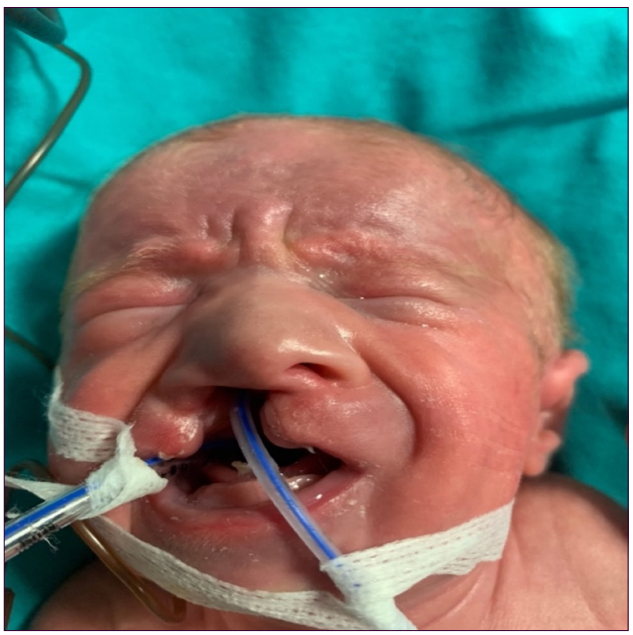
Bu vakada, aplasia kutis ve göz bulgularının eşlik ettiği Trizomi 13 vakası sunulmuştur.

OLGU

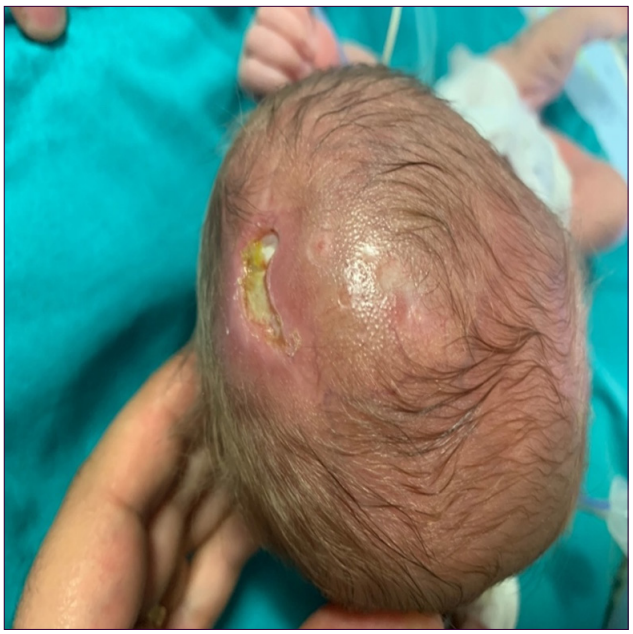
Tokat Gaziosmanpaşa Üniversitesi Hastanesi'nde, otuz yaşındaki annenin ikinci gebeliğinden ikinci yaşayan olarak 32 haftalıkken sezaryen ile doğdu. Anne ve baba arasında akrabalık yok.

Takipli gebelik olup; taramalar sonucu IUGR eşlik etmesi nedeniyle perinatoloji takibi önerilmiş. Perinatoloji takibinde yarık damak dudak tespit edilmiş olup gebeğin terminasyonu önerilmiş fakat aile gebeliğin devamını istemiş. Annede gestasyonel diyabet mevcut olup gebelikte insülin kullanım öyküsü var. Fetal distres nedeniyle acil sezaryen doğum sonrası bebeğinin APGAR skoru 1. dakikada 5 ve 5. dakikada 6 idi. Solunum çabasının yeterli olmaması nedeniyle ameliyathane salonunda entübe edildi.

Fizik muayenesinde; ağırlığı 1405 gram (3 persentil altı), boyu 41 cm (3 persentil altı), baş çevresi 26 cm (3 persentil altı) idi. Dismorfik görünümü ve ön fontonel boyutu 3×4 cm olan hastada mikrosefali, skalpte sol paryetal alanda aplazia kutis konjenita, sağ gözde mikroftalmi, sol gözde anoftalmi, yarık damak ve dudak mevcuttu (**Resim 1,2**). Üç ekstremitede polidaktili olduğu görüldü (**Resim 3**).



Resim 1.



Resim 2.

Resim 3.

Abdomen ultrasonografi (USG), kranial USG ve beyin bilgisayarlı tomografide patoloji görülmedi. Ekokardiyografisi normal olarak değerlendirildi. Sağ gözde konjenital katarakt ve mikroftalmi mevcutken; sol göz anoftalmikti.

TARTIŞMA

Trizomi 13'ün yaklaşık insidansı 10.000 canlı doğumda birdir (1). Patau sendromlu hastalarda genelde, fazla olan kromozom anne kaynaklıdır. Hastaların çoğu izole vakalar olduğundan dolayı, anne-baba karyotip analizi endikasyonu bulunmamaktadır (8).

Kesin tanı için periferik kan örneğinden kromozom analizi yapılması gereklidir. Patau sendromlu vakaların %90'ında Trizomi 13, %5-10'unda dengesiz Robertsonian 13;14 translokasyonu ve az sayıda vakada mozaik trizomi 13 saptanır (6). Vakamızda; hastanın periferik kandan yapılan moleküler karyotip analizi 47, XX, + 13 ile uyumlu olup, FISH analizi sonucu trizomi 13 ile uyumlu olarak değerlendirilmiştir.

Trizomi 13 sendromunun en sık görülen bulguları motor ve mental gerilik, mikrosefali, mikroftalmi, holoprozensefali, hipotelorizm, yarık damak ve/veya yarık dudak, kardiovasküler, genitoüriner ve oküler malformasyonlardır. Mikroftalmi, yarık damak ve yarık dudak, polidaktili olması bu sendrom için karakteristiktir (9,10). Patau sendromu olgularında %60-80 oranında yarık damak-dudak, %60-70 oranında holoprozensefali, %60-70 oranında göz anomalileri, %60-70 oranında polidaktili görülmektedir (11). Vakamızda yarık damak-dudak, polidaktili, sol göz anoftalmi, sağ göz mikroftalmi ve konjenital katarakt, mikrosefali, mikrognati eşlik etmektedir.

Bununla birlikte Trizomi 13 vakalarında görülme sıklığı yüksek olan kardiyak anomali, santral sinir sistemi gelişim anomalisi, üriner sistem anomalisi ise görülmemiştir.

İleri anne yaşı bu sendrom için en önemli risk faktörleri arasındadır. Anne yaşı kromozom ayrılmamasına (nondisjunction) neden olmaktadır. Bu olgularda ortalama anne yaşı 30.9 olarak bildirilmiştir (12). Bizim vakamızda anne yaşı 30 idi.

SONUÇ

Trizomi 13 antenatal dönemde tanı konulabilen ve yüksek derecede ölümcül seyreden bir kromozom bozukluğudur. Olgumuzda ileri anne yaşı da göz önüne alındığında sonraki gebeliklerde genetik danışmanlık alması önerilmiştir.

ETİK BEYANLAR

Aydınlatılmış Onam: Bu çalışmaya katılan hasta(lar)dan yazılı onam alınmıştır.

Hakem Değerlendirme Süreci: Harici çift kör hakem değerlendirmesi.

Çıkar Çatışması Durumu: Yazarlar bu çalışmada herhangi bir çıkara dayalı ilişki olmadığını beyan etmişlerdir.

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Yazar Katkıları: Yazarların tümü; makalenin tasarımına, yürütülmesine, analizine katıldığını ve son sürümünü onayladıklarını beyan etmişlerdir.

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CASE REPORT
OLGU SUNUMU

Acute Pleuro-Pericarditis due to Parvovirus B19: A Case Report of A 17-Year-Old Boy

Parvovirus B19'a Bağlı Akut Pleuro-Perikardit: 17 Yaşındaki Bir Erkek Çocuğun Olgu Sunumu

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Abstract

We report on a seventeen-year-old boy with acute pleuro-pericarditis with human parvovirus B19 (PVB19) infection. He presented with chest pain, fever and shortness of breath. On physical examination, he had orthopnea, increased temperature (38,4 °C), tachycardia and hepatomegaly. Echocardiography showed a pericardial effusion of 12 mm. Thoracic ultrasound revealed left pleural effusion of 10 mm. Serum anti-Parvovirus 19 IgM and Parvovirus B19 DNA were positive. Two weeks later; serum anti-Parvovirus B19 IgM was negative, pericardial effusion and pleural effusion were resolved. In our knowledge this case represents the first report of acute pleuro-pericarditis associated with Parvovirus B19 infection in a pediatric patient.

Keywords: Chest pain, parvovirus B19, pleuro-pericarditis

Öz

Parvovirüs B19 (PVB19) enfeksiyonu olan akut plöro-perikarditli on yedi yaşında bir erkek çocuğu rapor ediyoruz. Göğüs ağrısı, ateş ve nefes darlığı şikayetleri ile başvurdu. Fizik muayenede ortopne, ateş yüksekliği (38,4 °C), taşikardi ve hepatomegali vardı. Ekokardiyografide 12 mm perikardiyal efüzyon görüldü. Göğüs ultrasonunda 10 mm'lik sol plevral efüzyon saptandı. Serum anti-Parvovirus 19 IgM ve Parvovirus B19 DNA pozitifti. İki hafta sonra; serum anti-Parvovirus B19 IgM negatifti, perikardiyal efüzyon ve plevral efüzyon düzeldi. Bildiğimiz kadarıyla bu vaka, bir pediatrik hastada Parvovirus B19 enfeksiyonu ile ilişkili ilk akut plöro-perikardit raporunu temsil etmektedir.

Anahtar Kelimeler: Göğüs ağrısı, parvovirus B19, plöro-perikardit

INTRODUCTION

Viruses are common etiology of pericarditis among children (1,2). Although enteroviruses are the most common pathogens of this condition, another viruses such as Herpes Simplex, Rubella can be cause of the viral pericarditis (2). While Human Parvovirus B19 is largely seen in the population, it can be etiological cause of erythema infectiosum (fifth disease), symmetric polyarthropathy, transient erythroblastopenia, pericarditis, hydrops fetalis and fetal myocarditis (3). Currently, cases of perimyocarditis (4), pericarditis (5), pleuro-pericarditis and pericarditis-related heart failure (6) associated with Human Parvovirus B19 infection have also been reported in adult patients, on the contrary data is mostly limited in pediatric patients. We presented a seventeen-year-old boy who presented with acute pleuro-pericarditis associated with Human Parvovirus B19.

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CASE REPORT

A seventeen-year-old boy presented with a 4-days history of precordial chest pain, dyspnea, fever and difficulty standing in supine position. Neither anemia nor articular pain was associated. On physical examination, patient had orthopnea, a weak apperance, increased temperature (38.4°C) and a respiratory rate of 28/min. His heart rate was 122/min, blood pressure was 90/68 mmHg, and peripheral pulses were weak. Heart sounds were muffled in cardiac examination. Other findings of physical examination were normal. Laboratory results were as follows: hemoglobin 14.1 g/dl, hematocrit 41.5 %, red blood cells 4.74x106/µl, white blood cells 14,500/mm3 (9% lymphocyte, 84% neutrophil, 6% monocyte, 1% eosinophil), platelet count 228,000/mm3, erythrocyte sedimentation rate 6 mm/h (normal <25 mm/h) and C-reactive protein 113 mg/l (normal <5 mg/l). Serum biochemistry showed normal levels of electrolytes, urea, creatinine, glucose, liver enzymes, troponin and creatinine kinase. Values for thyroid functions were in the normal range. Telecardiography of the patient was with in normal limits with cardio-thoracic ratio of 50% and electrocardiography showed ST-segment elevation in the lateral/inferior leads (**Figure 1**). Echocardiography performed pericardial effusion measuring 11-12 mm in diameter with a little amount of fibrin, no signs of tamponade and normal systolic functions (**Figure 2**). Thoracic ultrasound revealed pleural effusion. Urinary culture and hemoculture were negative. Tuberculin skin test showed no signs of tuberculosis. Conderning involvement of collagen tissue diseases, anti-DNA, antinuclear antibodies, anti-cardiolipin antibodies and rheumatoid factor were negative. Serum IgM antibodies for EBV, HIV, VZV, adenovirus and coxsackievirus were found to be negative ELISA. DNA analysis by polymerase chain reaction (PCR) and serum anti-Parvovirus B19 IgM by ELISA and yielded positive results. Peripheral smear of the patient was normal. The patient's condition improved with the administration of non-steroidal anti-inflammatory drugs (NSAIDs) and bed rest. He was discharged after a two-weeks stay in hospital with complete resolution of symptoms.

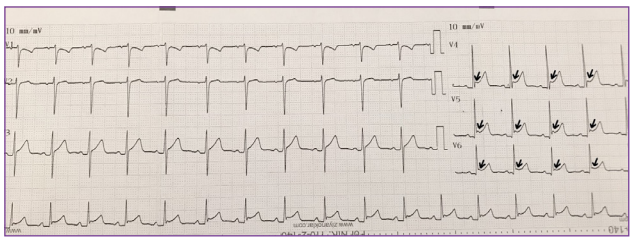

Figure 1. ST-segment elevation in the lateral/inferior leads

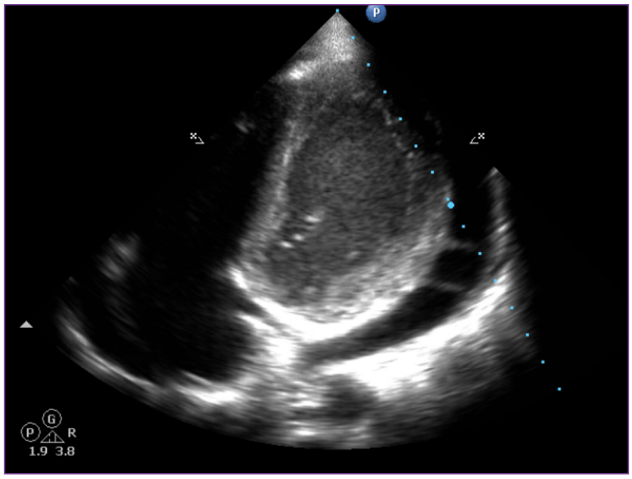

Figure 2. Two-dimensional echocardiogram showing a large Pericardial effusion and a little amount of fibrin

DISCUSSION

Viral pericarditis is the most common cause of acute pericarditis in the pediatric population (7). Although enteroviruses may be responsible for this clinical condition, coxsackie B virus is the most common causes of pericarditis and the disease may be caused by other viruses such as EBV, HIV, adenovirus and VZV (1,7,8). As patients with viral pericarditis often present with fever and precordial chest pain, they are generally less toxic appearing than patients with bacterial pericarditis. But, if myocarditis accompanies, clinical presentation can worsen (7). In our case, the main presenting symptoms were precordial chest pain, dyspnea, and fever. Although Parvovirus B19 infection frequently causes erythema infectiosum (fifth disease) in pediatric patients, less frequently, it can also lead to polyarthropathy, hydrops fetalis, fetal myocarditis, transient erythroblastopenia (4,9,10). The infection is generally diagnosed with physical examination, clinical symptoms, serological tests, and by the detection of viral DNA with PCR (11). Pleuro-pericarditis associated with Human Parvovirus B19 infection has also been reported in adult patients (4,6). In this comprehensive review of literature, no case of pleuro-pericarditis was retrieved in pediatric patients. Seishima et al. (6) reported Human Parvovirus B19 infection in a male patient who presented with fatigue, polyarthralgia and edema, and developed acute heart failure due to Parvovirus B19 associated pericarditis five days after admission. In another study , a 34-year old man who was diagnosed to have perimyocarditis induced with human Parvovirus B19 by showing anti-Parvovirus B19 Ig M and Ig G antibodies in the blood with ELISA. In the present case, the patient presented with complaints of precordial chest pain, dyspnea, fever and the diagnosis of pleuro-pericarditis was determined by clinical, electrocardiographic, echocardiographic and laboratuar findings. At first, bacterial and other viral pathogens, neoplasias and collagen tissue diseases were excluded.

CONCLUSION

This case seems to be the first reported pediatric case of pleuro-pericarditis due to Parvovirus B19 infection. Parvovirus B19 infection might be considered in the etiology of acute pleuro-pericarditis in pediatric patients.

ETHICAL DECLARATIONS

Informed Consent: Written informed consent was obtained from all participants who participated in this study.

Referee Evaluation Process: Externally peer-reviewed.

Conflict of Interest Statement: The authors have no conflicts of interest to declare.

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

Author Contributions: All of the authors declare that have all participated in the design, execution, and analysis of the paper, and that they have approved the final version.

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