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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

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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)
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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

The Role of Thiol-Disulfide and Ischemia-Modified Albumin in The Differential Diagnosis of Acute Scrotum in Children

Çocuklarda Akut Skrotumun Ayırıcı Tanısında Tiyol-Disülfid ve İskemi Modifiye Albüminin Rolü

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ABSTRACT

Aim: Testicular torsion, which is among the most common causes of acute scrotum, is a true surgical emergency. However, despite advanced imaging techniques and laboratory tests, the differential diagnosis from other pathologies causing acute scrotum is difficult. Therefore, more specific laboratory tests are needed to confirm the diagnosis. We aimed to investigate the usability of a laboratory test in the differential diagnosis of acute scrotum in children by measuring thiol-disulfide and ischemia modified albumin (IMA) levels.

Material and Method: Of 60 male children, 30 with acute scrotum and 30 healthy boys who were admitted for circumcision were included. The levels of native thiol (SH), total thiol (SH+SS), dynamic disulfide (SS), SS/SH+SS percentage rate, albumin and IMA were measured. Patients' pre-operative and postoperative first-day blood test results were compared.

Results: The SH (p=0.025), SH+SS (p=0.032), SS (p=0.045), albumin (p<0.001) and IMA (p<0.001) levels of the acute scrotum group were significantly higher than those of the control group. The IMA levels of the epididymo-orchitis subgroup of acute scrotum were found to be higher than those of the testicular torsion subgroup. Pre- and post-operative results of the acute scrotum group were similar for all variables (p>0.05).

Conclusion: The evaluation of thiol-disulfide homeostasis and IMA levels and the detection of changes in favour of oxidative stress might help in the differential diagnosis of acute scrotum, but those results cannot help differentiate testicular torsion from an epididymo-orchitis diagnosis.

Keywords: Acute scrotum, children, IMA, thiol-disulfide

ÖZ

Amaç: Akut skrotumun en sık nedenleri arasında yer alan testis torsiyonu, gerçek bir cerrahi acildir. Ancak ileri görüntüleme teknikleri ve laboratuvar tetkiklerine rağmen akut skrotuma neden olan diğer patolojilerden ayırt edilmesi zordur. Bu nedenle, tanıyı doğrulamak için daha spesifik laboratuvar testlerine ihtiyaç vardır. Çalışmada, tiyol-disülfid ve iskemi modifiye albümin (İMA) düzeylerini ölçerek çocuklarda akut skrotumun ayırıcı tanısında bir laboratuvar testinin kullanılabilirliğini araştırmayı amaçladık.

Gereç ve Yöntem: 30'u akut skrotumlu ve 30 sünnet için başvuran sağlıklı erkek çocuk olamk üzere 60 erkek çocuk çalışmaya dahil edildi. Native tiyol (SH), total tiyol (SH+SS), dinamik disülfid (SS), SS/SH+SS yüzde oranı, albümin ve IMA seviyeleri ölçüldü. Hastaların ameliyat öncesi ve ameliyat sonrası ilk gün kan testi sonuçları karşılaştırıldı.

Bulgular: Akut skrotum grubunda SH (p=0,025), SH+SS (p=0,032), SS (p=0,045), albümin (p<0,001) ve İMA (p<0,001) düzeyleri, anlamlı olarak kontrol grubundan daha yüksek bulundu. Akut skrotumun epididimo-orşit alt grubunun İMA düzeyleri testis torsiyonu alt grubuna göre daha yüksek bulundu. Akut skrotum grubunun ameliyat öncesi ve sonrası sonuçlarında anlamlı farklılık yoktu (p>0.05).

Sonuç: Tiyol-disülfid dengesi ve IMA düzeylerinin değerlendirilmesi sonucu oksidatif stres lehine değişikliklerin saptanması akut skrotumun ayırıcı tanısında yardımcı olabilir, ancak bu sonuçlar testis torsiyonunu epididimo-orşit tanısından ayırmaya yardımcı olamaz.

Anahtar Kelimeler: Akut skrotum, çocuklar, IMA, tiyol-disülfid

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INTRODUCTION

Acute scrotum is a pathology that presents itself in the inguinoscrotal region with sudden pain, swelling and redness due to various aetiological reasons.[1,2] The most common cause of acute scrotum is testicular torsion.[3] Testicular torsion is a true surgical emergency because the longer the testis remains torsed, the less the chance of recovery.[3] While advanced imaging techniques and laboratory studies are used in the differential diagnosis of pathologies causing acute scrotum, it is not always easy to confirm this diagnosis.[4] Surgical exploration of acute scrotum (as a treatment) ensures that testicular torsion is not overlooked; however, it is still controversial due to some considering it an unnecessary surgical intervention.[5] Therefore, there is a need for laboratory parameters that confirm the diagnosis of testicular torsion.

In inflammatory events, the activation of neutrophils and macrophages and excessive production of reactive oxygen species (ROS) increase oxidative stress (OS).[6-8] Damage due to ROS is prevented by enzymatic or nonenzymatic antioxidant mechanisms, including super-oxide dismutase, catalase and glutathione S-trans enzyme systems and important biological thiols, such as glutathione, cysteine, homocysteine, N-acetyl cysteine and gamma glutamine. Thiol is an organic compound that contains a -SH group, which plays a critical role in the prevention of OS in cells. SH-containing amino acid groups in proteins are primary targets of ROS.[9]

In the same medium with ROS, the -SH groups are oxidised and form reversible disulfide bonds. Loss of thiol groups is the main molecular mechanism that results in structural and functional changes in proteins. Antioxidants, especially thiol groups that try to prevent the destructive effects of free radicals, cannot maintain plasma and tissue levels during these interactions.[10] However, the formed disulfide bonds can be reduced to thiol groups by the cellular reduction effects of some antioxidants; thus, thiol-disulfide homeostasis is preserved.[11]

The ischemia modified albumin (IMA) test is an FDA-approved test among newly investigated cardiac markers.[12] The principle of the test is based on measuring the cobalt-binding capacity of albumin, leading to chemical changes in the albumin during ischemia. This new albumin molecule is also called IMA. The formation of this new albumin molecule, which has lost its ability to bind cobalt, is one of the earliest markers of ischemia.[13] However, recent studies suggest that IMA, which stands out as a marker of cardiac ischemia, might very well increase in different pathologies.[14–16]

Our study aimed to investigate the usability of the diagnostic as a laboratory parameter in the differential diagnosis of acute scrotum causing inflammation in children via thiol–disulfide and IMA levels.

MATERIAL AND METHOD

In this study, a total of 60 male children, including 30 boys who were admitted to the paediatric surgery clinic due to acute scrotum (patient group) and 30 healthy boys who were admitted for circumcision (control group), were included. Acute scrotum was diagnosed with physical examination and absence of blood flow of testis on the scrotal doppler ultrasonography. The study was conducted prospectively in our hospital between 1st April 2017 and 1st April 2018. Study permission was obtained from the ethics committee of the Ankara Children's Hematology Oncology Education and Training Hospital (decision no. 2017/03 dated 27/03/2017). Informed consent forms were obtained from the parents of the patients and controls included in this study.

The thiol–disulfide and IMA levels of the patients who were operated on for acute scrotum were compared with those of the control group. Thirty boys who were operated for acute scrotum were were divided into two sub-groups as testicular torsion or epididymorochitis by diagnosed at surgery. The thiol–disulfide and IMA levels were compared between those groups too. The patients' blood samples were taken in the pre-operative period and on the first post-operative day. Two ml of blood were taken from the cases and placed in yellow cap gel tubes (BD Vacutainer plastic SST II tube®). Blood samples were collected after centrifuging at 3,600 rpm for 10 minutes, and 2 ccs of serum were obtained and stored at -80°C in the biochemistry laboratory. After all the samples were collected, they were thawed at the same time, and the blood thiol–disulfide parameters were determined by Erel and Neselioglu's newly developed automatic measurement method.[17] The IMA levels were studied in a Roche Hitachi Cobas c501 automatic analyser with the colorimetric method, as described by Bar-Or et al.[18] at Ankara Ataturk Training and Research Hospital's biochemistry laboratory.

The levels of SH, SH+SS, SS, SS/SH+SS, albumin and IMA were measured from the sera of patients and controls. These results of patients and control grups were examined by comparing the differences.

Statistical Analyses

Statistical analysis was performed using the Statistical Package for the Social Sciences (SPSS) 17.0 (Chicago Inc., 2008) programme. Continuous variables' normal distribution was checked with the Shapiro–Wilk test. While the SH+SS and albumin levels of the two groups were found to have normal distribution, the IMA, SH, SS and SS/SS+SH ratio values did not. For the normally distributed variables, the mean values of the variables were analysed using the Student's t test and ANOVA testing. Other variables were analysed using the Mann–Whitney U test and Kruska–Wallis H Test, with $p<0.05$ considered significant.

RESULTS

Demographics data

The mean age of the patients in the acute scrotum group was 12.12 years (standard deviation=4.63, range: 0.2–18 years), and the control group was 7.6 years (standard deviation=2.54, range: 3–13 years). There was no statistical difference between the groups. (p=0,46).

Laboratory results

The SH, SH+SS, SS, SS/SS+SH percentage ratio, IMA and albumin levels of the acute scrotum and control groups and their statistical analysis are shown in **Table 1**.

The SH (p=0.025), SH+SS (p=0.032), SS (p=0.045), albumin (p<0.001) and IMA (p<0.001) levels of the acute scrotum group were found to be significantly higher than those of the control group. Although the SS/SH+SS percentage ratio was higher than that of the control group, it was not statistically significant (p=0.38).

Patients in the acute scrotum group were divided into two sub-groups: children with testicular torsion and children with epididymo-orchitis. The two groups were also compared in terms of their SH, SH+SS, SS, SS/SH+SS percentage ratio, albumin and IMA levels (**Table 2**).

There was no statistically significant difference between the two sub-groups in terms of SH, SH+SS, SS, SS/SH+SS percentage ratio and albumin levels, whereas the IMA levels of the epididymo-orchitis group were significantly higher than those of the testicular torsion group.

Table 1. The comparison of the acute scrotum (n=30) and controls (n=30)' laboratory findings

Variables	Groups	n	Mean	SD	p value
Native thiol (SH)					0.025*
	Acute scrotum	30	421	49.41685	
	Control	30	393.723	54.74252	
Total thiol (SH + SS)					0.032**
	Acute scrotum	30	467.6	57.92349	
	Control	30	434.62	58.5673	
Disulfide (SS)					0.045*
	Acute scrotum	30	23.2667	6.15284	
	Control	30	20.4333	4.27247	
Disulfide (SS) / Total thiol (SH+SS) %					0.308*
	Acute scrotum	30	4.9339	0.89738	
	Control	30	4.7209	0.93069	
Ischemia-modified Albumin (IMA)					0.00*
	Acute scrotum	30	0.5826	0.09985	
	Control	30	0.7003	0.14878	
Albumin					0.00**
	Acute scrotum	30	4.76	0.26987	
	Control	30	4.41	0.32626	

n: frequency, SD: standard deviation, *: Mann Whitney U test, **: Student t test

In the acute scrotum group, the pre- and post-operative levels of SH, SH+SS, SS, SS/SH+SS percentage rate, albumin and IMA were compared with a paired correlation test (see **Table 3**). All pre- and post-operative variables were similar (p>0.05).

Table 2. Testicular torsion (n=16) and epididymo-orchitis (n=14)' laboratory findings

Variables	Groups	n	Mean	SD	p value
Native thiol (SH)					0.308**
	Testicular torsion	16	429.75	40.84354	
	Epididymo-orchitis	14	411	57.62666	
Total thiol (SH + SS)					0.417*
	Testicular torsion	16	475.794	46.07285	
	Epididymo-orchitis	14	458.236	69.70064	
Disulfide (SS)					0.76**
	Testicular torsion	16	22.9375	4.50879	
	Epididymo-orchitis	14	23.6429	7.79158	
Disulfide / Total thiol %					0.428**
	Testicular torsion	16	4.81	0.77489	
	Epididymo-orchitis	14	5.0755	1.03107	
Ischemia-modified Albumin					0.014**
	Testicular torsion	16	0.5417	0.07212	
	Epididymo-orchitis	14	0.6294	0.10878	
Albumin					0.093*
	Testicular torsion	16	4.8375	0.26552	
	Epididymo-orchitis	14	4.6714	0.25549	

n: frequency, SD: standard deviation, *: Mann Whitney U test, **: Student t test

Table 3. Acute scrotum group (n=30)'s pre- and post-operative laboratory findings

Variables	Mean	SD	p value
Native thiol (SH) (pre-op)	427.25	40.66905	0.465
Native thiol (SH) (post-op 1st day)	370.84	40.38713	
Total thiol (SH + SS) (pre-op)	475.58	46.42073	0.437
Total thiol (SH + SS) (post-op 1st day)	412.59	43.70198	
Disulfide (SS) (pre-op)	24.2	5.0728	0.223
Disulfide (SS) (post-op 1st day)	20.9	6.15449	
Disulfide/Total thiol % (pre-op)	5.0734	0,84905	0.059
Disulfide/Total thiol % (post-op 1st day)	5.0717	1,51395	
Ischemia-modified Albumin (pre-op)	0.545	0.06173	0.665
Ischemia-modified Albumin (post-op 1st day)	0.6024	0.06547	
Albumin (pre-op)	4.89	0.2079	0.606
Albumin (post-op 1st day)	4.46	0.21187	

Sixteen patients with testicular torsion were evaluated in terms of the time period between the onset of symptoms and their admission to the emergency department. The cases were divided into three groups (Group 1: 0–6 hours; Group 2: 7–24 hours; Group 3: more than 24 hours) and evaluated for statistical differences in their laboratory findings. There was no difference between the three groups in terms of their SH (p=0.890), SH+SS (p=0.894), SS (p=0.987), SS/SH+SS percentage ratio (p=0.940), albumin (p=0.776) and IMA (p=0.293) levels (one-way ANOVA test).

DISCUSSION

Testicular torsion, which is among the most common causes of acute scrotum, is a surgical disease affecting newborns, children and adolescents. Twisting of the torsion of the spermatic cord around the longitudinal axis might result in venous congestion, oedema and arterial occlusion, and if left untreated, gonadal necrosis may develop.[19] The longer the testicle remains torsed, the more the testicular tissues remain ischemic, which can result in necrosis formation, making testicular torsion a true surgical emergency.[4] Despite the advanced imaging techniques and laboratory studies currently used in the differential diagnosis of pathologies causing acute scrotum, there are no biochemical markers to demonstrate testicular torsion.

The state of dynamic thiol–disulfide balance plays a critical role in antioxidant protection, detoxification, signal transduction and apoptosis in the regulation of enzymatic activity, transcription factors and cellular signalling mechanisms.[20] In addition, distortions in the dynamic thiol–disulfide balance have been shown to play a role in the pathogenesis of many diseases, such as diabetes, cardiovascular diseases, cancer, rheumatoid arthritis, chronic kidney disease, AIDS, Parkinson's disease, Alzheimer's disease, Friedrich ataxia, multiple sclerosis, amyotrophic lateral sclerosis and liver diseases.[21] Therefore, the determination of the dynamic thiol-disulfide balance might provide valuable information regarding various normal or abnormal biochemical processes.[21] Several studies have shown the predictive value of thiol-disulfide haemostasis in organ ischemia. The measurement of plasma or serum thiol-disulfide balance via the colorimetric method was developed by a Turkish scientist.[21]

There are many studies on OS markers and their relationship with various diseases, such as acute appendicitis, in children,[22–24] yet no studies exist on the relationship between acute scrotum and OS markers in children. Additionally, many studies have explored the relationship between thiol-disulfide and testis torsion; however, our study is important as the first to show thiol–disulfide and IMA levels in the differential diagnosis of acute scrotum in children.

A rat model experimental study by Menteşe et al. showed that thiol-disulfide haemostasis might be a haematologic parameter in the prognosis of early testicular ischaemia, revealing a positive correlation between the decrease in SH+SS values and histopathologic injury in the testis following ischaemia–reperfusion.[25] Their study also showed that ischaemia-modified albumin could be predictive of testicular injury in short- (2 hr) and long-term (2 months) testicular torsion, which could be a marker for fertility capacity.[25]

In our study, the SH+SS/SS homeostasis markers' of the SH+SS, SH and SS levels of the patients in the acute scrotum group were significantly increased compared to those of the control group, except for the SS/SH+SS percentage rate, which was similar to that of the control group. The IMA and albumin levels also increased statistically.

There was no difference between the two sub-groups of acute scrotum regarding their SH+SS, SH, SS, SS/SH+SS percentage ratio, albumin and IMA levels. These results indicate that in the acute scrotum group, OS shifted towards the oxidant side; hence, there was an OS increase. However, there was no difference in the levels of thiol-disulfide balance and IMA when the testicular torsion and epididymo-orchitis cases were compared. Therefore, thiol-disulfide balance and IMA levels cannot be used as markers for the differentiation of testis torsion and epididymo-orchitis, which are the most common causes of acute scrotum in children. There was no statistical difference in the SH+SS, SH, SS, SS/SH+SS percentage ratio, albumin and IMA levels in the acute scrotum cases' pre- and postoperative blood samples. These results indicate that the thiol–disulfide balance in the acute scrotum cases shifted towards the oxidative side, resulting in increased OS levels.

CONCLUSION

The development of a specific test in the differential diagnosis of acute scrotum in children would assist with early diagnosis and thus early surgery in testicular torsion as well as prevent unnecessary surgical interventions. Physical examination of findings, imaging methods and current laboratory tests, along with the evaluation of thiol-disulfide homeostasis and IMA levels and the detection of changes in favour of OS, might help in the differential diagnosis of acute scrotum. However, these results cannot help to distinguish testicular torsion from an epididymo-orchitis.

ETHICAL DECLARATIONS

Ethics Committee Approval: Study permission was obtained from the ethics committee of the Ankara Children's Hematology Oncology Education and Training Hospital (decision no. 2017/03 dated 27/03/2017).

Informed Consent: Informed consent forms were obtained from the parents of the patients and controls included in this study.

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ORIGINAL ARTICLE
ORİJİNAL ARAŞTIRMA

Comparison of Tube Thoracostomy and Thoracoscopic Debridement in the Treatment of Empyema in Children

Çocuklarda Ampiyem Tedavisinde Tüp Torakostomi ile Torakoskopik Debridmanın Karşılaştırılması

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ABSTRACT

Objective: Empyema is an important problem that develops after pneumonia in children. Antibiotherapy and drainage of purulent fluid form the basis of treatment. Various methods are used for drainage. Thoracoscopic debridement is widely used today with the development of minimally invasive methods. In this study, patients who were treated for empyema in the period before the use of thoracoscopic debridement in our clinic were compared with the patients who were treated in the period after the use of thoracoscopic debridement.

Material and Method: For this purpose, cases before thoracoscopic debridement (Group 1, n = 25) and patients after thoracoscopic debridement (Group 2, n = 28) were compared in various aspects. While tube thoracostomy was applied to all cases in the first group, thoracoscopic debridement was applied to the cases in the second group whether or not tube thoracostomy was applied.

Results: Fever, leukocytosis, respiratory distress and purulent drainage lasted shorter in the group in which thoracoscopic debridement was applied, in other words, clinical improvement was faster. In the group in which thoracoscopic debridement was applied, the duration of tube thoracostomy was shorter and the need for thoracotomy was less than in the group not applied.

Conclusion: Thoracoscopic debridement accelerates the recovery of empyema in children. In this, it is effective to debride fibrin and septations in the pleural space more effectively and quickly under the camera view. Therefore, thoracoscopic debridement should be the first option in the treatment of empyema without delay.

Keywords: Empyema, Thoracoscopic debridement, Tube thoracostomy, children

ÖZ

Amaç: Ampiyem çocuklarda, pnömoni sonrası gelişen önemli bir sorundur. Antibiyoterapi ve pürülan sıvının drenajı tedavinin temelini oluşturur. Drenaj amacıyla çeşitli yöntemler kullanılır. Minimal invaziv yöntemlerin de gelişmesiyle günümüzde torakoskopik debridman yaygın olarak kullanılmaktadır. Bu çalışmada, kliniğimizde torakoskopik debridman kullanılmaya başlamadan önceki dönemde ampiyem nedeniyle tedavi edilen olgularla, torakoskopik debridman kullanılmaya başladıktan sonraki dönemde tedavi edilen olgular karşılaştırılmışlardır.

Gereç ve yöntem: Bu amaçla kliniğimizde torakoskopik debridman öncesi olgular (Grup 1, n=25) ile torakoskopik debridman sonrası olgular (Grup 2, n=28) çeşitli yönleriyle karşılaştırılmışlardır. İlk gruptaki tüm olgulara tüp torakostomi uygulanırken, ikinci gruptaki olgulara öncesinde tüp torakostomi uygulansın ya da uygulanmasın torakoskopik debridman uygulanmıştır.

Bulgular: Torakoskopik debridman uygulanan grupta, uygulanmayan gruba göre ateş yüksekliği, lökositoz, solunum sıkıntısı ve pürülan drenaj daha kısa sürmüş, yani klinik iyileşme daha hızlı olmuştur. Torakoskopik debridman uygulanan grupta, uygulanmayan gruba göre tüp torakostomi süresi daha kısa, torakotomi ihtiyacı daha az saptanmıştır.

Sonuç: Torakoskopik debridman, çocuklarda ampiyemde iyileşmeyi hızlandırmaktadır. Bunda, plevral boşluktaki fibrin ve septasyonların kamera görüşü altında daha etkili ve hızlı debride edilmesi etkilidir. Bu nedenle ampiyem tedavisinde ilk seçenek zaman kaybetmeden torakoskopik debridman olmalıdır.

Anahtar kelimeler: Ampiyem, torakoskopi, debridman, tüp torakostomi, çocuk

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INTRODUCTION

Empyema is an accumulation of purulent fluid in the pleural space. It occurs usually after pneumonia in children. Empyema develops in 2-8% of children hospitalized for pneumonia (1). Despite advances in diagnosis and treatment, empyema remains as an important cause of morbidity (2). While the incidence of pneumonia has decreased in children recently, the rate of parapneumonic effusion and empyema has risen (3). The conventional treatment of empyema involves drainage of fluid from the pleural space through tube thoracostomy. However, the success rate of tube thoracostomy alone is rather low in the presence of dense and loculated pleural fluid. Administration of fibrinolytic agents along with tube thoracostomy may also fail, especially in advanced stage empyema (4). With the development of minimally invasive methods, currently thoracoscopic debridement is widely used in empyema (5,6,7,8). Owing to thoracoscopy, fibrin and septa can be successfully debrided by opening the loculated areas in the pleural space under direct vision.

In this study, the aim was to investigate the effects of thoracoscopic debridement on clinical improvement and duration of tube thoracostomy in the treatment of empyema in children. For this purpose, patients who were treated for empyema before the use of thoracoscopic debridement in our clinic were compared with those who were treated in the period after the use of thoracoscopic debridement started.

MATERIAL AND METHOD

Medical records of patients who were diagnosed with empyema and underwent tube thoracostomy or thoracoscopic debridement as the first treatment were retrospectively reviewed. The patients who were treated for empyema before 2001 when thoracoscopic debridement was implemented in our clinic (Group 1, n=25) were compared with those who received treatment in 2001 and after (Group 2, n=28) (53 cases in total). All empyema patients who underwent tube thoracostomy or thoracoscopic debridement as the first treatment were included in the study and no cases were excluded from the study.

Treatment approach in empyema cases

Patients suspected of empyema due to clinical and physical examination findings were evaluated by ultrasound and/or computed tomography scan in addition to chest X-rays. The investigation was directed to find out whether there was pleural fluid, pneumothorax, cavitation, loculation, atelectasis and pleural thickening in imaging methods. First, thoracentesis was performed and the characteristics of the fluid were evaluated. Cell count and type were evaluated by microscopic examination, and microorganisms were searched by gram staining. In the biochemical examination, pH, protein, glucose and LDH values in the fluid were tested and simultaneously compared with the protein, glucose and LDH values in blood biochemistry. Furthermore, antibiotic susceptibility tests were performed for each culture on the collected fluids. Single, dual or multiple broad-spectrum antibiotherapy was administered to the patients depending on their clinical conditions and antibiogram results. Patients were followed up in the intensive care unit during the hospitalization, and were transferred to the ward when their clinical conditions were stable. Vital functions (body temperature, heart rate, blood pressure, respiratory rate and peripheral oxygen saturation) and fluid balance of the patients were monitored hourly in the intensive care unit and at appropriate intervals during their ward stay.

Tube thoracostomy was performed as the first step in treatment of all patients in the first group upon detection of fluid. In the second group, thoracoscopic debridement was performed without prior tube thoracostomy in the patients who had loculated or thick pleural fluid in the radiological examinations and/or when no fluid could be removed by thoracentesis (in cases where fluid was observed radiologically but could not be removed by thoracentesis, it was assumed that the fluid was loculated and fibrinous). While tube thoracostomy was performed first in the other cases, thoracoscopic debridement was performed afterwards, since there was no clinical improvement. Patients in both groups were followed up with intermittent chest X-rays. The amount, characteristics of the drainage material and air outlet from the tube were recorded daily. Hematological and biochemical values of the patients were checked at certain intervals. Tube thoracostomies were terminated in the patients whose clinical and radiological findings regressed (lung expansion, fluid and improvement in pneumothorax) and whose drainage from the tube ceased. Empyema of the patients was staged according to clinical, laboratory, radiological examination and operational findings.

Thoracoscopic debridement

Thoracoscopic debridement was performed under general anesthesia in the lateral decubitus position in all cases. During the procedure, CO_2 gas at a pressure of 5–8 mmHg was insufflated into the pleural space. A 5 mm trocar was placed on the area where the highest amount of fluid was found radiologically (usually the point where the 4th and 6th intercostal space intersected with the midaxillary line) and the pleural space was visualized with a 30°, 5 mm-telescope. In cases with thoracic tube inserted, the tube was pulled out and the tube tract was used for the entrance of the first trocar. Once a vision was achieved by making some dissection with the camera in the pleural space, the second and,

if necessary, the third trocar (3 or 5 mm) was placed depending on the localization of the loculated fluid and septa. Septations in the pleural cavity were dissected under direct vision using a 3 or 5-mm endoscopic dissector, scissors and aspirator inserted through the trocars; purulent fluid was aspirated, and fibrin was debrided. Later, the pleural space was irrigated with isotonic fluid with the help of a sump drain inserted through one of the trocar holes. At the end of the procedure, an appropriately sized thorax tube was inserted through one of the trocar holes into the pleural space with the help of a camera.

Statistical Analysis

Statistical analysis was performed using SPSS 15.0 for Windows program. Chi-Square and Mann-Whitney tests were used to compare the groups. A p value of <0.05 was considered statistically significant.

RESULTS

Therapeutic intervention was performed in 53 cases for treatment of empyema. The mean age of the patients was 4.6 (1-17) years; the female/male ratio was 25/28. There were 25 cases in Group 1, and 28 cases in Group 2. Pleural effusion was located in right side in 29 cases and in left side in 23 cases. Bilateral pleural effusion was present in one case. Pre-intervention clinical and laboratory findings, and empyema stages of both groups were compared. The results were presented in **Table 1**. It was seen that the data obtained were similar in both groups and no statistically significant difference was found.

Post-intervention clinical and laboratory findings, tube thoracostomy and hospital stay durations of both groups were compared. The results were presented in **Table 2**. The mean CRP value measured on the first day after the intervention, the incidence of bronchopleural fistula development, the number of patients who received blood transfusion, the length of stay in the intensive care unit, the length of hospital stay after the intervention, and the total length of hospital stay were similar in both groups. Apart from these, other data were found to be significantly lower in Group 2. Improvement in clinical findings such as fever, respiratory distress, and leukocytosis was significantly faster in Group 2. Furthermore, duration of thoracic tube was significantly shorter in Group 2. Although the length of hospital stay was shorter in Group 2, it was not statistically significant.

In group 2, thoracoscopic debridement was performed first in 20 patients who had a loculated or thick pleural fluid on radiological examinations and/or had undergone a failed thoracentesis, without prior tube thoracostomy (in cases where fluid was observed radiologically but could not be removed by thoracentesis, the fluid was assumed to be loculated and fibrinous). Tube thoracostomy was performed in the other 8 cases first, and thoracoscopic debridement was performed when there was no clinical improvement. Thoracotomy was performed in 6 cases in Group 1 due to the lack of clinical and radiological improvement (due to persistence of clinical findings in 2 cases, and continuation of clinical findings and bronchopleural fistula in 4 cases). In Group 2, fistula repair was performed by thoracotomy upon the persistence of bronchopleural fistula despite clinical improvement in only 1 case. In addition, in 1 case in Group 2, the fistula was repaired by performing thoracoscopy for the second time as the bronchopleural fistula persisted. Clinical improvement was achieved in all other cases, and bronchopleural fistulas regressed spontaneously. All cases were discharged with healing, and no patient died.

Table 1. Pre-intervention clinical and laboratory findings, and empyema stages of both groups

	Group 1 (n=25)	Group 2 (n=28)	p
Duration of complaints (mean±SD) (day)	11.2±6.08	11.21±6.82	0.82
High fever at first admission (n)	21	19	0.21
Leukocyte count at first admission (mean±SD) (/mm^3)	21768.18±11376.51	16747.69±8245.62	0.11
CRP value at first admission (mean±SD) (mg/dl)	16.38±12.1	19.46±8.12	0.57
Pleural LDH (mean±SD) (U/L)	7462.62±10043.03	4810.84±9765.95	0.29
Pleural Protein (mean±SD) (g/L)	4.43±0.75	4.57±1.34	0.73
Serum LDH (mean±SD) (U/L)	831.33±814.58	879±495.94	0.30
Serum Protein (mean±SD) (g/L)	6.06±0.61	6.04±1.26	0.51
Stage 1 empyema (n)	0	0	-
Stage 2 empyema (n)	21	25	0.69
Stage 3 empyema (n)	4	3	0.69
Cavitation (n)	6	6	0.82
Number of antibiotics used per patient (mean±SD)	2.52±0.77	2.21±0.56	0.06
Time from the onset of the complaint to tube thoracostomy (Group 1) and thoracoscopy (Group 2) (mean±SD) (day)	12.80±5.91	14.86±7.95	0.43

Table 2. Post-intervention clinical and laboratory findings, tube thoracostomy and hospital stay durations of both groups

	Grup 1 (n=25)	Grup 2 (n=28)	p
The duration of fever reduction after tube thoracostomy (Group 1) and thoracoscopy (Group 2) (mean±SD) (day)	10.71±9.19	2.05±1.12	<0.0001
Respiratory distress and regression of oxygen demand after tube thoracostomy (Group 1) and thoracoscopy (Group 2) (mean±SD) (day)	4.29±3.09	1.33±0.51	0.01
Leukocyte count on the 1st day after tube thoracostomy (Group 1) and thoracoscopy (Group 2) (mean±SD) (/mm3)	17821.74±12456.5	10320.42±4736.51	0.04
CRP value on the 1st day after tube thoracostomy (Group 1) and thoracoscopy (Group 2) (mean±SD) (mg/dl)	8.75±6.54	7.52±4.9	0.84
Duration of purulent drainage from thoracic tube (mean±SD) (day)	9.36±9.42	2.32±1.09	<0.0001
The duration of thoracic tube withdrawal after tube thoracostomy (Group 1) and thoracoscopy (Group 2) (mean±SD) (day)	13.16±8.73	5.79±4.95	<0.0001
Total duration of thoracic tube (mean±SD) (day)	13.16±8.73	7.82±7.1	0.003
Bronchopleural fistula (n)	7	6	0.75
Number of patients applied blood transfusion (n)	6	11	0.23
Amount of blood given per kg of body weight (mean±SD) (ml)	29.24±12.99	14.1±4.51	<0.0001
Thoracotomy (n)	6	1	0.043
Length of stay in intensive care (mean±SD) (day)	5.72±4.33	4.82±4.27	0.22
Length of stay in hospital after tube thoracostomy (Group 1) and thoracoscopy (Group 2) (mean±SD) (day)	15.08±9.89	11.36±9.21	0.11
Duration between first admission to hospital and withdrawal of the thoracic tube (mean±SD) (day)	14.2±9	8.75±6.91	0.008
Total length of hospital stay (mean±SD) (day)	16.68±10.08	14.96±10.81	0.44

DISCUSSION

The primary objective in treatment of empyema is the treatment of infection with antibiotherapy and the evacuation of pleural fluid to ensure adequate lung re-expansion. Thoracentesis, tube thoracostomy, fibrinolytic therapy, thoracoscopic debridement, thoracotomy and decortication can be used for the treatment of empyema in children (9). However, there is no consensus on which treatment should be used and when. The stage of empyema, the condition of the involved lung, the presence of a bronchopleural fistula, and the clinical condition of the patient affect the treatment.

The conventional treatment of empyema involves drainage of the pleural space through tube thoracostomy. However, the success rate of tube thoracostomy alone is rather low in the presence of dense and loculated pleural fluid. Although there are cases where tube thoracostomy has been successful, long-term use of antibiotics, the need for repeated tube thoracostomy, and the need for a long hospital stay constitute significant disadvantages (10,11). After tube thoracostomy, a second tube may be required in 15-40% of the cases, and conversion to thoracotomy for open decortication or lobectomy may be needed (2,12). The most important reason for the failure of the treatment performed with tube thoracostomy is the presence of fluid that is too thick to drain from the tube and septations that allow only some of the fibrin-containing fluid. Thus, tube thoracostomy becomes ineffective, duration of tube and time for clinical recovery are prolonged. For such patients, thoracoscopic debridement is recommended as a treatment option to eliminate loculations in the pleural space and to remove fibrin and pus that are too thick to be drained from the thoracic tube (2,10-14). By dissecting loculations with thoracoscopic debridement, gelatinous, organized pleural material and empyema fluid are evacuated, allowing the lung to re-expand.

Thoracoscopy was performed in 9 cases by Kern and Rogders for the first time in the treatment of empyema in children in 1993 and they reported that a rapid recovery was achieved (15). This procedure has been widely used in children since then (2,7,8,11,13,14). Thoracoscopic debridement allows reconstruction of a single pleural cavity by debriding intrapleural loculations and membranous structures. Furthermore, the thorax tube is placed more easily and conveniently under the camera view. This enables a more successful drainage. Thoracoscopic debridement has a higher chance of success than tube thoracostomy alone and shortens the duration of treatment (6-8). Thoracoscopic debridement reduces the length of hospital stay, facilitates return to normal activity, reduces the need for repeated thoracentesis and tube thoracostomy, as well as pain and anxiety in the child; It is less invasive compared with thoracotomy, reduces the need for blood transfusion and analgesia, and provides a better cosmetic appearance (2,11,12,14)

Although fibrinolytic therapy has recently become popular in empyema, studies have shown that duration of tube thoracostomy and hospital stay times are shorter in thoracoscopic debridement compared to fibrinolytic therapy (16,17). In tube thoracostomy or

in intrapleural fibrinolytic therapy along with tube thoracostomy, the dissection of septations and cleaning of the pleura can take days, while thoracoscopy can be performed more effectively in a few hours. Therefore, the duration of clinical recovery and tube thoracostomy is shortened. Studies have shown that the success rates of thoracoscopy and fibrinolytic therapy in empyema are similar, and they do not have any superiority over each other (18-20). There was no difference between the two methods in terms of length of hospital stay time, duration of tube drainage, duration of fever, and need for analgesics and oxygen (18). Generally, the criticism on this subject argued that thoracoscopy is a high-cost operation performed under general anesthesia. However, in practice, it should be accepted that tube thoracostomies are also performed under general anesthesia in the operating room and perioperative complications may occur. (17). In addition, it should be kept in mind that operation costs are lower and fibrinolytic therapy costs are higher in developing countries compared to developed countries. In cases where fibrinolytic therapy is unsuccessful, the cost and length of hospital stay increase with thoracoscopy. In a meta-analysis, it was found that thoracoscopy and fibrinolytic therapy in empyema had similar rates in terms of complications, and the number of repeated attempts in thoracoscopy and the length of postoperative hospital stay were shorter (17). Thoracoscopic debridement is recommended as the first-line treatment method, especially in advanced stage empyema (21).

Pre-intervention clinical and laboratory findings, and empyema stages of both groups were similar in both groups and no statistically significant difference was found. This indicates that both groups had similar homogenous characteristics and severity of empyema in both groups was similar. In terms of staging, no patients with Stage 1 empyema were found in either groups, while the number of patients with Stage 2 and Stage 3 empyema was similar. Thus, both groups could be compared in terms of the effectiveness of the treatment method applied. Incidence of cavitation, which is an indicator of parenchymal necrosis, was similar in both groups. Both groups were similar in terms of this finding, which indicates the severity of the underlying pneumonia.

Clinical recovery is faster in cases who underwent thoracoscopic debridement compared to tube thoracostomy, and patients return to their normal lives sooner (6,15,22). In our study, post-procedure fever, leukocytosis, respiratory distress and duration of regression in oxygen need were shorter in the thoracoscopy group; that is, clinical improvement was faster in this group. After the procedure (tube thoracostomy in the first group, and after thoracoscopy in the second group) leukocyte values were also lower in the second group. With thoracoscopic debridement, purulent and infected material was removed from the body more effectively and rapidly, which reflected to the clinical course and fever, leukocytosis and respiratory distress were reduced more rapidly.

By dissecting the loculations with thoracoscopic debridement, gelatinous, organized pleural material and empyema fluid are evacuated, allowing the lung to re-expand (23). Thus, purulent drainage takes a short time. In our study, duration of the purulent fluid drainage time after the procedure (tube thoracostomy in the first group, and after thoracoscopy in the second group) was shorter in the second group. Fibrinoid structures and septations were removed more effectively with thoracoscopy. This indicates that thoracoscopic debridement is more effective in the drainage of empyema and achieves drainage in a short time. In the treatment of empyema, thoracoscopy is a safe and effective method that prevents the chest tube from staying for a long time (24). In our cases, duration of the tube thoracostomy after thoracoscopic debridement was shorter compared to the cases in the first group. Thoracoscopic debridement accelerated the drainage of empyema and tube thoracostomies were terminated in a shorter time.

Parenchymal necrosis leads to bronchopleural fistula, resulting in a prolonged stay of thoracic tube in the chest, it also causes the signs of infection to persists (11,13,14). In our study, no significant difference was found in both groups in terms of bronchopleural fistula development. Bronchopleural fistula developed in all cases with cavitation, which is an indicator of parenchymal necrosis. The development of a bronchopleural fistula is related to the severity of the underlying parenchymal disease, regardless of the procedure. This indicates that the disease in both groups had similar severity and that the groups were comparable to each other.

After tube thoracostomy, a second tube may be required in 15-40% of the cases, and thoracotomy and lobectomy for decortication may be needed (2,12). However, problems such as disruption of the integrity of the thorax due to a large incision, pain and prolonged recovery in the postoperative period, and unwanted cosmetic appearance may occur with thoracotomy (2). In our study, thoracotomy was performed in 6 patients who did not show clinical improvement in the first group, whereas in the second group only 1 patient required thoracotomy due to the persistence of bronchopleural fistula despite clinical improvement. The need for thoracotomy was significantly less common in the second group. Thoracoscopic debridement reduced the need for thoracotomy. This indicates that thoracoscopic debridement is effective and sufficient in the treatment of empyema.

Thoracoscopic debridement reduces the need for blood transfusion compared to thoracotomy (14). While there was no difference between the number of cases requiring blood transfusion in both groups, the amount of blood transfused per kg of body weight was significantly higher in the first group. This indicates that there was a higher blood need for the cases in the first group. In the first group, the prolongation of the clinical improvement and therefore the persistence of the infection and the greater need for thoracotomy increased the need for blood transfusion.

When compared to other treatment methods, thoracoscopic debridement shortens the length of hospital stay (2,12,15-17). In both groups, there was no significant difference in terms of the time interval from the procedures to discharge, the length of stay in the intensive care unit and the length of hospital stay. In the second group, when the patients who underwent tube thoracostomy before thoracoscopic debridement are removed, the length of stay in the intensive care unit is significantly shortened. In the second group, patients who underwent tube thoracostomy before thoracoscopic debridement and were monitored in the intensive care unit caused prolonged length of stay in the intensive care unit. When the time interval from hospitalization to the removal of the thoracic tubes was considered as the surgical recovery time, it was found to be shorter in the thoracoscopy group. Surgical recovery of empyema was faster in patients who underwent thoracoscopic debridement with or without tube thoracostomy. Purulent drainage regressed in a shorter time in the second group owing to thoracoscopy.

CONCLUSION

In our study, clinical recovery and tube thoracostomy was shorter in the thoracoscopic debridement group compared to the other group, and the need for thoracotomy was lower. Debridement of fibrin and septations in the pleural space more effectively and rapidly under the camera view helps to achieve this. Thoracoscopic debridement contrubutes recovery from empyema in children. Tube thoracostomy alone causes both slow clinical recovery and progression of the disease and consequently loss of time. Therefore, we think that thoracoscopic debridement should be the first choice in the treatment of empyema and no time should be lost with tube thoracostomy beforehand.

Limitations

This study performed retrospectively and number of patients is small.

ETHICAL DECLARATIONS

Ethics Committee Approval: In this research, the data before 2020 was used and the research was concluded before 2020. According to the Regulation on Clinical Researches published in the Official Gazette of the Republic of Turkey with the number 28617 dated 3 November 2015, the ethics committee approval was not obtained in accordance with the article "Retrospective studies are outside the scope of the regulation (article 2-(2))". This study was prepared in accordance with the Law on Protection of Personal Data, by anonymizing patient data and in accordance with the 2013 Brazil revision of the Helsinki Declaration and guidelines for Good Clinical Practice.

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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ORIGINAL ARTICLE
ORİJİNAL ARAŞTIRMA

Evaluation of Children with Extremity Fracture Occurred as a Result of Motor Vehicle Injury

Motorlu Taşıt Yaralanması Nedeniyle Ekstremite Kırığı Gelişen Çocukların Değerlendirilmesi

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ABSTRACT

Aim: The aim of the study was to examine and evaluate patients with extremity fractures who admitted to the Pediatric emergency service due to a motor vehicle accident, whether there was any accompanying additional organ injury, and fracture patterns according to the severity of the trauma.

Material and Method: The study was conducted between January 2015 and 2020 retrospectively. Relevant data were analyzed with IBM SPSS V23 statistics.

Results: Of the 205.710 patients who admitted to Pediatric emergency service in the course of this study, 1.378 (0.66%) experienced motor vehicle injuries. 161 (0.08%) of these cases were evaluated on the grounds of extremity fractures. Mean age of the cases were 133 months (minimum: 3, maximum: 2018 months) and 90 (56%) cases were male. Trauma type was mostly outside-vehicle traffic accident (50%). 55% of the cases were severe mechanism of injury. The most common lower extremity fracture type was the tibia (26%) fracture. The most common fractures in the upper extremity were humerus (16%) and radius (16%) fractures. Of the patients with pelvis fracture, 65% had other types of organ injuries ($p<0.05$). Surgical treatment was admitted to 68% of those with femoral fractures and 43% of those with tibia fractures ($p <0.05$). While lower extremity fractures were more common in outside-vehicle traffic accident (66%) and motorcycle accident (60%), upper extremity fracture was more common in intra-vehicle traffic accident (54%) ($p <0.05$).

Conclusion: Since the literature evaluating the extremity fractures in motor vehicle accidents is very limited, relevant data are also very limited. Extremity fractures that occur especially after motor vehicles are an important cause of injuries and deaths, and impose a heavy burden upon both families and the government in terms of hospital stay and hospital costs.

Keywords: Pediatric emergency, motor vehicle injury, extremity fractures

ÖZ

Amaç: Çocuk Acil Servise motorlu araç kazası nedeniyle başvuran ekstremite kırığı mevcut olan hastaları, beraberinde ek organ yaralanması olup olmadığını, trvamanın şiddetine göre kırık paternlerini inceleyip değerlendirmeyi amaçladık.

Gereç ve Yöntem: Ocak 2015 ve Ocak 2020 tarihleri arasında motorlu araç kazasıyla başvuran ekstremite kırığı olan hastalarda retrospektif olarak yapılmıştır. Veriler IBM SPSS V23 ile analiz edildi.

Bulgular: Çalışma süresince Çocuk Acil Servise 205.710 hasta başvurmuş olup, 1.378 (% 0,66)'i motorlu taşıt yaralanmasıdır. Bu olgularında 161 (%0,08)'i ekstremite kırığı nedeniyle değerlendirilmiştir. Olguların yaş ortalaması 133 ay (minimum:3; maksimum:218ay) dır ve 90 (%56) olgu erkektir. Travma şekli daha çok ADTK (%50) idi. %55 i yüksek enerjili travmaydı. Alt extremite kırıklarından en sık görüleni tibia (%26) kırığıydı. Üst extremitede kırıklarından en sık görüleni humerus (%16) ve radius (%16) kırıklarıydı. Pelvis krığı olan hastaların %65 inde başka organ yaralanması vardı ($p<0,05$). Hastaların büyük kısmına (%66) tedavide atel uygulanırken, %26 sına cerrahi operasyon yapılmıştır. Femur kırığı olanların %68 ine, tibia kırığı olanların %43 üne cerrahi tedavi uygulandı ($p<0,05$). Hastaların %50,3 ü acilden taburcu edildi. Alt extremite kırığı ADTK (%66) ve motosiklet kazası (%60) sonucu daha çok görülürken, Üst extremite kırığı AİTK (%54) sonucu daha çok görüldü ($p<0,05$).

Sonuç: Motorlu taşıt kazalarında extremite kırıklarının incelendiği literatür çok kısıtlı olduğundan, verilerde çok sınırlıdır. Özellikle motorlu araç sonrası gelişen extremite kırıkları sakatlıklar ve ölümlerin önemli bir nedeni ve hastanede kalış ve hastane maliyeti açısından da hem ailelere hem de devlete ağır bir yük oluşturur.

Anahtar kelimeler: Çocuk acil, motorlu araç yaralanması, ekstremite kırıkları

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INTRODUCTION

Motor vehicle injuries are caused by motor-driven motorcycles, automobiles, trucks, minibuses, trains, trams, tractors, and land vehicles. Motor vehicle injuries play critical role in casualties and labor loss in developed and developing countries (1). Across the globe, due to trauma caused by motor vehicle injuries, more than 3.000 people die every day, and about 1.2 million people each year (2,3).

Of the emergency service applications in developing countries, 30-86% are due to trauma caused by motor vehicle injuries. Current data on injuries indicate that driving motor vehicle without helmet and seat belt accounts for more than 50% of driver casualties (4).

Serious injuries caused by motor vehicle accident are traumatic brain injury, spinal cord injury, generalized burns in the body, amputation and blindness (5). Fractures occur at a ratio of 10-25% among all childhood traumas. Occurrence frequency is 50% in boys and 30% in girls. Fracture pattern has a changeability depending on the countries, climatic characteristics and cultural features (6). Extremity fractures constitute 80-90% of all fractures. The type and pattern of extremity fractures also vary as per the age, mechanism of injury, and involvement of surrounding tissues (7). Bone periosteum of children are thicker, stronger, has a significant osteogenic potential, and are more active metabolically. In regard of these characteristics, children fractures are easier to heal. Thicker periosteum prevents fracture displacement and compound fractures (8).

Extremity fractures caused by motor vehicle accidents are essential as they can lead to morbidity and disabilities. Lower extremity fractures, upon which limited number of studies were conducted in literature, were analyzed in one of the studies evaluating few adult and pediatric patients. Of the samples, most of them were adults between the ages of 20-40, stressing the fact that this is due to population being consisted of actively working adults who have a higher potential to use motor vehicles.

In this study, we conducted a retrospective analysis regarding the patients with extremity fractures who admitted to the Pediatric emergency service due to a motor vehicle accident, whether there was any accompanying additional organ injury, and fracture patterns according to the severity of the injury, and aimed to emphasize the importance of protective and preventive measures and equipment and traffic rules to prevent mortality and disabilities.

MATERIAL AND METHOD

Study Design

This study was conducted retrospectively between January 2015 and 2020 at urban, tertiary pediatric emergency service. Ethics committee approval was received by the medical faculty scientific research ethics committee prior to the study (No: 09.05.2020/ 91610558-604.01.02-).

Patient Selection and Data Collection

All patients younger than 18 years old with a fracture of an extremity due to a motor vehicle accident who admitted directly or brought by ambulance to the pediatric emergency service were included in the study. Demographic data of the patients such as age, gender, type of trauma, fracture extremity, fracture type, number of fractures, treatment and follow-up were recorded.

Definitions

Motor vehicle injuries are injuries caused by motor-driven motorcycles, automobiles, trucks, minibuses, trains, trams, tractors, and land vehicles (9).

Severe mechanism of injury is defined as overturning as a result of a motor vehicle accident, ejection from the vehicle, having a casualty inside the vehicle, collapse failure of more than 50 cm on the driver's side, collapse failure of more than 30 cm on the passenger side, being stuck the vehicle for more than 20 minutes, motorcycle accident without helmet, running down a pedestrian with a vehicle or a motorcycle, and speeding up to 65 km/h (10).

Multiple fractures are evaluated as the presence of more than one long bone fracture in the patient.

Statistical Analysis

Data were analyzed with IBM SPSS V23 statistics. Chi-square and One-way Anova tests were used in the comparison of categorical data. T test was used in the comparison of ages of the patients. Analysis results were presented as frequency (percentage) for categorical data, mean and standard deviation for numerical data. Significance level was taken as $p<0.05$

RESULTS

Of the 205.710 patients who admitted to Pediatric emergency service in the course of this study, 1.378 (0.66%) were motor vehicle injuries. 161 (0.08%) of these cases were evaluated on the grounds of extremity fractures (**Figure 1**). Mean age of the cases were 133 months (minimum: 3, maximum: 2018 months) and 90 (56%) cases were male. Demographic information of the patients who constitute the study group were presented in **Table 1**.

Of the patients, 52% (80) had low extremity fracture localization. The most common lower extremity fracture type was the tibia (26%) fracture, followed by femoral (n=28, 17%), fibula (n=28, 17%), and pelvis fractures (n=20, 12%) respectively. The most common upper

Table 1: Demographic data of the patients		
Age (months) min-max	130±59.6 (3-218)	
Gender	**n (%)**	
Male	90 (56)	
Female	71 (44)	
Application time		
08:00-16:00	63 (39)	
16:00-00:00	79 (49)	
00:01-07:59	15 (9)	
Trauma types		
Intra-vehicle traffic accident	71 (44)	
Outside-vehicle traffic accident	80 (50)	
Motorcycle	10 (6)	
Severe mechanism of injury		
Yes	89 (55)	
No	72 (45)	
Type of Severe mechanism of injury		
Overturning as a Result of a Motor Vehicle Accident	21 (13)	
Ejection from the vehicle	7 (4)	
Casualty status	12 (8)	
Collapse failure of more than 50 cm	3 (2)	
Collapse failure of more than 30 cm on the passenger side	2 (1)	
Being stuck in the vehicle for more than 20 minutes	4 (3)	
Motorcycle Accident without Helmet	4 (3)	
Running Down a Pedestrian With a Vehicle or a Motorcycle	15 (9)	
Speeding up to 65 km/h	21 (13)	
Fracture Localization		
Upper	67 (42)	
Lower	84 (52)	
Upper and lower extremity	10 (6)	
Fracture Type		
Compound Fracture	6 (4)	
Closed Fracture	153 (95)	
Compound+closed fracture	2 (1)	
Checkup		
Discharged from emergency department		
Yes	81 (50.3)	
No	80 (49.7)	
Follow-up in other units		
Pediatrics	4 (3)	
Pediatric intensive care	13 (8)	
Orthopedics	34 (21)	
Pediatric surgery	17 (11)	
Treatment	**Yes n (%)**	**No n (%)**
Air Splint	106 (66)	55 (34)
Plaster	22 (14)	139 (86)
Surgery	42 (26)	119 (74)
Other organ injury	**Yes n (%)**	**No n (%)**
Liver	14 (9)	147 (91)
Spleen	4 (2.5)	157 (97.5)
Lungs	23 (14)	138 (86)
Intestines	1 (1)	160 (99)
Stomach	0 (0)	161 (100)
Pancreas	7 (4)	154 (96)
Kidney	1 (1)	160 (99)
Central Nervous System	4 (2.5)	157 (97.5)

extremity fracture type was humerus (n=26, 16%) and radius (n=26, 16%) fractures, followed by clavicula (n=23, 14%) and ulna (n=8, 5%) fractures respectively. While shaft fractures were more common in femoral fracture type, distal fractures were more common in the tibia and fibula fracture types, and proximal fractures were more common in the humerus fracture types (**Figure 2, 3**).

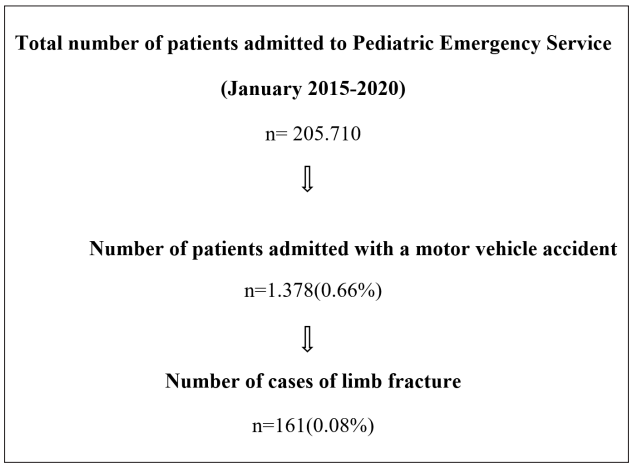

Figure 1. Patients Admitted to Pediatric Emergency Service

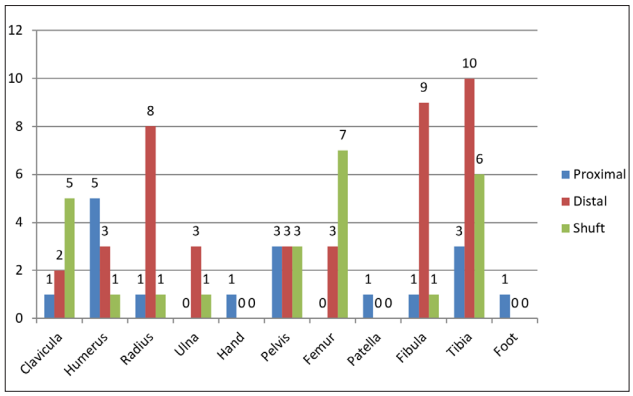

Figure 2. Extremity bone fracture incidence ratio for the right side (%)

*Total ratio for Patella, Hand, and Foot were indicated in the proximal part, without distinction of proximal, distal, shaft fractures.

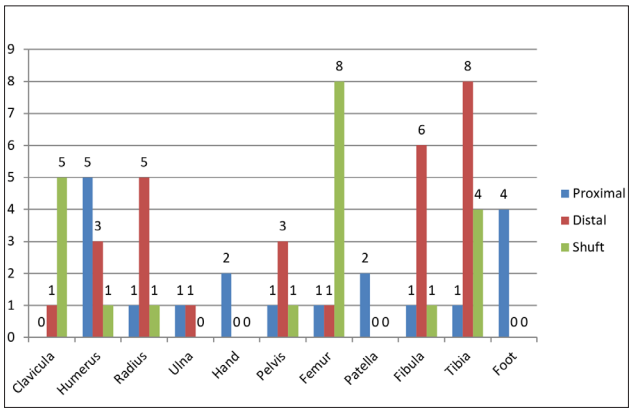

Figure 3. Extremity bone fracture incidence ratio for the left side (%)

*Total ratio for Patella, Hand, and Foot were indicated in the proximal part, without distinction of proximal, distal, shaft fractures.

Evaluating the fracture localization as per the age groups, it was observed that the upper extremity fractures in 13 (50%) patients under the age of 5 were more common while lower extremity fractures in 29 (54.7%) patients aged 5-11 and 44 (53.4%) patients over the age of 11 were more common. Of the patients with lower extremity fractures, 44 (52.4%) consisted of children over the age of 11 (**Table 2**). The ratio of all the fracture types were more common in children over the age of 11 (**Figure 4**).

Table 2. Fracture localization as per the age groups

Age Groups	Fracture localization			p
	Upper Extremity n (%)	Lower Extremity n (%)	Both n (%)	
5 years >				0.109
yes	13(19.4)	11(13.1)	2(20)	
no	54(80.6)	73(86.9)	8(80)	
5-11 y				0.080
yes	20(29.9)	29(34.5)	4(40)	
no	47(69.1)	55(65.5)	6(60)	
11y<				0.076
yes	34(50.7)	44(52.4)	4(40)	
no	33(49.3)	40(47.6)	6(60)	

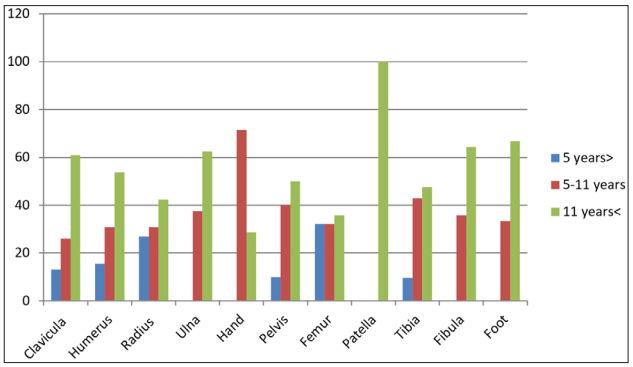

Figure 4. Extremity bone fractures by age groups (%)

Femoral, pelvis, fibular fractures and other organ injuries were statistically significantly high in the patients who admitted hospitals due to severe mechanism of injury ($p<0.05$). Additionally, 83% of the compound fractures occurred due to severe mechanism of injury (**Table 3**).

While upper extremity fractures were statistically significantly high in severe mechanism of injury patient group who experienced overturning as a result of a motor vehicle accident, ejection from the vehicle, and those who had a casualty inside the vehicle; lower extremity fractures were more common in the patient groups who experienced motorcycle accident without helmet, pedestrian versus motor vehicle accident and those speeding up to 65 km/h ($p<0,05$). While most of the patients who had upper extremity fractures were treated with air splint or encased in plaster, lower extremity fracture cases required surgical treatment (**Table 4**).

Of the patients with femoral fracture, 68% were surgically treated ($p<0.05$). The ratio of other organ injuries occurred in the patients with pelvis fracture were 65 % ($p<0.05$) (**Table 5**).

Table 3. The relationship between severe mechanism of injury and fracture location, fracture type, and other organ injury

	Severe mechanism of injury		p
	Yes n (%)	No n (%)	
Femoral fracture			0.006
Yes	22(25)	6 (8)	
No	67(75)	66 (92)	
Pelvis fracture			0.001
Yes	18(20)	2 (3)	
No	71(80)	70 (97)	
Other organ injury			<0.001
Yes	42(47)	9 (13)	
No	47(53)	63 (87)	
Tibia fracture			0.060
Yes	18(20)	24(33)	
No	71(80)	48(67)	
Fibula fracture			0.007
Yes	9(10)	19(26)	
No	80(90)	53(74)	
Humerus fracture			0.118
Yes	18(20)	8(11)	
No	71(80)	64(89)	
Radius fracture			0.483
Yes	16(18)	10(14)	
No	73 (82)	62(86)	
Ulna fracture			0.299
Yes	3(3)	5(7)	
No	86(97)	67(93)	
Multiple fracture			0.332
Yes	35(39)	23(32)	
No	54(61)	49(68)	
Compound Fracture	5(6)	1(1)	
Closed Fracture	82(92)	71(99)	0.574
Compound+closed fracture	2 (2)	0(0)	
Total	89	72	

Table 4. Comparison of fracture location and severe mechanism of injury type and treatment modality

Type of severe mechanism of injury	Fracture localization Upper ext.	Lower ext.	Upper+lower ext.	p
Overturning as a result of a motor vehicle accident	11(52)	9(43)	1(5)	
Ejection from the vehicle,	4(57)	3(43)	0(0)	
Casualty status	7(58)	3(25)	2(17)	
Collapse failure of more than 50 cm	1(33)	1(33)	1(33)	
Collapse failure of more than 30 cm on the passenger side	1(50)	0	1(50)	0.028
Being stuck in the vehicle for more than 20 minutes	0	2(50)	2(50)	
Motorcycle accident without helmet	1(25)	3(75)	0	
Running down a pedestrian with a vehicle or a motorcycle	6(40)	8(53)	1(7)	
Speeding up to 65 km/h	3(14)	17(81)	1(5)	
Treatment modality				
Air Splint				
Yes	51(76)	47(56)	8(80)	
No	16(24)	37(44)	2(20)	
Plaster				
Yes	6(9)	14(17) 70(83)	2(20)	
No	61(91)		8(80)	
Surgical Treatment		29(35)		
Yes	6(9)	55(65)	7(70)	
No	61(91)		3(30)	

Table 5. Correlation of fracture location and surgical treatment.

Fracture localization	Surgical Treatment		p
	Yes n (%)	No n (%)	
Femoral fracture			<0.001
Yes	19(68)	9(32)	
No	23(17)	110(83)	
Tibia fracture			0.004
Yes	18(43)	24(57)	
No	24(20)	95(80)	
Pelvis fracture			0.670
Yes	4(20)	16(80)	
No	3(2)	138(98)	

DISCUSSION

Motor vehicle accidents have an important place among childhood traumas and they are the leading cause of death in children aged 2-14 years in developed countries (11). Motor vehicle accidents consist of 30% of the trauma-related casualties aged between 0-19 (12). Fractures are common among injuries in the childhood age group (13). Extremity fractures are among the most common reasons for hospitalization in children (14). While there were not many articles previously studying extremity fractures occur in motor vehicle accidents, in the present study, male dominance (56%) was observed in line with the results of studies evaluating childhood fractures (15,6).

Lower extremity fractures constitute approximately 20% of all fractures occur in children and may cause significant mortality and morbidity (16). In the study of Ngunde et al., the incidence ratio of lower extremity fractures in patients experienced a motor vehicle accident was found to be 35% (4). In a study conducted with adult patients, the lower extremity fracture was detected 3 times more than the upper extremity fracture occurred in motor vehicle accident (3). Of the patients included in this study, 52% had lower extremity fracture. Evaluating the age groups, lower extremity fracture was mostly seen in the patient group over 11 years of age. We can conclude that in conjunction with the growth, children will be as tall as the vehicles and along with it, lower extremity risk also increases.

Lower extremity fractures were also significantly higher in patients who admitted to hospital for severe mechanism of injury (particularly a motorcycle accident without a helmet, running off a pedestrian, speeding up to 65 km / h). In line with the literature, mostly tibia fracture (26%) was observed in the lower extremity (15,17).

In accordance with the literature (18), shaft fractures were more common in the femur, while distal fractures were prominent in the tibia and fibula. In addition, femoral fractures caused by a motor vehicle accident were mostly treated surgically in this study, as was detected in other studies conducted (19).

43% of tibia fractures required surgical operation. For that reason, it is of great importance that the parents adhere to the traffic rules, display sensitivity as regards the safety equipment such as safety belt, helmet, and bring them to bear on their children without compromising.

Pediatric pelvic fractures occur in 0.2% to 2% of all pediatric fractures and usually occur due to severe mechanism of injury (20). Of the patients included in

this study, 12% had pelvis fracture, and pelvis fractures occurred in 90% of the patients as a result of severe mechanism of injury in accordance with the literature (21,22). Pediatric pelvis fractures usually heal without needing a surgical operation (23,24). Of the patients included in a study evaluating pelvis fractures, 94% healed without needing a surgical operation. In this study, in line with the literature, 80% of the patients were treated without surgical operation (25). Although it is a common fact that pelvis fractures heals itself without needing a surgical operation, it should not be forgotten that bleeding risk is considerably high and usually convoys other organ injuries (26). In the present study, in line with the literature (27), the ratio of other organ injuries was found 65% while multiple fracture ratio was found 25%.

In other studies evaluating childhood extremity fractures, the upper extremity fracture was more prominent than the lower extremity fracture. It can be concluded that this is because not only motor vehicle accidents but all traumas were evaluated in the study (16). In this study, it was found that most common upper extremity fractures (with a ratio of 42%) were humerus (26%) and radius (26%) fractures, followed by clavicula (14%) and ulna (5%) fractures respectively. In another study evaluating other motor vehicle accidents6, clavicula was more common, while in another study evaluating upper extremity fractures (3), it was found that radius (27%) was the most common fracture type, followed by humerus (25%), ulna (18%), and clavicula (17%) fractures respectively. In a study evaluating upper extremity fractures with adult patient groups (28), humerus (44%) and ulna (28%) fractures were the most common fracture types.

Multiple fractures are mostly associated with severe mechanism of injury, and their incidence ratio varies between 1.7 to 23% in the pediatric age group.3 In this study, approximately 1/3 of the patients had multiple fractions and of this cases, 60% had severe mechanism of injury. Patients with upper extremity fractures were mostly treated with plaster and air splint, while patients with lower extremity and multiple fractures were treated mostly with surgical operations.

Compound fractures have considerably high morbidity and mortality possibilities requiring patient to be hospitalized longer along with the high hospitalization costs. Of the compound fractures, 90% occur as a result of traffic accident. Risk of contamination, infection, nonunion of the fracture, risk of delayed union, neurovascular complications and increased amputation rate increase the significance of the compound fractures for the childhood age group (29). In this study, the ratio of compound fractures was found to be 4%. Of these fracture incidences, 83% occurred as a result of severe mechanism of injury.

The most substantial limitation is that the study was conducted in a single center. Multi-centered studies are required.

CONCLUSION

Since the literature analyzing extremity fractures in motor vehicle accidents is very limited, relevant data are also very limited in this regard. In particular, extremity fractures, injuries and additional organ injuries that develop after motor vehicles create a heavy burden on both families and the government in terms of hospitalization and hospital costs, resulted by a critical ratio of casualties. On that account, collecting relevant data of this group of patients is of very high concern in terms of the management of this group of patients, taking precautions to prevent traffic accidents and narrating traffic rules to parents and children via their parents.

ETHICAL DECLARATIONS

Ethics Committee Approval: Ethics committee approval was received by the medical faculty of Gazi University scientific research ethics committee prior to the study (No: 09.05.2020/ 91610558-604.01.02-).

Informed Consent: Informed consent forms were obtained from the parents of the patients and controls included 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 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

Treatment Approaches in Ovarian Masses in Children

Çocuklarda Over Kitlelerinde Tedavi Yaklaşımları

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Abstract

Aim: The aim of this retrospective study is to evaluate clinical features, treatment approaches and outcomes of children with ovarian mass.

Material and Method: In our clinic, the oncologic charts of children with a mass in the ovary between 2009 and 2020 were analyzed retrospectively. The patients' demographic features, symptoms and signs, diagnosis, treatments and outcomes were recorded.

Results: The age of 55 patients included in the study ranged from two months to 18 years (median, 12.9 years). While the ages of 38 patients were ≥ 10 years (69%), 17 of them were <10 years (31%). Fifty-one of the patients underwent elective surgery (92.7%), and four had emergency surgery (7.3%). Of the surgeries performed, 28 were salpingo-oophorectomy (51.9%), 23 were oophorectomy (42.6%), and 4 were cystectomy (5.6%). The types of ovarian mass were germ cell tumors (n: 44, 80%), epithelial tumors (n: 4, 7.3%), stromal tumors (n: 3, 5.5%), simple cyst (n: 2, 3.6%), and others (n: 2, 3.6%). Two patients with malignant tumor (one dysgerminoma patient with ataxia telangiectasia, and the other with yolk sac tumor) died while the others were alive.

Conclusion: In children, benign tumors are more prominent and surgical treatment is sufficient. In malignant tumors, overall survival rates have increased with multidisciplinary approaches.

Keywords: Ovary, mass, children, treatment approaches

Öz

Amaç: Bu geriye dönük çalışmanın amacı, over kitlesi olan çocukların klinik özelliklerini, tedavi yaklaşımlarını ve sonuçlarını değerlendirmektir.

Gereç ve Yöntem: Kliniğimizde, 2009 ile 2020 yılları arasında, overde kitle saptanan çocukların dosyaları geriye yönelik incelendi. Hastaların demografik özellikleri, semptom ve bulguları, tanıları, tedavileri ve sonuçları kaydedildi.

Bulgular: Çalışmaya dâhil edilen 55 hastanın yaşı iki ay ile 18 yıl arasında değişiyordu (ortanca, 12,9 yıl). Hastaların 38'inin yaşı ≥ 10 yıl iken (%69), 17'sinin yaşı <10 yıldı (%31). Hastaların 51'inde elektif cerrahi (%92,7), ve dördünde acil cerrahi uygulandı (%7,3). Gerçekleştirilen ameliyatların 28'i salpingo-ooferektomi (% 51,9), 23'ü ooferektomi (% 42,6) ve 3'ü kistektomi (% 5,6) idi. Overdeki kitlelerin tipleri, germ hücreli tümörler (n: 44,% 80), epitel tümörleri (n: 4,% 7,3), stromal tümörler (n: 3,% 5,5), basit kist (n: 2, %3,6), ve diğerleri (n: 2, %3,6) idi. Malign tümörü olan iki hasta (biri ataksi telenjiektazili disgerminoma hastası ve diğeri yolk sac tümörlü) kaybedilirken, diğer hastalar yaşamaktaydı.

Sonuç: Çocuklarda, benign tümörler daha ön plandadır ve cerrahi tedavi yeterlidir. Malign tümörlerde ise multidisipliner yaklaşımlarla genel sağ kalım oranları artmıştır.

Anahtar Kelimeler: Over, kitle, çocuk, tedavi yaklaşımı

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INTRODUCTION

In children, unlike adults, there is not much experience with adnexal masses and generally the limited experience in children is mostly on ovarian tumors (1, 2). The majority of the ovarian masses are benign in children (1-4). Although the most common complaints in ovarian masses are abdominal pain and palpable mass in children, urinary system symptoms such as dysuria or pollakiuria, or gastrointestinal system symptoms such as constipation, or menstrual disturbances such as dysmenorrhea, and menorrhagia or oligomenorrhea can also be presented. It should be kept in mind that precocious puberty, gonadotropin-independent, may be a sign of an ovarian mass, especially in juvenile granulosa cell tumors (1-5). Another interesting presenting complaint may be galactorrhea (6). The ovarian torsions that appear together with ovarian mass that require urgent surgical approach should not be forgotten (7).

Tumor markers such as human chorionic gonadotropin (HCG), alpha-fetoprotein (AFP), lactate dehydrogenase (LDH), and inhibin have the clinical utility in childhood germ cell tumors (8). Especially, elevations of AFP in yolk sac tumor, LDH in dysgerminoma, and HCG in choriocarcinoma are very useful in diagnosis, evaluation of treatment response and in follow-up.

Malignant ovarian tumors in children constitute 25% to 35% of all ovarian masses (2, 9). The functional lesions including corpus luteum cyst, hemorrhagic cyst, follicular cyst, paraovarian cyst, and only ovary torsion accounts for about half of all childhood ovarian masses (5, 10). Others are epithelial tumors such as serous cyst adenom, mucinous cyst adenom, mucinous cyst, serous cyst, and germ cell tumor such as mature cystic teratoma, mature teratoma, dysgerminoma, yolk sac tumor, and immature teratoma (5, 10, 11). Different childhood malignant diseases including neuroblastoma, rhabdomyosarcoma, Ewing's sarcoma, rhabdoid tumor, and carcinoid tumor may cause metastatic ovarian tumor in children also (12).

The aim of this study is to evaluate clinical features, treatment approaches and outcomes of children with ovarian mass, retrospectively.

MATERIAL AND METHOD

Approval was obtained from the local ethics committee for this retrospective study (No: 2021/129, Date: 27 Jan, 2021). Written informed consent was not obtained as a retrospective study. This study complies with the guidelines for human studies and was conducted ethically in accordance with the World Medical Association Declaration of Helsinki.

From 2009 to 2020, the oncologic charts of patients who were diagnosed and followed up with an ovarian mass at the department of pediatric hematology and oncology were retrospectively reviewed. The patients with functional lesions including corpus luteum cyst, hemorrhagic cyst, follicular cyst, paraovarian cyst, and only torsion were excluded.

The ages of the patients were recorded as demographic data. The patients were divided into two groups as age <10 years and ≥ 10 years. In addition, the symptoms and findings of the patients at admission, tumor markers at admission, if any; the largest diameter of the mass; surgical findings; diagnosis; treatment approaches and results were recorded.

The main diagnoses of the patients were given as simple cyst, germ-cell tumors, stromal tumors, and epithelial tumors, as definitive diagnosis.

"Federation Internationale de Gynecologie Oncologique" classification system was used for staging except non-Hodgkin lymphoma. For non-Hodgkin lymphoma, Murphy staging system was used.

The BEP chemotherapy protocol containing cisplatin, etoposide and bleomycin was used in patients with germ cell tumors, and the LMB89 protocol was used for non-Hodgkin lymphomas (13).

Statistical analysis

SPSS-15 software (SPSS Inc., Chicago, Illinois, USA) was used for all statistical analyses. According to whether the distribution was normal or not, the mean value and standard deviation or median value plus the minimum and maximum values were used in numerical data. For categorical data, frequency and percentage values were used. Kaplan–Meier survival analysis was used for survival analyses.

RESULTS

In this period, 55 children with ovarian mass were included in the study. The age of the patients ranged from two months to 18 years (median, 12.9 years). In addition, 17 patients (31%) were <10-year-old, while 38 patients (69%) were ≥ 10 years.

The patients' symptoms and signs are summarized in **Table 1**. The most common symptom was abdominal pain (n: 50, 91%). Four patients (7.3%) underwent emergency surgery for ovarian torsion.

The largest tumor diameter ranged from 3 cm to 28.5 cm (median, 11 cm). High AFP levels in 11 patients, high HCG levels in 10 patients and high LDH levels in 12 patients were found. In one of our patients with dysgerminoma, the LDH level was very high (value: 37155 IU/L, N: 125-243 IU/L).

Table 1. The patients' symptoms and signs at admission

Symptom/Signs	n (%)
Abdominal pain	50 (91)
Palpable mass	11 (20)
Menstrual disturbances	2 (3.6)
Enuresis	1 (1.8)
Weight loss	1 (1.8)
Constipation	1 (1.8)
Anorexia	1 (1.8)

While 51 of the patients underwent elective surgery (92.7%), four had emergency surgery (7.3%). The operations of the patients were performed by a pediatric surgeon (n = 45), a gynecologist (n = 9) and a general surgeon (n = 1). Of the surgeries performed, 28 were salpingo-oophorectomy (51.9%), 23 were oophorectomy (42.6%), and four were cystectomy (5.6%). Lymph node dissection was performed in three patients (5.5%) and these were performed by gynecologists.

The types of ovarian mass were germ cell tumors (n: 44, 80%), epithelial tumors (n: 4, 7.3%), stromal tumors (n: 3, 5.5%), simple cyst (n: 2, 3.6), and others (n: 2, 3.6) (**Table 2**).

Table 2. The tumor types of the children with ovarian mass

The tumor types	n (%)
Germ-Cell tumors	44 (80)
Benign	
Mature cystic teratoma	25 (45.5)
Mature teratoma	1 (1.8)
Malign	
Dysgerminoma	10 (18.2)
Yolk Sac tumor	7 (12.7)
Immature teratoma	1 (1.8)
Epithelial tumors	4 (7.3)
Benign	
Mucinous cystadenoma	2
Borderline	
Serous borderline tumor	1
Mucinous borderline tumor	1
Stromal tumors	3 (5.5)
Benign	
Serous cystadenofibroma	2
Malign	
Juvenile granulosa cell tumor	1
Simple cyst	2 (3.6)
Others	2 (3.6)
Non-Hodgkin lymphoma	2

Fourteen patients received chemotherapy. Twelve patients with germ cell tumors received the BEP protocol, while two patients with non-Hodgkin lymphoma received the LMB89 protocol. Two patients with malignant tumor died, the others were alive. Of these two patients who died, the patient with yolk sac tumor died with progressive disease. In the other patient with ataxia telangiectasia and dysgerminoma who died, central nervous system tumor developed as a secondary cancer approximately two years later and however, the guardian of the patients refused any treatment. As a result, this patient died due to a central nervous system tumor (14). Overall survival rate was 80.8% with median follow-up time 2.5 years (range, 1.5 months - 14 years) (**Figure 1**).

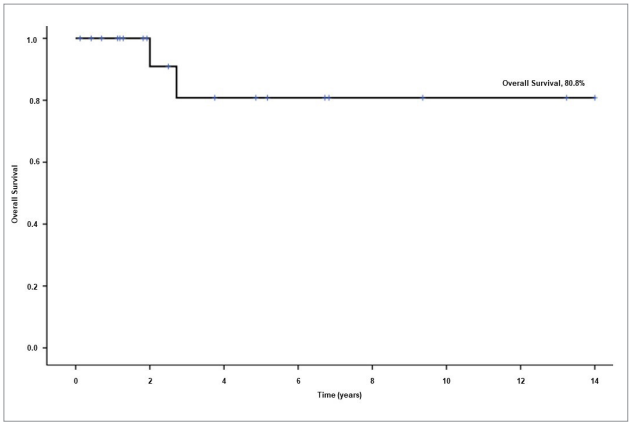

Figure 1. The overall survival of patients with malignant ovarian mass

DISCUSSION

In children, it should be kept in mind that, although most of the adnexal masses are benign unlike adults, approximately 1% may be malignant (15). In addition, almost all adnexal masses arise from the ovaries (1-5). In children, ovarian masses can be classified as functional lesions, simple cysts, epithelial tumors, stromal tumors, germ cell tumors, metastatic tumors and others, and 45% of them are functional lesions (5). Herein, the aim is to evaluate clinical features, treatment approaches and outcomes of children with ovarian mass, retrospectively. In our study, a significant portion of our patients were benign in nature, and most of the patients with malignancy had germ cell tumors.

Although the symptoms at the time of diagnosis may be variable in children with ovarian mass, the most commons are abdominal pain and palpable mass, also the ovarian mass should be kept in mind in children with menstrual disturbances or ovary torsion (1-5). Similarly, abdominal pain and palpable mass were the most common complaints in our patients. However, on rare occasions, menstrual disturbances, urinary system symptoms such as enuresis, gastrointestinal system symptoms such as constipation, weight loss, and anorexia were other symptoms.

Generally, AFP, HCG and LDH are commonly used tumor markers. Significant elevation of AFP is seen in yolk sac tumor, while mild elevations may be seen in immature or mature teratoma. Treatment response should also be monitored, taking into account the appropriate half-life

for that age, especially in patients with elevated AFP (8, 16). Another important tumor marker is HCG. High HCG level is usually seen in choriocarcinoma. However, mild HCG elevations can be seen in dysgerminoma and sometimes in yolk sac tumor. Both AFP and HCG elevations are generally seen in mixed germ cell tumors. Another important tumor marker that is sometimes forgotten is LDH. A marked elevation of LDH can be detected in mature or immature teratoma or yolk sac tumor, especially in dysgerminoma. In our patients, high AFP levels in 11 patients, high HCG levels in 10 patients and high LDH levels in 12 patients were found. In one of our patients with dysgerminoma, the LDH level was very high.

General treatment approach for ovarian masses in children is surgery. While only cystectomy is performed for ovarian cysts larger than five cm, salpingooforectomy or oophorectomy is preferred for other masses (1-5, 11, 17, 18). Lymph node dissection is not the preferred approach in children with malignant ovarian mass. In our study, except for four patients, all patients underwent elective surgery. Emergency surgery was performed in four patients due to ovarian torsion. The most preferred surgical intervention was salpingooforectomy or oophorectomy. Lymph node dissection was performed in three patients and primary operations of these patients were performed by gynecologists.

Ovarian tumors other than functional ovarian lesions in children, can be classified as simple cysts, epithelial tumors, stromal tumors, germ cell tumors, metastatic tumors and others (1-5, 10). In the study by Sonmez et al (5), functional lesions (44.6%), epithelial lesions (32.1%) and germ cell tumors (23.2%) were seen in order of frequency. In children, epithelial tumors are often benign and cystadenoma is the most common. Rarely, borderline tumor or more rarely malignant tumor may occur (10). In germ cell tumors, it is usually immature teratoma, dysgerminoma, yolk sac tumor, juvenile granulosa cell tumor, and mixed germ cell tumor at varying rates in different studies (11, 19). In our study, functional lesions were excluded. Germ cell tumors are important part of our patients (80%). According to the literature, the possible reason for this high rate is the complete recording of all benign ovarian masses such as "mature cystic teratoma" due to its close relationship with the pediatric surgery department. Dysgerminomas and yolk sac tumors were the most common malignant ovarian tumors.

In children, the most experience with chemotherapy is in malignant germ cell tumors. For many years, the BEP protocol containing cisplatin, etoposide and bleomycin or the JEB protocol containing carboplatin, etoposide and bleomycin have been preferred. In our study, BEP protocol was preferred, except for two patients with non-Hodgkin lymphoma and the JEB protocol was preferred in a patient with dysgerminoma due to the accompanying ataxia telangiectasia (14).

Success rates are good, depending on the stage, especially in germ cell ovarian tumors. In our study, the overall survival rate is 80.8%. One patient with ataxia telangiectasia was died. This patient was lost because of the development of a second cancer due to telangiectasia.

CONCLUSION

Most of the ovarian masses in childhood are benign. Harmonious collaboration is important in diagnosis and treatment of both benign and malignant ovarian masses.

ETHICAL DECLARATIONS

Ethics Committee Approval: Local Ethic Committee of XXX University, Faculty of Medicine (No: 2021/129, Date: 27 Jan, 2021).

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

Referee Evaluation Process: External double-blind referee assessment.

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

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ORIGINAL ARTICLE
ORİJİNAL ARAŞTIRMA

Clinical Characteristics and Long-Term Outcomes of Systemic Lupus Erythematosus in Children

Çocuklarda Sistemik Lupus Eritematozusun Klinik Özellikleri ve Uzun Dönem Sonuçları

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ABSTRACT

Aim: Systemic lupus erythematosus (SLE) is a common multisystemic autoimmune disease characterized by the presence of autoantibodies and multiorgan system involvement. The aim of this study was to describe the presenting clinical manifestations, laboratuary findings, clinical course and prognosis of SLE in children.

Material and Method: We performed a retrospective study patients with SLE, diagnosed before the age of 18 years. Clinical and laboratory data were collected from initial admission to study initiation.

Results: Thirty-five children and adolescents SLE (cSLE) were recorded, 85.7% of female. The median age at disease onset was 12(range 4-17) years, and median follow up duration was 5 (1-14) years. The most common clinical feature was arthritis (65.1%), followed by constitutional symptoms (48.6%), malar rash (31.4%), photosensitivity (5.7%), alopecia (5.7%) and oral ulcers (5.7%). Renal involvement accounted 4/5 of the patients (80%). Hematuria and proteinuria were the most frequent presenting findings (48.5% and 45.7% respectively). Renal biopsy was performed in 27 patients. According to WHO classification: 1 patient had classV nephritis, 10 had class IV, 4 had class III and 12 had class II nephritis. 20% of children developed neurologic symptoms. One patient died during the follow-up period. At the last follow up none of the patients had renal failure but, proteinuria persisted in 4 of them (11.4%). All neurological findings of patients are in remission apart from sequelae.

Conclusion: Clinical outcome was favorable in our patients. Renal involvement is common but progression to end stage kidney disease, at least in the short term, is rare.

Keywords: Children, initial presentation, prognosis, systemic lupus erythematosus

ÖZ

Amaç: Sistemik lupus eritematozus (SLE), otoantikorların varlığı ve çoklu organ sistemi tutulumu ile karakterize, yaygın bir multisistemik otoimmün hastalıktır. Bu çalışmanın amacı, çocuklarda SLE'nin klinik belirtilerini, laboratuvar bulguları, klinik seyrini ve prognozunu tanımlamaktır.

Gereç ve Yöntem: 18 yaşından önce tanı almış SLE'li hastaları retrospektif olarak incelendi.. Klinik ve laboratuvar verileri, ilk kabulden çalışmanın başlamasına kadar geçen sürede hastaların klinik ve laboratuvar verileri kaydedildi.

Bulgular: Otuz beş çocuk ve adölesan SLE (cSLE) hastanın %85,7'si kadındı. Ortalama hastalık başlangıç yaşı 12 (4-17 arası) yıl ve medyan takip süresi 5 (1-14) yıldı. En sık görülen klinik özellik artrit (%65,1) idi, bunu konstitusyonel semptomlar (%48,6), malar döküntü (%31,4), ışığa duyarlılık (%5.7), alopesi (%5,7) ve oral ülserler (%5,7) izliyordu. Böbrek tutulumu hastaların 4/5'ini (%80) oluşturuyordu. Hematüri ve proteinüri en sık başvuru bulgusuydu (sırasıyla %48,5 ve %45,7). 27 hastaya böbrek biyopsisi yapıldı. WHO sınıflamasına göre: 1 hastada sınıf V, 10 hastada sınıf IV, 4 hastada sınıf III ve 12 hastada sınıf II nefrit vardı. Çocukların %20 'sinde nörolojik semptomlar gelişti. Takip süresi boyunca bir hasta öldü. Son takipte hiçbir hastada böbrek yetmezliği gelişmedi, ancak 4'ünde (%11,4) proteinüri devam etti. Hastaların sekel dışında tüm nörolojik bulguları remisyondadır.

Sonuç: Hastalarımızda klinik sonuç olumluydu. Böbrek tutulumu yaygındır, ancak en azından kısa vadede son dönem böbrek hastalığına ilerleme nadirdir.

Anahtar Kelimeler: Çocuklar, ilk başvuru, prognoz, sistemik lupus eritematozus

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INTRODUCTION

Systemic lupus erythematosus (SLE) is a complex systemic autoimmune disease characterized by the presence of autoantibodies and multiorgan system involvement. Approximately 15-20% of patients present in the first two decades of life (1). It is the most common rheumatic disease associated with morbidity and mortality in children. Although the specific cause of SLE is unknown, genetic and environmental factors are involved in the pathogenesis. The disease is characterized by immune dysregulation involving both the innate and adaptive immune systems with polyclonal B-cell activation, which results in the production of self-reactive autoantibodies (2).

The clinical presentation at onset of SLE is heterogeneous; it may range from rash and arthritis to severe life-threatening multi-organ involvement. Childhood-onset SLE (cSLE) are known to have a more severe course with higher prevalence of renal and neuropsychiatric manifestations. In addition, cSLE is often more acute and severe, affecting multiple organs in children compared to adults. Therapeutic approaches involve immunosuppression and immunomodulation and are tailored to the severity of disease and the extent of organ damage (3). The aim of treatment is to target specific organ manifestation in order to achieve low disease activity.

Because of the different presentations, manifestations, and laboratory abnormalities in patients with cSLE, diagnosis can be challenging. The aim of this study was to describe the presenting clinical manifestations, laboratory findings, clinical course and prognosis of cSLE.

MATERIAL AND METHOD

This study was conducted with a retrospective review of the medical records of 35 SLE patients who were followed regularly in a tertiary referral hospital between January 1995 and December 2017. Inclusion criteria were: Fulfillment of the revised American College of Rheumatology (ACR) 1982 criteria for diagnosing SLE (4), age of onset of symptoms <18 years. The inclusion criteria were: fulfillment of revised American College of Rheumatology (ACR) 1982 criteria for diagnosis of SLE (4) , age at onset of symptoms at less than 18 years of age. Patients with missing data and who were followed up for less than six months were excluded from the study.

All patients were systematically reviewed for: demographic characteristics, age at disease onset, disease duration, follow-up duration, clinical features and laboratory variables as well as the treatment and outcomes. Clinical manifestations such as skin rashes, a fever above 38°C, photosensitivity, ulcers, arthritis/ arthralgia, serositis, nephritis (defined as mesangial, focal proliferative, diffuse proliferative or membranous glomerulonephritis according to the WHO (World Health Organisation) classification on histopathology, or persistent proteinuria of >0.5 g/day, proteinuria of >3.5 g/day, or cellular casts of any kind according to the ACR criteria) were recorded. Laboratory findings included urinary testing (with abnormalities defined as any one of the following: urinary casts, hematuria (>5 red blood cells/high power field), proteinuria (≥1+), pyuria (>5 white blood cells/high power field), complete blood and differential counts, erythrocyte sedimentation rate (ESR), serum C3 and C4 levels, antinuclear antibodies (ANA), anti-cardiolipin (aCL) IgG/M testing and double-stranded DNA (ds-DNA) antibody were recorded. Antinuclear antibodies and anti-dsDNA were determined by indirect immunofluorescence or enzyme-linked immunosorbent. Serum C3 and C4 were measured by immunodiffusion or turbidimetric immunoassay.

Data on induction and maintenance therapy were collected. Patients with WHO Class II LN were treated with combinations of tapering doses of oral prednisone and/ or azathioprine (AZA) depending on extra-renal disease. Patients with proliferative nephritis (WHO Class III and IV LN) were treated with intravenous methylprednisolone (30 mg/kg) pulse followed by 2 mg/kg/day (max 60 mg) oral prednisolon at the first month of therapy, and gradually tapered along with monthly intravenous cyclophosphamide (CYP) infusions for the first six months, followed by maintenance with either three monthly CYP infusions for 18 to 24 months or AZA (2–2.5 mg/kg/day) and low-dose oral prednisone. mycophenolate mofetil (MMF) was used both for induction and maintenance therapy in association with oral prednisone in a few patients. A minority of patients with refractory disease were treated with rituximab. Angiotensin-converting enzyme inhibitors (ACEIs) and/or angiotensin-receptor blockers (ARBs) were used in most patients for hypertension and/or proteinuria. Anticoagulants were used in patients with secondary antiphospholipid syndrome if indicated. Hydroxychloroquine was used both for the relief of joint and skin manifestations, and also to prevent flares of the disease.

The study was approved by the ethical committee of Ankara City Hospital (E2-21-532). All procedures were carried out in accordance with the ethical rules and the principles of the Declaration of Helsinki.

Statistical Analysis

Descriptive statistical analysis methods were used to evaluate the demographics and clinical data. The mean, median, standard deviation, range were calculated. Frequency tables were used to describe the categorical data. Data were analyzed using SPSS v.21 (SPSS Inc., Chicago, IL, USA).

RESULTS

This study consisted of 35 (30 female, female to male ratio was 6:1) patients with cSLE. The median age at disease onset was 12 (4-17) years. Only one child was below five years of age at diagnosis. At the time of diagnosis, 18 of patients ages were over 17 and 18 of them were girl. The median duration of follow-up was 5 (1-14) years. The follow up time was more than two years in 29/35 patients, while it was longer than five years in 21/35 of them.

The most common extrarenal clinical feature was non–deforming arthritis (65.1%), followed by, constitutional symptoms (48,6%) such as fever, fatigue and anorexia. The malar rash (31.4%) was the most common cutaneous manifestation followed by photosensitivity (8.6%) and alopecia (5.7%). Serositis was found in 17.1% of the patients.

Renal Involvement

Approximately 4/5 patients (80%) had renal involvement. Eighty–three percent of females and 100% of males had renal disease at diagnosis.

Hematuria and proteinuria were the most common presenting findings (48.5% and 45.7% respectively) of renal involvement. Seven patients (20%) had nephrotic syndrome. Three patients presented with nephritic features (hypertension and gross hematuria). Three patients had renal impairment at presentation (one Class IV nephritis, two Class II nephritis).

Table 1 summarizes the clinical and laboratory features of the patients at disease onset.

Renal biopsy was performed in 27 patients. According to WHO classification: 1 patients had class V nephritis, 10 had class IV, 4 had class III and 12 had class II nephritis. The renal findings of patients biopsied are given in **Table 2**.

Neurological Involvement

Neurological involvement (NP-SLE) was seen in seven patients (20%); 3 patients had headache, 2 had chorea, 1 had acute confusional state and 1 had seizure. Median age of these patients was 13 years.

During the treatment, psychosis developed in one patient. Her symptoms were not accompanied with flaring in immunological parameters and primary illness (lupus nephritis). She was managed symptomatically with antipsychotics and reduced steroid dosage.

Hematological and Immunological Findings

Hematological abnormalities were revealed in 21 patients (60%) at the time of diagnosis. Anemia was the most common hematological manifestations at onset (74%). Hemolytic anemia with positive Coomb's test was detected in 13 patients (37.1%). Twelve patients had leucopenia (34.3%), and 15 patients (42.9%) had thrombocytopenia.

Table 1. Clinical features of systemic lupus erythematosus patients at presentation

Characteristics	n (%)
Demographic data	
Age at diagnosis, median (range) years	12 (4-17)
Follow-up duration, median (range) years	5 (1-14)
Female/Male	30/5 (6/1)
Fever, fatigue, anorexia	17 (48.6%)
Cutaneous findings	
Malar rash	11 (31.4%)
Photosensitivity	3 (8.6%)
Alopecia	2 (5.7%)
Oral ulcers	2 (5.7%)
Weight loss	2 (5.7%)
Arthritis-arthralgia	23 (65,1%)
Serositis	6 (17.1%)
Pericardial	4 (11.4%)
Pleural	2 (5.7%)
Renal involvement	28 (80%)
Nervous system involvement	7 (20%)
Hematological and immunological involvement	21 (60%)
Cardiovascular involvement	6 (17.1%)

Table 2. Renal biopsy findings as per WHO classification among patients who had undergone a kidney biopsy in our series of pediatric SLE patients (n=27)

Histopathology	Number of patients n (%)	Follow up (at the end of the first year)	Follow up (at the end of the five years)
Minimal mesangial lupus nephritis (Class I)	0		
Mesangial proliferative lupus nephritis (Class II)	12 (44.4%)	11 normal 1 nephritic proteinuria	8 normal 1 nephritic proteinuria (Class III)* 1 nephrotic syndrome (Class IV)*
Focal lupus nephritis (Class III)	4 (14.8%)	3 normal 1 nephritic proteinuria	4 normal
Diffuse lupus nephritis (Class IV)	10 (37.0%)	6 normal 3 nephritic proteinuria 1 nephrotic syndrome	4 normal 2 nephritic proteinuria 2 nephrotic syndrome (Class IV)*
Membranous lupus nephritis (Class V)	1 (3.7%)	1 normal	
Advanced sclerosing lupus nephritis (Class VI)	0		

One patient presented with microangiopathic hemolytic anemia (MAHA), one with macrophage activation syndrome; a 15-years old male patient presented with microangiopathic hemolytic anemia and nephritic syndrome. He had thrombocytopenia (45,000/mm³), schistocytes and fragmanted erythrocytes on blood smear, elevated LDH (2255 IU, N: 0-248 IU/L), blood urea and creatinine (urea 69 mg/dL, creatinin 1,22 mg/dL), hypocomplementemia and elevated ANA, antids-DNA titer. Renal biopsy showed Class II lupus nephritis.

A 13 years old girl presented with macrophage activation syndrome. She has neither hypocomplementemia nor renal involvement but severe cutaneous erythema, vasculitis and onycomycosis. ANA and antidsDNA were strong positive. C1q deficiency was detected. She was treated with pulse methylprednisolon and cyclosporine at the acute attack then therapy continued with azathioprin and oral prednisolon. Two years later azathioprin switched to (MMF) because of the severe neutropenia. One year later, at the second attack of macrophage activation syndrome she died.

ANA positivity was detected in 32 patients (91.44%) and 88.6% of them had elevated anti-dsDNA titer. Low serum C3 levels were found in 24 (68.6%), low serum C4 levels were found in 27 (77.1%) patients (66.4±40,1 mg/dL; 12.5±8,09 mg/dL respectively).

Anticardiolipin antibodies (aCL) were positive 10 of the 35 patients. aCL was positive in 5 (71.4%) of 7 NP-SLE patients, while 5 (17.8%) of 28 non NP-SLE patients. Antiphospholipid syndrome (APLAS) developed in two patients. One of them had presented with catastrophic APLAS (seizure, necrotizing pancreatitis, unilateral renal artery thrombosis), the other patient presented with digital ulcers.

Treatment

All patients with class IV nephritis were treated with pulse metilyprednisolon and intravenous CYP for six months as an induction therapy then shifted to either azathioprine (6 patients) or mycofenolate mofetil (4 patients). All patients received hydroxychloroquin during follow up.

Prognosis

One patient died during the follow-up period from macrophage activation syndrome. At the last follow up none of them had renal failure but, proteinuria persisted in 4 of them (11.4%). Five of the 7 patients with neurological findings (71.4%) completely recovered, and two patients had long-term neurological complications, including one with cognitive impairment and a hemiplegic gait and one with visual impairment.

DISCUSSION

Childhood-onset systemic lupus erythematosus is a chronic, potentially fatal autoimmune disease with a great variability in disease presentation and course. In this study we described the characteristics of 35 children with cSLE seen in a tertiary referral hospital. Our report suggests that female gender was predominant and constitutional symptoms are the most common features at presentation. The current study showed that most cases of cSLE appeared after the pubertal period (>12 years of age). It might be explained by the hormonal effects on SLE patients after puberty has been also observed in most studies (5).

The presentation, clinical manifestations, immunological findings and treatment issues of cSLE are similar to those of adult SLE patients (6). On the other hand, pediatric and adult SLE differ in multiple aspects, and it is important to recognize these differences for optimal treatment and prognosis of these patients. In general, children with SLE tend to have a more severe disease at onset with higher rates of organ involvement and a more aggressive clinical course than adult-onset SLE patients (7, 8). In the first year of diagnosis, 35–90% of children will present with constitutional and generalized symptoms, 20–80% will have nephritis, 20–74% musculoskeletal symptoms, 60-80% any skin involvement, 15-30% neuropsychiatric disease, 5-30% cardiovascular disease, and 18-40% pulmonary disease (2).

The arthritis of SLE is typically non-erosive and non-deforming and involves larger joints such as knees, ankles, wrists and small joints of hands and feet. Arthralgia and arthritis were noted in 65.1% of our patients.

As expected, renal involvement accounted 4/5 of our patients (80%). Approximately 45% of patients had hematuria and proteinuria and 20% of patients had nephrotic syndrome at baseline. Renal impairment was seen in three patients at presentation. Ten patients had class IV nephritis according to WHO classification. Although renal involvement at the time of disease onset were associated with worse outcome, at the last follow up none of them had renal failure but, proteinuria persisted in 11.4% of them. A number of studies, but not all, have reported that the predominant clinical features of patients with Class IV, include hypertension, impaired kidney function, anemia, proteinuria, hypocomplementemia, and high anti-dsDNA titer (9, 10). Inconsistency in these results could be related to variations in the genetics, demographic area, and/or ethnicity of the study populations. Recently Srivastava et al. reported the renal outcome of 205 cSLE patients, revealing 46.7% Class IV nephritis. In this study renal survival at five, 10 and 15 years was 93.8%, 87.1% and 84%, respectively (11). Although multiple factors

individually predicted poor outcome, only raised serum creatinine at onset and damage accrual remained significant. In our series a transient elevation of serum creatinine was observed in three patients, and renal survive was found in 100% after a median follow up of five years.

Neuropsychiatric (NP) manifestations are found in approximately 25 to 90% of children and adolescent patients with SLE. Nearly half of the NP involvement will occur within the first year from the time of diagnosis. Headaches are common (50-75%), but true lupus headache is considered to be associated with significant intracranial pathology including cerebrovascular disease such as vasculitis and cerebral vein thrombosis. Following headache, psychiatric manifestations of pSLE are the most common NP manifestation. This may occur as part of the primary disease process, or secondary to central nervous system sequelae of SLE, e.g. seizures, infection or adverse effect of treatment with steroids, or immunosuppressant (12). Cerebrovascular disease, seizures and chorea are seen in about 25%, 20% and 5% of NP-pSLE patients respectively . In our study NP-SLE was seen in seven patients (20%); 3 patients had headache, 2 had chorea, 1 had acute confusional state and 1 had seizure.

Involvement of peripheral nervous system and the CNS has been reported to occur in 20% to 95% of patients with cSLE. Neuropsychiatric symptoms in children with SLE cause high morbidity and disability. The majority of patients with NP-SLE have the initial signs and symptoms within the first year of diagnosis of SLE; approximately 25% initially develop NP-SLE more than 2 years after disease onset (2) . Neuropsychiatric involvement may result in more damage and poorer outcome (13) . Various autoantibodies have been implicated in the pathogenesis of NP-SLE, including antiribosomal P protein, antineuronal, antiganglioside, antiphospholipid (aPL), and antiendothelial antibodies (14-16). One of the most intriguing issues is the association of neuropsychiatric manifestations with aPL. These autoantibodies can lead to thrombosis of cerebral vessels, both arterial and venous and a nonthrombotic, immune-mediated neurologic impairment has been associated with aPL. The risk of NPSLE is three times greater in children with systemic lupus erythematosus and a positive lupus anticoagulant which are also specific risk factors for stroke in children and adults (17) . In our study, tests for aCL were carried out 10 of 35 patients with SLE and 5 (71.4%) of 7 patients with NP-SLE.

Zuniga et al. defined seizures, migraine and depression as the most common NP symptoms in 30 of 90 cSLE patients (18) . Lupus anticoagulant (LAC) was found positive in high titers in these patients, and they proposed that immunesupressive treatment could avert or delay NP symptoms. Avcin et al. also found a persistently positive aCL in 50%, anti-β2GPI in 29%, and LAC in 16% of children with SLE (19) . They found that the prevalence of anti-β2GPI antibodies was found to be higher in the group of SLE patients with neuropsychiatric disease compared with those without. Their study suggested an association between LAC and cerebrovascular disease at the time of SLE diagnosis and chorea over the disease course, but not between aPL and other neuropsychiatric manifestations (20) .

Recent cSLE studies have shown 5-year survival rates of >95%, with 10-year survival rates reported to be as high as 86%. Mortality rates have been shown to be associated with socioeconomic status and individual access to health care, educational background, racial/ethnic background, endemic infection rates, disease activity, and renal or central nervous system involvement (20) . Common causes of death within the first 2 years are pancreatitis, pulmonary hemorrhage, infection, thromboembolic disease, and active neuropsychiatric disease. The causes of late death (>5 years) are complications of end-stage renal disease, atherosclerosis, suicide, and less commonly active SLE or infection (21). Singh et al. performed a renal biopsy in 53 of 72 cSLE patients and observed Class IV nephritis in 35. Mortality occurred in 22 children (30%), with half of these occurring at presentation. The two important causes of death were infection and end-stage renal disease (22). In our series of 35 children only one patient died from macrophage activation syndrome.

CONCLUSION

The current study provides longterm outcome of LN in cSLE. Long-term patient and renal survival rates in our series are favorable. Children with cSLE and having aCL positivity should be followed closely for NP symptoms.

ETHICAL DECLARATIONS

Ethics Committee Approval: This study was approved by the the Ethics Committee of Ankara City Hospital (E2-21-532).

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

Referee Evaluation Process: External double-blind referee assessment.

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

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ORIGINAL ARTICLE
ORİJİNAL ARAŞTIRMA

Ergenlerde, Aktif ve Pasif Sigara İçiminin Solunum Fonksiyon Testleri Üzerine Etkisi

The Effects of Active and Passive Smoking on Pulmonary Function in Adolescent

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Öz

Amaç: Bu çalışmanın amacı ergenlerde aktif ve pasif sigara içiminin solunum fonksiyonları üzerine etkisini belirlemektir.

Gereç ve yöntem: Bu çalışma, Haziran 2013-2015 tarihleri arasında, Ankara Eğitim ve Araştırma Hastanesi Çocuk Polikliniğine başvuran ergenler dahil edildi. Olguların ve ailelerinin sosyodemografik özelliklerini içeren anket formu ebeveynler tarafından doldurduldu. Olgular, aktif sigara içenler, pasif sigara içenler ve hiç içmeyenler olmak üzere üç gruba ayrıldı. Tüm olguların volumetrik spirometre ile solunum fonksiyon testleri yapıldı.

Bulgular: Çalışmaya alınan toplam 222 ergenin yaş aralığı 11-17 yaş (median:15 yaş) olup 102'si (%45,9) erkek ve 120'si (%54,1) kızdı. Aktif sigara içenler %29,7 [n= 66, median 16 yaş, 42 erkek (%64) ve 24 kız (%36)], pasif sigara içenler %55,4 [n= 123, median 14 yaş, 52 (%42,3) erkek ve 71 kız (%57,7)] ve hiç içmeyenler %14,9 [n=33, median 14 yaş, 8 erkek (%24,2) ve 25 kız (%75,8)] idi. Pasif sigara içenlerin FEF25-75 ve FEV1 değerleri aktif sigara içenlere göre farklı bulunmadı. Ancak, hiç içmeyenlerle karşılaştırıldığında anlamlı daha düşüktü (sırası ile p=0,049; p=0,003). Pasif sigara içen grubun FVC değerleri aktif sigara içen ve hiç sigara içmeyen gruba göre anlamlı olarak düşük saptandı (sırası ile, p=0,020; p=0,010).

Sonuç: Ergenlik döneminde sigara içme oranı yüksek saptandı (%29,7). Pasif sigara içen grubun solunum fonksiyon testlerinin anlamlı düşük çıkması, sigaraya maruziyet oranının, maruziyet tipinden (aktif/pasif) daha etkili bir faktör olduğu kanısını ortaya koymuştur.

Anahtar Kelimeler: Ergen, sigara içimi, pasif sigara maruziyeti, solunum fonksiyon testleri

Abstract

Purpose: To determine the effects of smoking and exposure type (active and passive) on pulmonary functions in adolescents.

Material and Method: Adolescents, who admitted to ages Ankara Training and Research Hospital's Pediatric Outpatient Clinics between June 2013 and 2015 were included in the study. The demographic parameters and sociodemographic characteristics of the cases and families were recorded in the questionnaired fulfilled by the parents. Three groups were constituted: Active smokers, passive smokers and nonsmokers. All of the cases' pulmonary function tests were determined with volumetric spirometry.

Results: A total of 222 adolescents, ages between 11-17 years [median 16 years,120 girls (54,1%), 102 boys (45,9%)] were included in the study. Active smokers were 29.7% [n: 66, median 16 years old, 24 girls (36%), 42 boys (64%)], passive smokers were 55.4% [n:123, median 14 years old, 71 girls (57,7%), 52 boys (42,3%)] and nonsmokers were14,9% [n: 33, median 14 years old, 25 girls (75,8%), 8 boys (24,2%)]. Passive smokers' FEF25-75 and FEV1 values did not show a significant difference according to active smokers, however were significantly lower than nonsmokers (respectively; p=0.049 and p=0.003). Passive smokers also had significantly lower FVC values than active smokers and nonsmokers (respectively; p=0.020; p=0.010).

Conclusion: Prevelance of smoking habit is high in adolescents (29.7%). Determining the respiratory function tests of passive smokers signicantly lower, suggest us that the amount of exposure is a more effective factor than type of exposure (active/passive).

Keywords;Adolescent, smoking, passive smoking, pulmonary function tests

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GİRİŞ

Pasif sigara içimi de aktif sigara içimi gibi ciddi ve ölümcül hastalıklara neden olabilen bir durumdur. Pasif içiciliğinin çocuklar ve bebeklerde özellikle solunum sistemi ve orta kulak hastalıklarına yakalanma riskini arttırdığı bildirilmiştir. Pasif içicilik, kısa ve az bile olsa zarar verebilmektedir ve bunun güvenli bir seviyesi de bulunmadığı kabul edilmektedir. Pasif içicilerin de aktif sigara içenler gibi erken ölüm ve artmış hastalıklar için büyük risk altında olduğu bilinmektedir (1).

Türkiye'de, 2012 yılından sonra tütün kullanımı oranları hem erkek hem de kadınlarda artış göstermiştir (2). Dünya Sağlık Örgütü (DSÖ) raporlarına göre, 2015 yılında ülkemizde 15 yaş ve üzeri olan erkeklerin %40,2'si, kadınların %12,4'ü, toplamda ise %25,4 oranında olmak üzere 14.892.000 tütün kullanıcısı bulunmaydı (3). Dünya genelinde 2016 yılında, 7,1 milyon kişinin tütün kullanımına bağlı hastalıklar sonucunda öldüğü bildirilmiştir. Bu ölümlerin 6,3 milyonunu aktif sigara içenler, 884 binini de pasif sigara içenler oluşturmuştur (4). Tüm dünyada çocukların %40'ının evde sigaraya maruz kaldığı tahmin edilmektedir (5).

Türkiye'de 2017 yılına gelindiğinde ise, tütün kullanımında daha da artma olduğu görülmektedir. Tütün kullanımı oranları 2017'de erkeklerde %42, kadınlarda %16,9 ve toplamda %29,5'e ulaşmıştır (6). Ülkemizde sigarayı bırakma oranının %27,2'den 13,6'ya düştüğüne, bunun nedenleri arasında televizyon ya da radyoda sigara karşıtı uyarılar görmenin azalması ve sigara reklamlarının artması olarak işaret edilmiştir (2). Ülkemizde çocukların durumuna bakıldığında, ergenlerde 13-15 yaş arası erkek çocuklarda günlük tütün kullanım oranı %23,2, kızlarda %12,1, toplamda %17,9'a yükseldiği görülmüştür (7).

Aktif sigara içen yetişkinlerde, solunum fonksiyon bozuklukları oluştuğu, birinci saniyedeki zorlu ekspirasyon volümü (FEV1), FEV1'in zorlu vital kapasite (FVC)'ye oranının (FEV1/FVC) ve zorlu ekspirasyonun orta yarısındaki ortalama akım hızı (FEF25-75)'nın daha düşük olduğu raporlanmıştır (8-13). Benzer şekilde ergenlerde de aktif ve pasif sigara içiminin FEV1, FEV1/FVC ve FEF25-75 değerlerini etkileyebileceği bildirilmiştir (12-16).

Birçok ülkede bu soruna karşı önemli adımlar atılmış ve pasif sigara içiminin önüne geçilmeye çalışılmıştır. Ülkemizde de bu önemli sorun dikkate alınmış ve önemli kararlar ve önlemler hayatımıza girmiştir. Buna rağmen pasif sigara içimi etkilerini sürdürmektedir (2, 6). Özellikle çocuklar ebeveynlerinin kullanımına bağlı olarak etkilenmektedirler.

Bu çalışmanın amacı; ergen yaş grubunda sigara içme sıklığını belirlemek ve sigaranın solunum fonksiyonları üzerindeki olumsuz etkilerini ortaya koymaktır.

GEREÇ VE YÖNTEM

Çalışmaya, Haziran 2013-2015 tarihleri arasında Ankara Eğitim ve Araştırma Hastanesi Çocuk Polikliniklerine başvuran, 11-17 yaş aralığındaki 222 ergen dahil edildi. Ergenlerden ve ebeveynlerinden yazılı onam alındı. Çalışmaya katılmayı kabul eden ebeveynlere, 19 sorudan oluşan seçmeli anket formu verildi. Çalışmaya dahil edilen ergenlerin sosyodemografik özellikleri, ebeveynlerin okur-yazar durumu ve sigara içimi, anket formundaki verilerden elde edildi. Fizik muayenede enfeksiyonu veya öyküsünde astım, bronşit ve kronik hastalığı bulunanlar çalışmaya dahil edilmedi.

Olgular sigara içme durumlarına göre üç gruba ayrıldı:

1. **grup (sigara içen):** Günde en az bir adet sigara kullanan olgular. Bu grupta ebeveynde sigara kullanımı aranmadı.
2. **grup (pasif sigara içen):** Hiç sigara kullanmamış, ebeveynlerden en az bir tanesi günlük bir adet ve üzeri sigara kullanan olgular.
3. **grup (sigara maruziyeti olmayan):** Hiç sigara kullanmamış ve ebeveynlerinde sigara kullanımı olmayan olgular

Her üç gruba solunum fonksiyon testi yapıldı. Ölçüm kuru portabl flow volümetrik spirometre cihazı kullanılarak gerçekleştirildi. Çocuklara testin uygulanışı hakkında bilgi verildi. Solunum fonksiyon testi yapma sertifikası olan hemşire eşliğinde ölçümler yapıldı. En az üç deneme yapılarak aygıtın seçtiği en iyi değer çalışmaya alındı. Her üç gruptaki çocukların, zorlu vital kapasite (FVC), birinci saniyedeki zorlu ekspirasyon volümü (FEV1), FEV1/FVC ve zorlu ekspirasyonun orta yarısındaki ortalama akım hızı (FEF25-75) parametreleri ölçüldü. Yüzde değerler spirometre verileriyle otomatik olarak hesaplandı. "İnsan" öğesinin içinde bulunduğu bu çalışma, Helsinki Deklarasyonu prensiplerine uygun olarak yapılmıştır. Etik kurul izni: (18/01/2013) 513:4295

İstatistiksel Analiz

Verilerin analizi IBM SPSS (Stastistical Package For the Social Sciences) 20 paket programında yapıldı. Sürekli ve kesikli sayısal değişkenlerin dağılımının normale yakın olup olmadığı Kolmogorov Smirnov testiyle araştırıldı. Normal dağılıma uyan değişkenler (yaş) ortalama±standart sapma (SD), uymayanlar ortanca [minumum, maximum (min-max)], kategorik değişkenler ise olgu sayısı ve "%" biçiminde gösterildi. Sonuçlar %95 güven aralığında, anlamlılık $p<0,05$ düzeyinde değerlendirildi.

Sürekli verilerin normal dağılıma uygunluğu Shapiro-Wilk testi ile incelendi. Normal dağılıma uyan verilerin üç grupta karşılaştırılmasında Tek Yönlü Varyans Analizi (ANOVA) kullanıldı. Gruplar arasındaki farkın hangi gruplardan kaynaklandığı Tukey test ile incelendi.

Normal dağılıma uymayan verilerin üç grupta karşılaştırılmasında Kruskal Wallis Varyans Analizi kullanıldı. Gruplar arasındaki farkın hangi gruplardan kaynaklandığı Kruskal Wallis çoklu karşılaştırma testi ile incelendi. Nominal değişkenlerin grup karşılaştırmalarında (çapraz tablolarda) Ki-Kare test kullanıldı.

Değerlendirmelerde istatistiksel anlamlılık sınırı olarak p<0,05 kabul edildi.

BULGULAR

Çalışmaya dahil edilen 11-17 yaş arası 102 (%45,9) erkek ve 120 (%54,1) kız olgunun ortanca yaşları 15 yaş olarak bulundu. Cinsiyetlere göre istatistiksel fark saptanmadı (p=0,105). Olguların sosyodemografik özellikleri incelendiğinde aktif sigara içen grubun ortanca yaşı 16 (12-17) yıl olarak bulundu; aktif sigara içen grubun ortanca yaşı, pasif sigara içen ve içmeyen gruba göre istatistiksel olarak anlamlı daha büyük bulundu (sırasıyla p<0,001; <0,001) (**Tablo 1**). Aktif sigara içen grupta erkek cinsiyet sıklığı, pasif sigara içen ve içmeyen gruba göre istatistiksel olarak anlamlı büyük bulundu (sırasıyla, p<0,01; <0,001).

Aktif sigara içen grupta, sigara içme süreleri incelendiğinde olguların %21,2'si bir yıldan az, %56,1'i bir-üç yıl, %15,2'si dört-altı yıl, %7,6'sının altı yıldan fazla süredir sigara içtiği saptandı. Sigara içme sürelerine ve miktarlarına göre FEV1, FVC, FEV1/FVC, FEF25-75ve PEF değerleri ayrı ayrı karşılaştırıldığında anlamlı fark bulunmadı (p>0,05). Günde 10 adet altında içenlerin oranı %69,7 olarak bulundu. Ebeveynlerin sigara içme oranı değerlendirildiğinde kadınlarda %36,5 (n=81), erkeklerde %69,4 (n=154), olarak bulundu.

Pasif sigara içimi olan grubun FVC parametrelerinin ortalaması (± SD) 100,1±13,1 olup aktif sigara içen ve içmeyen gruba göre anlamlı olarak düşük saptandı (sırası ile, p=0,020; 0,010) (**Tablo 2**). Pasif sigara içimi olan grubun FEV1 ve FEF25-75 parametrelerinin ortalaması (± SD) 105,5±15,4 ve 104,9±28,2 olarak bulundu. Bu değerler sigara içmeyen gruba göre anlamlı düzeyde düşük saptandı (sırasıyla, p=0,003; 0,049).

Annelerin %53,1'i (n=43), babaların %48,7'si (n=75) 10 paket/yıl ve üstünde sigara içmektedirler. Aktif sigara içen ergenlerde babası 10 paket/yıl ve üstünde sigara içme oranı %52,5 (n=21) iken pasif sigara içen ergenlerde bu oran %47,4 (n=54) olarak bulundu (p=0,576).

Yine aktif sigara içen ergenlerde annesi 10 paket/yıl ve üstünde içenlerin oranı %57,7 (n=15) iken, pasif sigara içen ergenlerde bu oran %50,9 (n=28) olarak bulundu (p=0,568). Aktif sigara içen ve pasif sigara içen ergenlerin anne ve babalarının 10 paket/yıl ve üstünde sigara içme oranları arasında fark saptanmadı (p>0,05).

Tablo 1. Çalışma grubunun sosyodemografik özellikleri

Özellikler	Grup 1 Sigara içen n=66	Grup 2 Pasif sigara içen n=123	Grup 3 Sigara içmeyen n=33	Toplam n= 222	p
Ortanca yaş (en küçük-en büyük)	16 (12-17)	14 (11-17)	14 (12-17)	15 (11-17)	0,001
Cinsiyet, n (%)					
Erkek	42 (63,6)	52 (42,3)	8 (24,2)	102 (45,9)	<0,000
Kız	24 (36,4)	71 (57,7)	25 (75,8)	120 (54,1)	
Anne ortanca yaşı (en küçük-en büyük)	42 (30-64)	39 (27-52)	38 (30-60)	39 (27-64)	0,003
Baba ortanca yaşı (en küçük-en büyük)	44 (30-77)	42 (32-60)	44 (34-61)	43 (30-77)	0,014
Anne eğitimi n (%)					
Yok	16 (24,2)	49 (39,8)	15 (45,5)	80 (36,0)	
Orta	35 (53,0)	59 (48,0)	12 (36,4)	106 (47,7)	0,086
Lise ve üzeri	15 (22,7)	15 (12,2)	6 (18,2)	36 (16,3)	
Baba eğitimi n (%)					
Yok	41 (62,1)	66 (53,6)	13 (39,4)	120 (54,0)	
Orta	11 (16,7)	35 (28,5)	9 (27,3)	55 (24,8)	0,110
Lise ve üzeri	14 (21,2)	22 (17,9)	11 (33,3)	47 (21,2)	

Tablo 2: Çalışma grubunun solunum fonksiyon testlerinin değerlendirilmesi (ortalama±SD)

Özellikler	Grup 1 Sigara içen n=66	Grup 2 Pasif sigara içen n=123	Grup 3 Sigara içmeyen n=33	Toplam n= 222	p
FVC	105,7±15,7	100,1±13,1	107,9±12,9	102,9±13,9	0,002
FEV1	110,5±17,1	105,5±15,4	115,9±15,9	108,5± 16,4	0,002
FEV1/FVC	102,7±10,6	103,4± 8,2	105±8,2	103,4± 8,9	0,483
FEF25-75	111,9±34,3	104,9±28,2	118,8±26,7	109,1± 30,2	0,042
PEF	101,6±19,4	97,6±15,8	101,8±16,9	.99,4± 17,2	0,211

Anne ve babaların sigara içme süreleri 5 yıldan az olanlarla 5 yıldan fazla olanların FEV1, FVC, FEV1/FVC, FEF25-75 ve PEF değerleri karşılaştırılmış olup anlamlı fark bulunmadı (p>0,05). Yine günlük 1 paketten az ve 1 paketten fazla sigara içen anne ve babaların çocuklarının FEV1, FVC, FEV1/FVC, FEF25-75 ve PEF değerleri karşılaştırıldığında anlamlı fark saptanmadı (p>0,05).

TARTIŞMA

Sigara kullanıcıların çoğu 25 yaşından önce, bunların çoğunluğu da ergenlik döneminde sigaraya başlamaktadır (17). Türkiye'de farklı illerde yapılan çeşitli çalışmalarda 18 yaş altı sigara içme sıklığının %9,5 ile %41,2 arasında olduğu gösterilmiştir (17,18). Çalışmamıza katılan ergenlerde aktif sigara içme sıklığı %29,7 ve aktif sigara içenlerin yaş ortalaması 16'dır. DSÖ, 2008 yılında yayınlamış olduğu küresel tütün salgını raporunda, gelişmekte olan ülkelerde ergenlerde aktif sigara içme sıklığının hızla artmakta olduğunu ifade etmiştir (19). Sağlık Bakanlığı Sağlık İstatistikleri Yıllığı 2018 verilerine göre de 15-24 yaş aralığında her gün tütün kullanan birey sayısı 2012 yılında %14,3 iken 2016 yılında bu oran %18,1'e yükselmiştir (20). Türkiye İstatistik Kurumunun en son 2012'de yayımlanan araştırmasında, ülkemizde 15 yaş ve üzeri bireylerin %27'si her gün veya ara sıra tütün kullanmaktadır. Cinsiyete göre değerlendirildiğinde bu oran erkeklerde %41,4, kadınlarda %13,1'dir (21).

Çalışmamızda ebeveynlerin %52'sinin sigara içtiği ve içme oranının erkeklerde %69,4, kadınlarda ise %36,5 olduğu görülmüştür. Ülkemizde yapılan çeşitli çalışmalara göre bu oranlar farklılık göstermektedir. Özyurt'un bir ilkokulda okuyan çocuklara yönelik çalışmasında, çocukların büyük çoğunluğunun evinde en az bir kişi sigara içicisi olup çocuklar sigara dumanına maruz kaldığı belirtilmektedir (22). Küçük ve arkadaşlarının Yozgat'ta 873 hasta üzerine yaptığı bir çalışmada, çocukların %33,6'sının evde sigara dumanına maruz kaldığı ve maruz kalan çocukların da %38,9'unda solunum sistemi hastalığı görüldüğü belirtilmiştir (23). 2016 yılında Antalya'da 0-11 yaş çocukların evde sigara dumanından pasif etkilenmelerini araştıran başka bir çalışmada annelerin %31'inin, babaların %45,1'inin sigara içtiği ve sigara içen annelerin %80'inin, babaların ise %76,5'inin evde de sigara içtiği tespit edilmiştir (24). Nazlıer'in alt solunum enfeksiyonu nedeniyle hastaneye başvuran 5 yaş altı çocuklarda pasif içiciliğin etkisinin araştırılmasına yönelik doktora tezinde annelerin %30,6'sının babaların ise %82,9'unun sigara içtiği ve sigara içen annelerin %67,9'unun, babaların ise %69'unun çocukların yanında sigara içtiği görüldüğü belirtilmiştir (25).

Literatürde erişkinlerde sigaranın solunum fonksiyonları üzerine etkisini araştıran birçok çalışma olmasına rağmen ergenlerde yapılmış çalışma sayısı kısıtlıdır. 10-18 yaş arası 10.010 olgu ile yapmış bir çalışmada aktif sigara içenlerde FEV1 değerinin sigara içmeyenlere göre anlamlı olarak düştüğü tespit edilmiştir (26). Bird ve Orzco'nun 13-15 yaş arası 300 öğrenci ile yaptığı bir çalışmada da aktif sigara içenlerin FEV1, FEV1/FVC ve FEF25-75 değerleri sigara içmeyenlere göre anlamlı düşük bulunmuştur (8). Fakat Ghanem ve Hage'nin 18-24 yaş arası üniversite öğrencilerinde aktif ve pasif sigara içiminin etkilerini araştırdığı çalışmada sigara içen ve içmeyenlerin FEV1 düzeyleri arasında anlamlı fark bulunamamıştır (27). Benzer şekilde Jawed S ve arkadaşlarının 19-25 yaş arasındaki gençlerde yapmış olduğu başka bir çalışmada da aktif sigara içen ve içmeyenler arasında FEV1 değerlerinde anlamlı fark bulunamamıştır (28). Aynı çalışmada günde içilen sigara sayısı ile FEV1 ilişkisi değerlendirildiğinde günde 10 ve üzeri sigara içenlerin daha az sigara içenlere göre FEV1 değerleri anlamlı düşük bulunmuştur (28). Çalışmamızda aktif sigara içenlerin FEV1 değerleri, pasif sigara içenler ve hiç içmeyenlerle karşılaştırıldığında istatistiksel olarak anlamlı fark bulunmamış olsa da aktif sigara içenlerin FEV1 değerleri, sigara içmeyenlere göre düşük bulunmuştur. Bunun nedeni sigara içen ergenlerin günlük sigara miktarının ve sigara içme süresinin düşük olması olabilir.

Guerra ve arkadaşları prospektif olarak aktif ve pasif sigara maruziyetinin akciğer fonksiyonuna etkisini değerlendirdiği bir çalışmada 11, 16 ve 22 yaşlarındaki bireylerin FEV1/FVC değerlerini kıyaslamıştır. Çalışmada hem aktif sigara içenlerde hem de pasif sigara içenlerde aynı yaşlardaki içmeyenlerle FEV1/FVC değerleri kıyaslandığında aralarında fark bulunmamıştır. Halbuki aynı çalışmada 11 ve 26 yaşlarındaki FEV1/FVC oranları karşılaştırıldığında aktif ve pasif sigara içenlerde FEV1/FVC oranlarında anlamlı düşüş görülmüştür (29). Thacher ve arkadaşlarının 16 yaşındaki 2295 ergenle yaptığı başka bir çalışmada aktif sigara içenler ile sigara maruziyeti olmayanların FEV1/FVC oranları kıyaslanmış olup aktif sigara içenlerde anlamlı düşük bulunmuştur (30). Çalışmamızda aktif sigara içen, pasif sigara içen ve sigara içmeyenler arasında yapılan karşılaştırmada FEV1/FVC oranlarında istatistiki olarak anlamlı fark görülmese de sigara içenlerde FEV1/FVC oranı ortalaması düşük bulunmuştur.

Çalışmamızda pasif sigara içenlerin sigara içmeyenlere göre FEV1 değerleri daha düşük saptanmış olup Plata ve arkadaşlarının (31) Meksika'da 8-17 yaş arası 1632 erkek ve 1555 kız olgu ile yapmış olduğu bir çalışmada da çalışmamıza benzer şekilde pasif sigara içenlerin FEV1 ve FVC değerlerinin sigara içmeyenlere göre anlamlı olarak düştüğü tespit edilmiştir. Yine Merghani ve Saeed'in (32) yaptığı bir çalışmada da 9-14 yaş aralığında 135 erkek öğrenci değerlendirilmiş ve pasif sigara maruziyeti olan 69 çocuğun sigara maruziyeti olmayanlara göre ortalama FVC değerinin %8, FEV1 değerinin %7 düşük olduğu

gösterilmiştir. Uysal ve arkadaşlarının (33) yaptığı bir çalışmada ise pasif sigara içici olan grupla içici olmayan grup karşılaştırılmış olup solunumsal parametreler arasında anlamlı bir fark bulunamamıştır.

Pasif sigara dumanına maruz kalan ergenlerin FEV1 ve FEF25-75 değerlerinin sigara maruziyeti olmayanlara göre düşük olması erken dönemden itibaren pasif sigara maruziyetinin etkin olduğunu göstermektedir. Milanzi ve arkadaşlarının 12 ve 16 yaşlarındaki 552 ergenle yaptığı bir çalışmada çocukluk çağı pasif sigaraya maruz kalma ihtimali yüksek olanlarda düşük olanlara göre kıyaslandığında 12 ve 16 yaşları arasındaki yıllık FEV1 büyümesinde azalma olduğu görülmüştür (34).

Çalışmamızda bazı kısıtlılıklar bulunmaktadır. Adolesan dönemde sigara içen çocukların, ailelerinden çekinerek, bu konuda doğru bilgi vermeyebileceği gibi sigara içen anne ve babaların da suçluluk duygusuyla doğru söylemeyebilirler. Çalışmamızda çocukların sigara kullanımı ebeveyn ve ergenlerin beyanına göre öğrenilmiş olup ergenlere ve ailelere objektif olarak nikotin metaboliti bakılamamıştır.

SONUÇ

Sigara içme sıklığının ergenler ve erişkinlerde yükseldiği tespit edilmiştir. Pasif sigara içicilerin solunum fonksiyon testinin aktif sigara içenlere göre daha düşük çıkması maruziyet oranının, maruziyet tipinden (aktif/pasif) daha etkili bir faktör olduğu kanısını desteklemiştir. Genel olarak toplumda tütün kullanımının azaltılmasına yönelik özel olarak da pasif sigara içiciliği maruziyetinin ve oranının azaltılmasına yönelik, projelerin desteklenmesi ile ergen sağlığı korunabilir. Bununla birlikte, toplum, aile ve bireylere yönelik eğitim ve bilinçlendirme faaliyetleri ile daha etkin önlemlerin alınması gerektiği aşikardır.

Daha geniş ve kapsamlı çalışmalarla, ergenlerde aktif ve pasif sigara içimi prevalansının ve zararlı etkilerinin ortaya konularak bilgilendirme ve önleme çalışmalarının yapılması gerekmektedir.

ETİK BEYANLAR

Etik Kurul Onayı: Araştırma için etik kurul onayı alınmıştır (18/01/2013) 513:4295.

Aydınlatılmış Onam: Çalışma retrospektif olarak dizayn edildiği için hastalardan aydınlatılmış onam alınmamıştır.

Hakem Değerlendirme Süreci: Harici çift kör hakem değerlendirmesi.

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

Finansal Destek: Yazarlar bu çalışmada finansal destek almadıklarını beyan etmişlerdir.

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

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CASE REPORT
OLGU SUNUMU

Primary Lymphedema: A Newborn Case

Primer Lenfödem: Bir Yenidoğan Vakası

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Abstract

Lymphedema is a condition characterized by generalization or regional accumulation of protein-rich interstitial fluid as a result of impaired lymphatic circulation due to congenital or acquired reasons. Lymphedema infection may occur as a result of secondary neoplasm, surgery, trauma, and radiation while most childhood cases are primary lymphedema. In cases of primary lymphedema, there is an erroneous development or function of the lymphatic system and most of them are thought to have a genetic predisposition. We aim to present a case of primary congenital lymphedema with a history of kinship between parents and no dysmorphic findings, born with lymphedema on both feet.

Keywords: primer lymphedema, edema on the dorsum of feet, newborn, lymphoscintigraphy

Öz

Lenfödem; lenfatik dolaşımın konjenital veya edinsel bazı nedenlere bağlı olarak bozulması sonucu, proteinden zengin interstisiyel sıvının jeneralize veya bölgesel olarak birikimiyle karakterize bir durumdur. Lenfödem enfeksiyon, neoplazm, cerrahi, travma ve radyasyona sekonder olarak gelişebilirken, çocukluk çağındaki olguların çoğu primer lenfödem şeklindedir. Primer lenfödem olgularında lenfatik sistemin hatalı gelişimi veya işlevi söz konusudur ve çoğunda genetik yatkınlık olduğu düşünülmektedir. Amacımız, anne baba arasında akrabalık öyküsü olan ve dismorfik bulguları olmayan, her iki ayak sırtında lenfödem ile doğan, bir primer konjenital lenfödem olgusunu sunmaktır.

Anahtar Kelimeler: Primer lenfödem, ayak sırtında ödem, yenidoğan, lenfosintigrafi

INTRODUCTION

Primary lymphedema is a disease that can manifest from the intrauterine period, characterized by edema that begins unilaterally or from the dorsum of both feet and can progress towards the lower extremity, developing due to developmental anomaly of lymph vessels (1). Primary lymphedema may be associated with genetic diseases such as trisomy 13, 18, 21, primary congenital lymphedema, Turner syndrome, Noonan syndrome, lymphoedema-distichiasis, or yellow nail syndrome (2,3). Milroy syndrome developing due to gene mutation should be considered (4,5) as VEGFR-3 (FTL-4) with an autosomal dominant transition in cases with normal karyotype and positive family history, without syndromic findings. While primary lymphedema may be present from birth, it may occur later in the early period. Extremity involvement is unilateral or bilateral (6). Its timely diagnosis is especially important in order to prevent worsening of the situation in cases with visceral involvement. The purpose of treatment is based on preventing infections and complications that may develop. For this, the following are recommended: extremity involvement, exercise, massage, tightening bandage, and skincare (1).

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CASE REPORT

A male baby born from the fifth pregnancy of a forty-four-year-old mother at 39 weeks of gestation, weighing 3490 g by cesarean section, was admitted to the neonatal intensive care unit because of respiratory distress in the delivery room and edema on the dorsum of both feet. It was learned that there was a second degree of kinship between the mother and father in the patient's family history, the mother has no miscarriage and stillbirth and that they have 4 healthy children. There was no known history of any major diseases or infections in the prenatal history, except dietary regulated gestational diabetes. The patient was on follow-up on physical examination and did not accompany dysmorphic findings. Although the baby was more prominent on the right foot and leg, there was bilateral edema extending towards the below-knee region on both feet (**Figure 1, 2**). When the family was questioned, it was learned that there were no family members with similar complaints. Complete routine blood count, biochemical parameters, and urine tests were normal in routine laboratory examinations and examinations for edema etiology. Albumin and protein values were normal. No pathological finding was detected in the chest x-ray taken due to respiratory distress. Respiratory distress resolved in a few hours. Transfontanelle and all abdominal ultrasonography were normal. Echocardiography was normal. Arterial and venous colored Doppler ultrasonographies of bilateral lower extremities were also evaluated as normal in the examinations for the etiology of edema.

Lower and upper extremity lymphoscintigraphy was performed on the third day of his admission. In the lymphoscintigraphy images, it was observed that the radiopharmaceutical that was given on both upper extremities passed to the lymph node of the axillary; minimal transition to the inguinal lymph node was observed in the left lower limb and no transition to inguinal lymph nodes in the right lower limb. Primary congenital lymphedema was considered in the patient due to the absence of a significant transition on both lower extremities, especially on the right (**Figure 3**).

Figure 1.

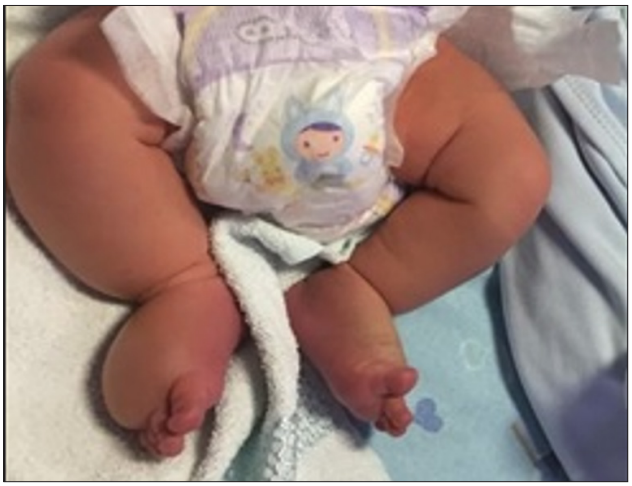

Figure 2.

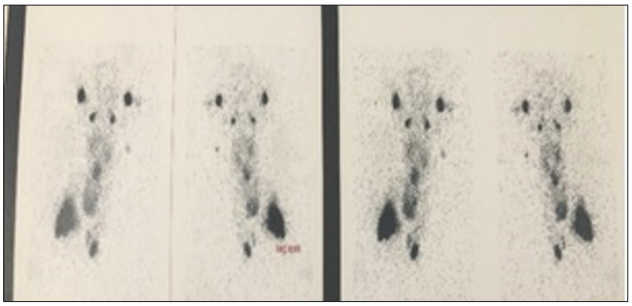

Figure 3.

DISCUSSION

Lymphedema is characterized by a generalized or regional accumulation of protein-rich interstitial fluid (7) as a result of impaired lymphatic circulation due to congenital or acquired reasons. It can occur either primary or secondary in two ways. While lymphedema may develop secondary to different acquired factors (infection, kidney failure), secondary to neoplasm, surgery, trauma, and radiation; most childhood cases are primary lymphedema (8-10). Primary lymphedema is seen at a rate of 1/100,000 (11). In primary lymphedema cases, there is an erroneous development of lymphatic structures (aplasia, hypoplasia, dilatation) or function, and most of them are thought to be a genetic predisposition. Typically, asymmetrical and bilateral lower extremity involvement occurs while swelling can be seen in all extremities. VEGFR-3 (FLT-4), the gene of this disease, is located in the region of 5q34-q35 of the

telomeric part of the 5q chromosome (12). It genetically shows an autosomal dominant transition. While it can be seen in both sexes, it is more common in girls (13). Primary lymphedema may also be associated with genetic diseases such as trisomy 13, 18, 21, primary congenital lymphedema, Turner syndrome, Noonan syndrome, lymphoedema dystiasis, or yellow nail syndrome (2, 3).

Most cases of lymphedema can be diagnosed by detailed history taking, physical examination, and ultrasonography. Lymphatic imaging is one of the examination methods used to confirm the diagnosis. Isotopic lymphoscintigraphy is considered the gold standard for the diagnosis of lymphedema since it is easy to use and minimally invasive, and is harmless to the lymphatic endothelium (14).

Treatment of primary lymphedema is conservative and generally successful in most patients. The main purpose of the treatment is to prevent the development of infections and complications related to edema. Elevation of the affected limb combined with skincare is sufficient for uncomplicated mild cases of lymphedema (15, 16). Over time, the subcutaneous tissue may harden due to protein accumulation and fibrosis in the extravascular area and hyperkeratosis may occur in the skin. The risk of cellulitis and lymphangitis increases with interruption of lymphatic drainage (17). Skincare reduces the risk of developing cellulitis and lymphangitis. In addition, compression bandages, manual lymphatic drainage (with special massage techniques), exercise, and strict diet (fluid and salt restriction) are other recommended treatment methods (18). In cases where medical treatment is unsuccessful, surgical intervention is among the treatment options (19).

Primary lymphedema was considered primarily due to lymphedema present from birth. Our patient was diagnosed with a clinical, physical examination, detailed history, and lymphoscintigraphy. The case is presented for a brief review of the primary lymphedema literature.

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 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

Korumasız Triküspid Kapak Orifisinin İki Yeni Olgusu

A New Two Cases of Unguarded Tricuspid Valve Orifice

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Öz

Korumasız triküspid kapak nadir görülen bir malformasyondur. Triküspid kapak ve subvalvüler aparatın tam veya kısmi agenezisi ile karakterizedir. Triküspid kapak yapılarının tamamen yokluğu genellikle pulmoner atrezi ve sağlam ventriküler septum ile ilişkilidir. Korumasız triküspid kapak orifis tanısı konulan iki vaka bildiriyoruz. Korumasız triküspid kapak orifisi literatürde nadir bildirildiği için bu olgular sunuldu.

Anahtar Kelimeler: Triküspid kapak, korumasız triküspid orifis, pulmoner atrezi

Abstract

Unguarded tricuspid valve orifice is a rare malformation. It is characterized by complete or partial agenesis of the tricuspid valvular and subvalvular apparatus. The complete absence of tricuspid valve structures is often associated with pulmonary atresia and intact ventricular septum. We report on two cases diagnosed as unguarded tricuspid valve orifice. These cases were presented because of unguarded tricuspid valve orifice is reported rare in the literature

Keywords: Tricuspid valve: unguarded tricuspid orifice, pulmonary atresia

GİRİŞ

Korumasız "Unguarded" triküspid kapak nadir bir kardiyak malformasyondur (1,2). Triküspid kapak ve subvalvüler aparatın tam veya kısmi agenezisi ile karakterizedir (2). Sağ atriyal ve sağ ventrikül genişlemesi ile sonuçlanan şiddetli bir triküspid kapak yetersizliği vardır. Bu lezyona genellikle pulmoner atrezi eşlik eder (2,3). Pulmoner atrezinin pulmoner kapak boyunca anterograd pulmoner kan akışının olmamasından kaynaklandığı düşünülmektedir (1,2). Hastalar siyanozdan konjestif kalp yetmezliğine kadar çeşitli semptomlara sahip olabilir (4).

OLGU 1

41. gebelik haftasında 3445 gr doğan ve doğumdan hemen sonra santral siyanoz fark edilen ve oksijen saturasyonları oksijensiz %75-85 arasında olan yenidoğan hastanın fizik muayenesinde 2/6 sistolik ejeksiyon üfürümü, gallop ritmi mevcuttu. Karın muayenesinde karaciğer kot altı 2 cm palpe edildi. Kan basıncı 70/50 mm/Hg, nabız sayısı 180 atım/dk, solunum sayısı 50/dk idi. Ekokardiyografik incelemede hipoplazik sağ ventrikül, unguarded triküspid kapak orifisi, triküspid orifis genişliği 12 mm, 2. derece triküspid yetmezliği, fonksiyonel pulmoner atrezi, hipoplazik pulmoner arter yatağı, patent duktus arteriozus (PDA) bağımlı pulmoner dolaşım, sağdan sola şantlı 5 mm boyutunda sekundum atriyal septal defekt (ASD) saptandı (**Resim 1**). Ekokardiyografide renkli doppler görüntülemede sağ ventrikülden sağ atriyuma ters akış kan akımı görüldü (**Resim 2**). Ekokardiyografi bulguları korumasız triküspid

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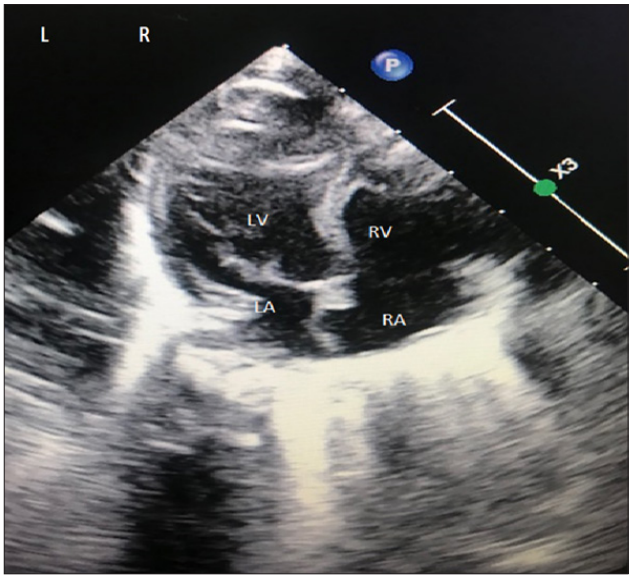
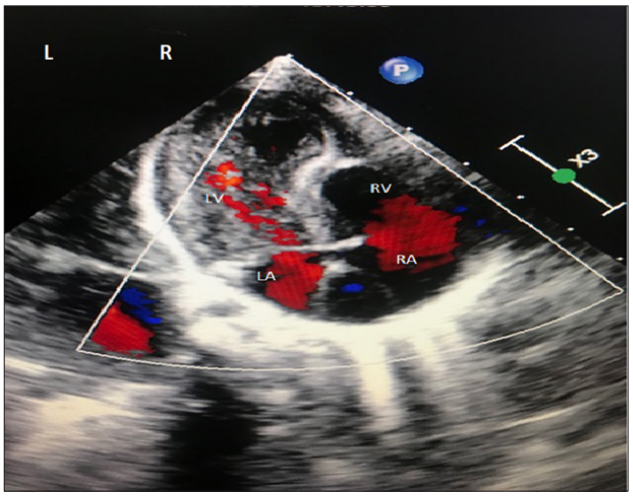
kapak orifisi ile uyumlu idi. Duktus arteriozusu açıklığını sağlamak ve pulmoner kan akımını arttırmak için prostaglandin E1 infüzyonu başlandı. Pulmoner arter ve dallarının net görüntülenmesi için kardiyak bilgisayarlı tomografi istendi. İncelemesinde sağ ventrikül hipoplazisi, ASD, PDA görüldü. Pulmoner arter çapları hipoplazik idi. Kalp damar cerrahisi tarafından modifiye Blalock Taussig (BT) şant ameliyatı yapıldı. Postoperatif takiplerinde beslenmesi iyi, oksijen ihtiyacı olmayan hasta operasyondan 14 gün sonra taburcu edildi. Kontrol seri ekokardiyografi incelemelerinde mevcut ekokardiyografi bulguları ile modifiye BT şant akımı görüldü. Poliklinik kontrolünde saturasyonları %90-92 civarında idi. Klinik olarak iyi olan hasta belirli aralıklarla kontrollere gelmektedir.



Resim 1. Ekokardiyografide apikal dört boşluk görüntüde: Sağ atriyumda genişleme, triküspid kapak, korda tendinealar ve papiller kasların olmadığı görülmektedir. LV: Sol ventrikül, LA: Sol atriyum, RA: Sağ atriyum, RV: Sağ ventrikül.



Resim 2. Ekokardiyografide renkli Doppler görüntülemede sağ ventrikülden sağ atriyuma geriye doğru kan akımı görülmektedir. LV: Sol ventrikül, LA: Sol atriyum, RA: Sağ atriyum, RV: Sağ ventrikül

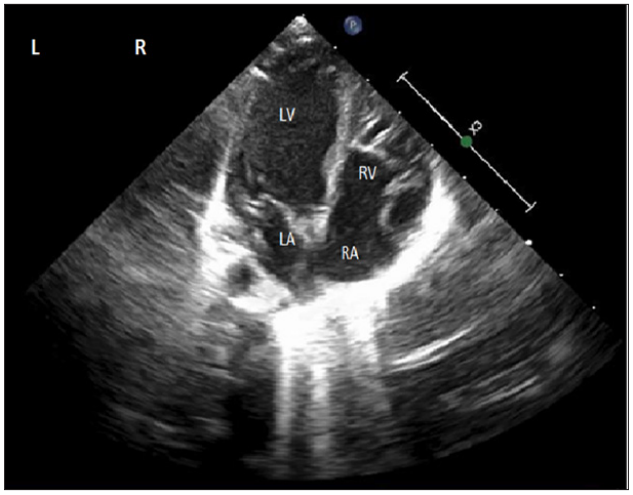
OLGU 2

34 yaşındaki gravide 4 parite 4 annenin 37+6 haftalık 3675 gram doğan ve doğumdan sonra oksijen saturasyonları oksijen desteği olmadan %75-85 arası olan hasta yenidoğan yoğun bakıma yatırıldı. Kan basıncı 70/50 mm/Hg, nabız sayısı 165 atım/dk, solunum sayısı 46/dk idi. Hastanın fizik muayenesinde triküspid odakta 3/6 şiddetinde pansistolik üfürüm vardı. Ekokardiyografide sağ ventrikül hafif hipoplazik, aortadan pulmoner arter içine doğru 2 mm genişliğinde restriktif PDA, pulmoner artere anterograd akım olduğu, 3 mm genişliğinde soldan sağa şantlı müsküler trabeküler ventriküler septal defekt (VSD) izlendi ve VSD üzerinde belirgin gradient alınmadı. Sağ ventrikül kavitesi 14 mm x18 mm (z skoru 1.3), sol ventrikül kavitesi 20mm x16 mm ölçüldü, triküspid kapak displastik, triküspid orifisi subapparatus dokusu iyi gelişmemiş, triküspid kapağın iki adelesi kalın aksesuar dokusu sağ ventrikül çıkış yolu altına doğru uzanıyordu,1 derece triküspid yetmezlik saptandı (**Resim 3,4**). Ekokardiyografi bulguları parsiyel korumasız triküspid kapak orifisi ile uyumlu idi. Hastaya duktus arteriozusun açıklığını sağlamak için prostaglandin E1 infüzyonu başlandı. Kardiyovasküler cerrahi tarafından sol modifiye BT şant ameliyatı yapıldı. Postoperatif 1. gün oksijen saturasyonu entübe iken %89 idi. Operasyondan 12 gün sonra beslenmesi iyi, vitalleri stabil ve oksijen ihtiyacı olmayan hasta taburcu edildi. Kontrol seri ekokardiyografi incelemelerinde mevcut ekokardiyografi bulguları ile modifiye BT şant akımı görüldü.

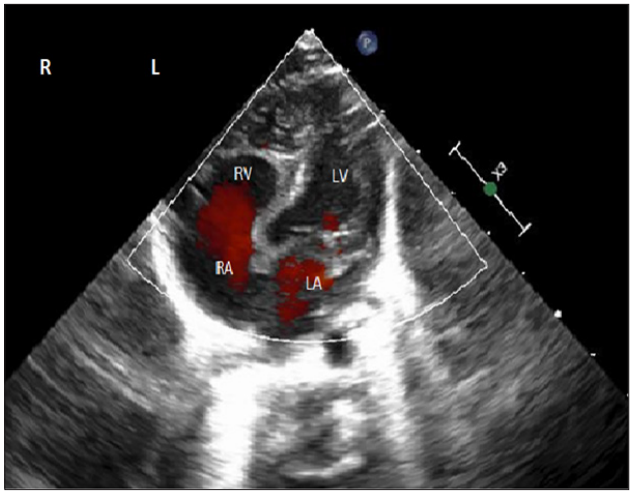
TARTIŞMA

Triküspid kapak displazisi, izole triküspid kapak yetersizliğinin en yaygın nedenidir. Triküspid kapak displaziler içinde korumasız triküspid kapak en nadir görünen formudur (3). Fetal ekokardiyografide kısmi veya tam triküspid kapak dokusu yokluğu 1964'te Kanjuh ve arkadaşları tarafından ''unguarded triküspid kapak'' olarak adlandırılmıştır (1). Normalde triküspid kapak sağ atriyoventriküler kapak olup fibröz anülüse bağlı üç yaprakçıktan (ön, arka ve septal) oluşmaktadır. Bu yaprakçıklar üç papiller kas ile desteklenmektedir (5). Korumasız triküspid kapak ayırıcı tanısında, Ebstein anomalisi, Uhl anomalisi ve aritmojenik sağ ventrikül displazisi yer almaktadır (3).

Ebstein anomalisinde septal ve arka yaprağının atriyoventriküler bileşke yerine aşağı (apikal) sağ ventrikül içine doğru yer değiştirmesi görülmektedir. Bunun sonucu olarak septal yaprakçık aşağı yerleşir ve displastiktir (6). Ebstein anomalisinde triküspid kapak hasarının şiddeti ve ek anomalilere bağlı klinik belirti ve başlama yaşı değişmektedir. Çoğu hasta bebeklik veya çocukluk döneminde teşhis edilmesine rağmen, ilk tanı yetişkinlikte de ortaya çıkabilir (7).



Resim 3. Ekokardiyografide apikal dört boşluk görüntüde triküspid kapak broşürleri, korda tendineler ve papiller kasların iyi gelişmediği görülmektedir. LV: Sol ventrikül, LA: Sol atriyum, RA: Sağ atriyum, RV: Sağ ventrikül.



Resim 4. Ekokardiyografide renkli Doppler görüntülemede sağ ventrikülden sağ atriyuma geriye doğru kan akımı görülmektedir. LV: Sol ventrikül, LA: Sol atriyum, RA: Sağ atriyum, RV: Sağ ventrikül

Uhl anomalisinde triküspid ve pulmoner kapakların anatomisi normaldir. Sağ ventrikül miyokardı kağıt gibi oldukça incedir, sağ ventrikül duvarı neredeyse endokard ve epikarddan oluşur (8).

Aritmojenik sağ ventriküler displazisi olarak da bilinen aritmojenik sağ ventriküler kardiyomiyopatisinde ventriküler aritmiler, sağ ventrikül disfonksiyonu ve ventriküler dilatasyon görülür. Miyokard dokusu yerine fibröz-yağ dokusu yer almaktadır. Sağ ventrikül miyokardında atrofi, fibroz-yağ infiltrasyonu ve fibrozis ile karakterize olan bir hastalıktır. Triküspid kapak normal görünümdedir. Kalp yetmezliğine neden olabilen ilerleyici bir hastalıktır. Aritmojenik sağ ventriküler kardiyomiyopati hastaları, ani kardiyak ölüm açısından yüksek risk altındadır (9).

Korumasız triküspid kapakta septal tüberküller, korda tendinalar ve papiller kasları yoktur. Triküspid kapağın yokluğu ve ilişkili pulmoner atrezi nedeniyle, çoğu zaman triküspid açıklık boyunca kanın ileri ve geri akımı vardır (1,2). EKG'de P pulmonale, sağ ventrikül hipertrofisi, sıklıkla atriyal fibrilasyon ya da sağ dal bloku örneği görülebilir. Telekardiyografide primer triküspid yetmezlikte; sağ atriyal genişleme, sağ ventriküler genişleme ve süperior vena kavada belirginleşme görülür (10). Klinik bulgular genellikle siyanoz ve/veya konjestif kalp yetmezliği ile erken çocukluk döneminde ortaya çıkar. Sağ ventrikül yetmezliği ve triküspid yetersizliği ile yetişkin yaşamı boyunca dekompanse olan az sayıda hasta vardır. Bu hastaların bir kısmı atriyal fibrilasyon ile gelebilir (11,12,13).

Korumasız triküspid kapak tanısı fetal ekokardiyografi ile tanısı konulabilir. Vikraman ve ark. sunduğu olguda 26 yaşında gravida 2 annenin, 2. trimesterde antenatal dönemde ultrasonografik incelemede rastlantısal fark edilen fetal sağ atriyumda genişleme görülmüş ve ileri inceleme için bir üst merkeze sevk edilen hastanın yapılan fetal ekokardiyografi ve renkli doppler incelemesinde korumasız triküspid kapak tanısı konulmuştur (14). Multidisipliner ekip yaklaşımı prenatal tanı ve postnatal hasta takip ve tedavisinde başarının artmasına önemli katkı sağlayacaktır.

SONUÇ

Triküspid orifisin tamamen yaprak dokusundan yoksun olduğu korumasız triküspid kapak lezyonu nadir görülen bir kapak anomalisi olması nedeniyle sunulmuştur. Yenidoğanda santral siyanozuna neden olabilen nadir bir konjenital kalp hastalığıdır. Triküspid kapak yetmezliği ve fonksiyonel pulmoner atrezi olan siyanotik yenidoğan bebeklerde bu hastalık akla getirilmelidir. Duktus bağımlı konjenital kalp hastalığı bulguları olacağı için korumasız triküspid kapak tanısının erken konulması ile tedavi seçenekleri belirlenebilir. Bu yazıda sunulan iki olgu da erken tanı sayesinde başarılı olarak tedavi edilmiştir.

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

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

Meckel's Diverticulitis Causing Small Bowel Intussusception: A Case Report

Meckel Divertikülitine Bağli İnce Bağırsak İntususepsiyonu: Olgu Sunumu

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ABSTRACT

Meckel's diverticulum (MD) is considered the most common congenital gastrointestinal malformation affecting 2% of the population. Complications including hemorrhage, diverticulitis, and intestinal obstruction occur in only 4% of patients with MD. This article presents the imaging findings of a small bowel intussusception caused by Meckel's diverticulitis in a 10-year-old girl with acute abdominal pain.

Keywords: Meckel's diverticulum, Meckel's diverticulitis, intussusception, intestinal obstruction.

ÖZ

Meckel divertikülü (MD), popülasyonun %2'sinde görülen en yaygın doğumsal gastrointestinal anomalisidir. MD hastalarının sadece %4'ünde kanama, divertikülit ve intestinal obstrüksiyon gelişir. Bu makale, akut karın ağrısı olan 10 yaşındaki kız olguda Meckel divertikülitine bağlı gelişen ince bağırsak intususepsiyonunun görüntüleme bulguları sunulmuştur.

Anahtar Kelimeler: Meckel divertikülü, Meckel divertiküliti, intususepsiyon, intestinal obstrüksiyon

INTRODUCTION

Meckel's diverticulum (MD) represents the most common gastrointestinal congenital malformation (1). The most frequent complications of MD are gastrointestinal hemorrhage, diverticulitis, and intestinal obstruction (1). To the best of our knowledge, the co-occurrence of Meckel's diverticulitis and small bowel obstruction in children have been reported only twice in English literature (2, 3).

This report shows the imaging findings of a small bowel intussusception caused by Meckel's diverticulitis in a 10-year-old girl with acute abdominal pain.

CASE REPORT

A ten-year-old girl who had undergone an appendectomy for acute appendicitis six months earlier, presented to the hospital complaining of abdominal pain and vomiting for the past five days.

Her abdomen was distended and she had a tenderness over the periumbilical region. The laboratory investigations showed mild anemia with an increased C-reactive protein value and white blood cell count. The urine analysis was normal. There were no significant findings in the chest X-ray. Abdominal radiographic imaging revealed no signs of pneumoperitoneum or dilated small-bowel loops with air-fluid levels.

The ultrasonographic (US) image demonstrated a target sign characteristic of intussusception caused by a blind-ended segment of a thick-walled bowel (**Figure 1A**). Surrounding soft tissue hyperechogenicity, reactive lymph nodes, and free intra-abdominal fluid was suggestive of associated inflammation.

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The contrast-enhanced abdominal CT images showed a fluid-filled, blind-ending intestinal structure with mural enhancement (**Figure 1B**).

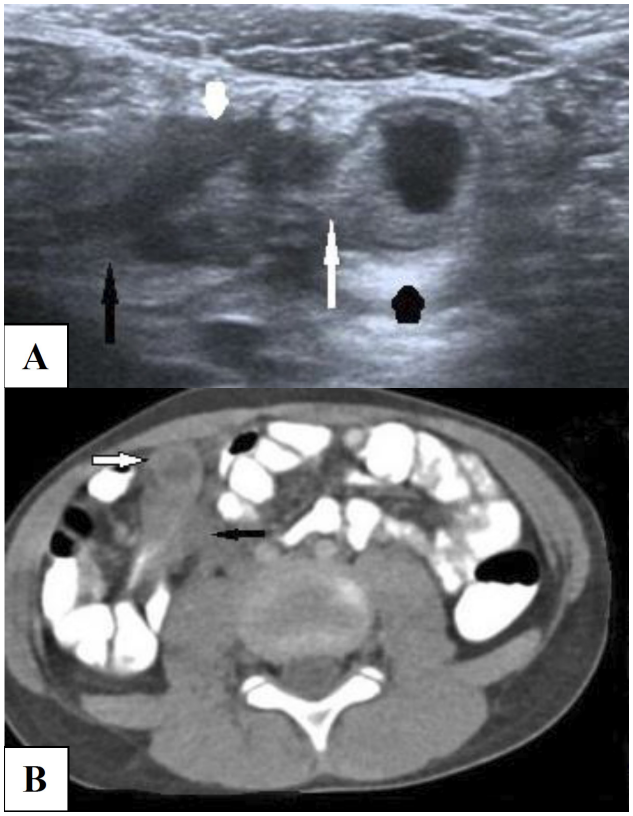

Figure 1. A. An ultrasound view shows the appearance of a blind-ending and thick-walled bowel segment (white arrow) in continuity with the terminal ileum (black arrow) surrounded by soft tissue hyperechogenicity (black arrowhead) and free fluid (white arrowhead). **B.** Contrast-enhanced abdominal CT axial image shows a blind-ending intestinal structure (white arrow) with reactive lymph nodes (black arrow).

The intraoperative findings revealed an ileo-ileal intussusception due to inflamed MD located 15 cm from the ileocecal valve. A segmental small bowel and MD resections with consecutive anastomosis were performed. Histopathology confirmed the diagnosis of Meckel's diverticulitis containing heterotopic gastric mucosa and inflammatory cells around the diverticulum. The postoperative period was uneventful, and the patient was discharged 6 days after the surgery. Informed consent was signed by the patient's father.

DISCUSSION

Meckel's diverticulum (MD) occurs as a result of incomplete obliteration of the omphalomesenteric duct (1). MD is generally recognized by the 'rule of twos' affecting 2% of the population, located two feet (60 cm) from the ileocecal valve, usually two inches (5 cm) long, and may contain gastric or pancreatic ectopic tissues (4).

MD is often asymptomatic; however 15% of patients more than four years old, 77% of patients aged one month to two years, and 85% of neonates develop symptoms due to complicated MD (2). The most common complications of MD are gastrointestinal bleeding, intestinal obstruction, and diverticulitis. Neoplasm is rare. Gastrointestinal bleeding is the most frequently encountered complication of MD in children (5,6). It occurs due to the acid secretion from the ectopic gastric tissue leading to peptic ulceration of the ileal mucosa (6). Meckel's diverticulitis occurs more frequently in adult patients (1). Complicated diverticulitis may be associated with perforation, formation of abscess, fistula, or bowel obstruction due to inflammatory adhesions (1-3). MD may act as a lead point causing ileo-vitelline, ileo-ileal, and ileocolic types of intussusceptions (6). MD tumors are very rare (1,2).

Radiologic diagnosis of complicated MD can be difficult, especially when the diagnosis is not initially suspected (6). One systematic review reported that 40% of fifty patients with MD had a preoperative diagnosis of acute appendicitis (6). Ultrasonography (US) should be the first step imaging modality for evaluation of the complicated MD in children. Inflamed MD is usually presented as a cystlike structure in the periumbilical region or right lower quadrant. A sonographic study by Daneman et al. reported that inflamed MD has a thicker and more irregular wall with increased vascularisation than that found in an enteric duplication cyst (7).

An intravenous contrast-enhanced abdominal CT is recommended in cases of high clinical suspicion and negative findings on US. Although it is difficult to determine the MD on CT, a blind-ending intestinal structure with mural enhancement and surrounding fat stranding are highly suggestive for Meckel's diverticulitis (8). A water-soluble oral contrast-enhanced abdominal CT facilitates the diagnosis of an abscess, fistula, or perforation due to Meckel's diverticulitis. Oral contrast media should be avoided in cases of acute gastrointestinal hemorrhage as it may obscure the active contrast extravasation from the site of bleeding (8). Contrast material accumulation into the blind-ending digestive structure connected to the ileum suggests an active bleeding MD (6,8).

Radionuclide studies based on Technetium-99m pertechnetate administration help identify the ectopic gastric mucosa in the MD. A review considering children with MD reported that the complicated MD was 2.3 times more likely to be diagnosed with CT than with scintigraphy (8).

To the author's knowledge, only two cases regarding the co-occurrence of Meckel's diverticulitis and small bowel obstruction in children have been published to date. The CT images of a Meckel's diverticulitis causing small bowel obstruction were first described by Shelat et al. (2). Kumar

et al. reported the abdominal X-ray findings of intestinal obstruction caused by inflamed MD (3). This report shows the images of three non-invasive radiological techniques of the small bowel intussusception caused by Meckel's diverticulitis in a child.

CONCLUSION

The present case suggests that MD should be considered as a possible cause for intussusception in children above two years of age. An early diagnosis is essential to ensure favorable outcomes in children with MD.

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 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

Acinetobacter lwoffii Septicemia in a Newborn

Yenidoğan Bir Bebekte *Acinetobacter lwoffii* sepsisi

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ABSTRACT

Acinetobacter lwoffii is gram-negative coccobacillus which is seen as a normal flora of the oropharynx and skin of the healthy individuals. It is a potential opportunistic pathogen in patients with impaired immune systems, and can cause of healthcare associated infections like septicemia. *A. lwoffii* infection is increasing particularly in premature and very low-birth weight neonates. Here, we present a case of a low birth weight neonate with *A. lwoffii* infection who was successfully treated.

Keywords: *Acinetobacter lwoffii*, septicemia, NICU

ÖZ

Acinetobacter lwoffii sağlıklı bireylerin cilt ve orofarenksinde flora üyesi olarak saptanabilen bir gram-negatif kokobasildir. İmmün baskılanmış kişilerde fırsatçı patojen olarak septisemi gibi sağlık bakımı ilişkili enfeksiyonlara sebep olabilir. Prematüre ve düşük doğum ağırlıklı bebeklerde *A. lwoffii* enfeksiyon sıklığı artmaktadır. Bu vaka sunumunda, düşük doğum ağırlıklı bir bebekte başarı ile tedavi edilmiş *A. lwoffii* enfeksiyonunu sunmak istiyoruz.

Anahtar kelimeler: *Acinetobacter lwoffii*, sepsis, yenidoğan yoğun bakım ünitesi

INTRODUCTION

Acinetobacter is an aerobic, non-fermentative, immotile, gram negative bacillus, which is widespread in nature. It has been identified as a cause of healthcare associated infections, especially in the colonization and infection in immunocompromised patients as an opportunistic pathogen (1,2). However, a limited number of cases of infection caused by *Acinetobacter lwoffii* has been reported, most of them are central intravascular catheter-related blood stream infections or bacteremia. On the other hand, community-acquired infections associated with *Acinetobacter lwoffii* such as pneumonia, meningitis, urinary tract infection, skin and wound infection and acute gastroenteritis were also reported (3). This report presents the case of a low birth weight neonate with multidrug-resistant *A. lwoffii* infection.

CASE REPORT

A male neonate was born at 26 week because of maternal preeclampsia. The mother had a history of chorioamnionitis. He was intubated due to respiratory failure. Ampicillin, amikacin treatment and fluconazole prophylaxis were started due to suspicion of early onset septicemia. Umbilical artery and vein catheters were inserted at the first day of life. The blood cultures, taken at the admission to neonatal intensive care unit (NICU), were negative. Ligation of ductus arteriosus was performed at seventh day of life. After surgery his vital signs deteriorated and abdominal distension developed. Sepsis was considered due to gastric residues, abdominal distention, and prolongation of capillary filling. Because of worsening clinical status, laboratory tests and blood cultures were repeated. The laboratory data were as

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follows: WBC 32,200/mm³, neutrophils 24,700 (76%); lymphocytes 1,300 (4%); monocyte 5900 (18%); hemoglobin 11 g/dL; platelets 55,000/mm³; mm/h; C-reactive protein 84 mg/dL; procalcitonin was 4.1 ng/mL. Metabolic acidosis was detected. Lumbar puncture was not performed, because of poor general condition and thrombocytopenia. Air around the intestinal wall was detected on abdominal radiography. He underwent surgery for perforated necrotizing enterocolitis. Broad spectrum antibiotics (meropenem, teicoplanin and fluconazole) were started immediately for suspicion of healthcare associated infection. Central vascular catheter was removed. The result of the blood culture was reported as *A. Iwoffii*. The microorganism was resistant to penicillin G, cefotaxime, ceftazidime, gentamicin, amikacin, cefepime and sensitive to meropenem (Minimal inhibitor concentration values: <0.5). Five days later his clinical findings improved, and inflammation markers returned to normal levels. Control blood culture was negative and meropenem treatment was completed to two weeks.

DISCUSSION

Acinetobacter Iwoffii, is an aerobic and gram-negative bacillus and recognized as normal flora of the skin, oropharynx, and perineum of healthy individuals. It has been previously reported that *A. Iwoffii* is found in environmental sources, particularly on the hands of nursing staff. It was associated with healthcare associated infections, particularly in immunocompromised hosts (4). Most of cases of *A. Iwoffii* bacteraemia are associated with central vascular catheter (2).

Factors such as longer stay in hospital, intensive care unit, and burn units, major surgical procedures, neutropenia, underlying chronic diseases, and previous antibiotic use are predisposing factors for *Acinetobacter* infections (5). Prematurity is thought to be associated with disseminated disease because of the immaturity of the immune system (6). Our patient was premature and extremely low birth weight infant. He was undergone ductal ligation and perforated necrotizing enterocolitis repair. He also had umbilical catheter. All these factors contributed to the development of *A. Iwoffii* septicemia.

The infections due to *Acinetobacter* spp. are often extremely difficult for clinicians to treat, because of rapid antimicrobial resistance development to all currently available antimicrobial agents like aminoglycosides, fluoroquinolones, ureidopenicillins and third generation cephalosporins (7). Carbapenems are the most effective antibiotics against these agents, while colistin can be used against carbapenem resistant *Acinetobacter* spp. on rare occasion (8).

Increasing rates of *Acinetobacter* spp are reported in NICUs. Judicious and timely antibiotic treatment is one of the important keys in controlling multi-drug resistant *Acinetobacter* infections (9). In the selection of empirical antibiotic treatment for healthcare associated neonatal sepsis cases, the resistance status of each unit should be considered.

ETHICAL DECLARATIONS

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

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