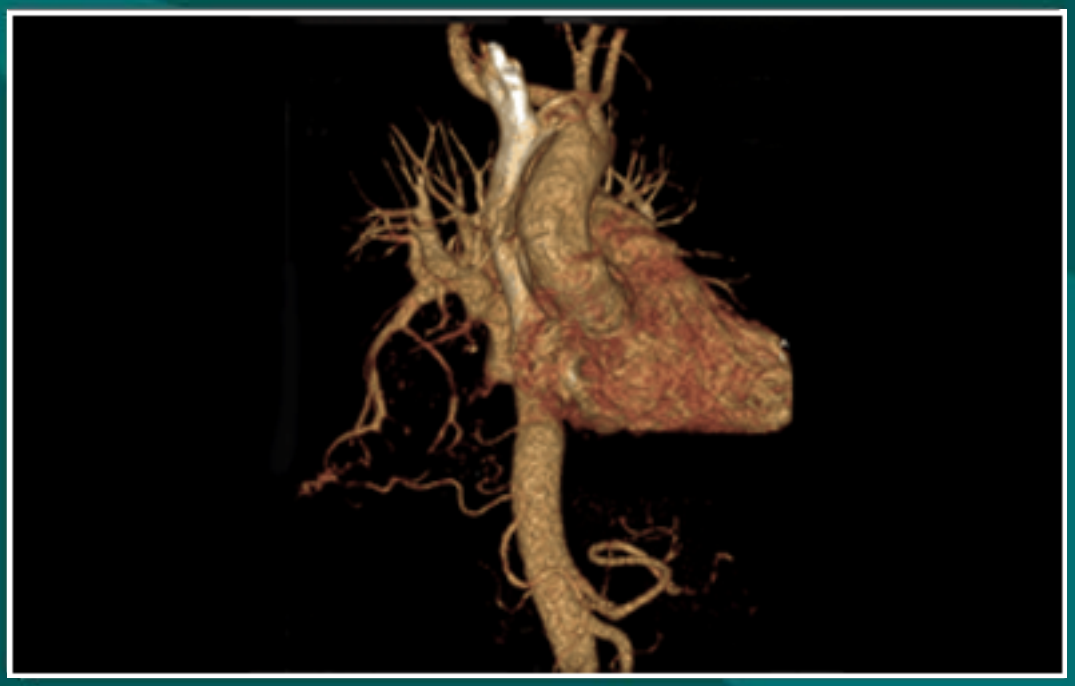
February 2017; Volume 8; Number 2

IJCRI

www.ijcasereportsandimages.com

International Journal of

CASE REPORTS AND IMAGES



Online submission at:
www.ijcasereportandimages.com/submit

ISSN: 0976-3198

International Journal of Case Reports and Images

EDITORIAL BOARD

International Journal of Case Reports and Images (IJCRI)

Dr. Achuta Kumar Guddati	USA	Dr. Hua Zhong	USA
Dr. Aditya Gupta	USA	Dr. Ho-Sheng Lin	USA
Dr. Adria Arboix	Spain	Dr. Imtiaz Wani	India
Dr. Adriana Handra-Luca	France	Dr. James Cheng-Yi Lin	Taiwan
Dr. Afra Hadjizadeh	Iran	Dr. Jonathan D. Solomon	USA
Dr. Ahmed El Said Lasheen	Egypt	Dr. Kyuzi Kamoi	Japan
Dr. Ali Soltani	USA	Dr. Luca Bertolaccini	Italy
Dr. Altacilio Aparecido Nunes	Brazil	Dr. Makoto Adachi	USA
Dr. Amin F Majdalawieh	UAE	Dr. Mehmet Uludag	Turkey
Dr. Athanassios Tsakris	Greece	Dr. Mehmet Inal	Turkey
Dr. Antonio La Cava	USA	Dr. Mohamed Radhi	USA
Dr. Asher Bashiri	Israel	Dr. Mohannad Al-Qudah	Jordan
Dr. Aziz Mustafa	Kosovo	Dr. Morikuni Tobita	USA
Dr. Christopher CK Ho	Malasiya	Dr. Naila Khalil	USA
Dr. Claudio Feliciani	Italy	Dr. Natalya Semiletova	USA
Dr. Daniela Cabibi	Italy	Dr. Oner Dikensoy	Turkey
Dr. Deepa Rastogi	USA	Dr. Ozlem Guneysel	Turkey
Dr. Deepak Sharma	USA	Dr. Paolo Cardelli	Italy
Dr. Emre Karaşahin	Turkey	Dr. Paul Rea	UK
Dr. Federico Bizzarri	Italy	Dr. Parijat Saurav Joy	USA
Dr. Gavin A. Falk	USA	Dr. Petru Matusz	Romania
Dr. Gerardo Gómez-Moreno	Spain	Dr. Pengcheng Luo	China
Dr. Gil Atzmon	USA	Dr. Piaray Lal Kariholu	India
Dr. Giovanni Leuzzi	Italy	Dr. Piraye Kervancioglu	Turkey
Dr. Giovanni Tuccari	Italy	Dr. Radhika Muzumdar	USA
Dr. Gokulakkrishna Subhas	USA	Dr. Rajesh Pareta	USA
Dr. Guo Wei	USA	Dr. Ranjan Agrawal	India
Dr. Hajimi Orita	Japan	Dr. Ranji Cui	China

Contact Details:

Editorial Office

Email: meditor@ijcasereportsandimages.com
Fax: +1-773-409-5040
Website: www.ijcasereportsandimages.com

Guidelines for Authors

Full instructions are available online at:
www.ijcasereportsandimages.com/submit/instructions-for-authors

Manuscript submission:
www.ijcasereportsandimages.com/submit

Disclaimer

Neither International Journal of Case Reports and Images (IJCRI) nor its editors, publishers, owners or anyone else involved in creating, producing or delivering International Journal of Case Reports and Images (IJCRI) or the materials contained therein, assumes any liability or responsibility for the accuracy, completeness, or usefulness of any information provided in International Journal of Case Reports and Images (IJCRI), nor shall they be liable for any direct, indirect, incidental, special, consequential or punitive damages arising out of the use of International Journal of Case Reports and Images (IJCRI) or its contents. While the advice and information in this journal are believed to be true and accurate on the date of its publication, neither the editors, publisher, owners nor the authors can accept any legal responsibility for any errors or omissions that may be made or for the results obtained from the use of such material. The editors, publisher or owners, make no warranty, express or implied, with respect to the material contained herein. (http://www.ijcasereportsandimages.com/disclaimer.php)

International Journal of Case Reports and Images

EDITORIAL BOARD

International Journal of Case Reports and Images (IJCRI)

Dr. Ricardo Correa	USA
Dr. Ricardo S. Macarenco	Brazil
Dr. Sanju George	UK
Dr. Saurabh Khakharia	USA
Dr. Sergio Gabriel Susmallian	Israel
Dr. Shashideep Singhal	USA
Dr. Shervin Assari	USA
Dr. Shilpa Jain	USA
Dr. Siddharth Mathur	USA
Dr. Sirinrath Sirivisoot	USA
Dr. Slobodan Marinkovic	Slovenia
Dr. Stefan Hagmann	USA
Dr. Stefano Romagnoli	Italy
Dr. Tapas Saha	USA
Dr. Teguh Haryo Sasongko	Malaysia
Dr. Tomoyuki Yano	Japan
Dr. Tun Hing Lui	China
Dr. Yulin Li	China

Contact Details:

Editorial Office

Email: meditor@ijcasereportsandimages.com
Fax: +1-773-409-5040
Website: www.ijcasereportsandimages.com

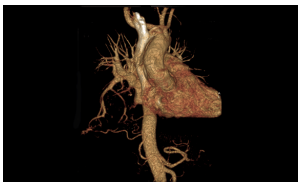
Guidelines for Authors
Full instructions are available online at:
www.ijcasereportsandimages.com/submit/instructions-for-authors

Manuscript submission:
www.ijcasereportsandimages.com/submit

Disclaimer
Neither International Journal of Case Reports and Images (IJCRI) nor its editors, publishers, owners or anyone else involved in creating, producing or delivering International Journal of Case Reports and Images (IJCRI) or the materials contained therein, assumes any liability or responsibility for the accuracy, completeness, or usefulness of any information provided in International Journal of Case Reports and Images (IJCRI), nor shall they be liable for any direct, indirect, incidental, special, consequential or punitive damages arising out of the use of International Journal of Case Reports and Images (IJCRI) or its contents. While the advice and information in this journal are believed to be true and accurate on the date of its publication, neither the editors, publisher, owners nor the authors can accept any legal responsibility for any errors or omissions that may be made or for the results obtained from the use of such material. The editors, publisher or owners, make no warranty, express or implied, with respect to the material contained herein. (http://www.ijcasereportsandimages.com/disclaimer.php)

International Journal of
Case Reports and Images

Cover Figure:



All Articles:

Open Access

Contents

Vol. 8, No. 2 (February 2017)

Cover Image

Computed tomography angiography (volume rendering) show the arterial blood supply of pulmonary sequestration from both right pulmonary artery and multiple branches from aorta.

Case Report

Clinical Images

Letters to the Editor

SUBMISSION INSTRUCTIONS
All manuscripts, including illustrations, should be submitted at: www.ijcasereportsandimages.com/submit or email to: submit@ijcasereportsandimages.com
Author Guidelines: www.ijcasereportsandimages.com/submit/instructions-for-authors.php
For any questions contact the Editorial Office via e-mail at: info@ijcasereportsandimages.com or Fax: 1-773-409-5040

CASE REPORT PEER REVIEWED | OPEN ACCESS

The idiopathic avascular osteonecrosis of the third metacarpal head (M. Mauclaire/Dieterich's disease)

Ingo Schmidt

ABSTRACT

Introduction: M. Mauclaire/Dieterich's disease is an uncommon condition mostly affecting the third metacarpal head, and can lead to secondary osteoarthritis of the metacarpophalangeal joint. For this condition, the total joint replacement is inevitable. Case Report: We report a 64-year-old male with a history of increasing pain in his third metacarpophalangeal joint right over a period of 10 years. Radiographically, there was a severe osteonecrosis of the third metacarpal head that was accompanied with pronounced osteoarthritis of the metacarpophalangeal joint. The patient was treated successfully with an unconstrained resurfacing total joint replacement. Conclusion: The preservation of motion in the metacarpophalangeal joints II–V has a top priority. A stable and functioning metacarpophalangeal joint is the key for satisfactory function of the overall finger. If a avascular osteonecrosis of a metacarpal head is not accompanied with secondary osteoarthritis, other joints preserving procedures are the methods of choice. In case of secondary osteoarthritis, the total joint replacement is inevitable. Resection arthroplasty or arthrodesis should be avoided as primary surgical procedure, and are to be considered as salvage options after a failed total joint replacement.

Ingo Schmidt

Affiliation: SRH Poliklinik Gera GmbH, Straße des Friedens 122, 07548 Gera (Germany).

Corresponding Author: Ingo Schmidt, Dr. Med, SRH Poliklinik Gera GmbH, Straße des Friedens 122, 07548 Gera (Germany); E-mail: schmidtingo62@googlemail.com

Received: 03 October 2016
-Accepted: 03 November 2016
Published: 01 February 2016

Keywords: Metacarpophalangeal joint, Idiopathic avascular osteonecrosis, Joint replacement

How to cite this article

Schmidt I. The idiopathic avascular osteonecrosis of the third metacarpal head (M. Mauclaire/Dieterich's disease). Int J Case Rep Images 2017;8(2):92–95.

Article ID: Z01201702CR10753IS

****

doi:10.5348/ijcri-201714-CR-10753

INTRODUCTION

The avascular osteonecrosis of the metacarpal head is a rare juvenile/adolescent lesion of the hand, and was first described in 1927 by Mauclaire [1]. Dieterich published in 1932 first results of eight treated patients and suggested that there is a strong correlation for manifestation of the third metacarpal head in young females [2]. These observations have been found its confirmation in all subsequent publications. Usually, the symptoms occur in childhood and adolescence, followed by patients in the middle decades of life, elderly patients from the 5th decade of life are tend to be underrepresented, it also may be present in the 1st, 2nd, 4th and 5th metacarpal, and bilateral occurrence was observed as well [3–11].

CASE REPORT

At presentation, 64-year-old male reported increasing pain in his metacarpophalangeal joint III right over a

period of 10 years. There was no history of any additional trauma. Professionally, he has been worked as a miner with jackhammers, and also he was a passionate boxer over a period of 20 years. The fist conclusion was incomplete. The extension of the third finger showed a deficit of 20 degrees to neutral, the flexion was limited to 60 degree. Radiographically, a severe osteonecrosis of the third metacarpal head was present that was accompanied with secondary osteoarthritis of the metacarpophalangeal joint (Figure 1A). The resurfacing joint replacement using the unconstrained SR™ MCP implant (formerly Avanta SR, Small Bone Innovations, Morrisville, PA, USA) with it uncemented cobalt-chrome (CoCr) alloy metacarpal hemispherical head that articulates against the cemented ultra-high molecular weight polyethylene (UHMWPE) phalangeal component was performed through a dorsal incision (Figure 1b-c).

Radiographically follow-up, at the fourth year showed that there was unchanged a correct positioning of the implant without any signs of loosening nor subsidence (Figure 2a). Fist conlusion and long finger extension were completely restored (Figure 2b-c). Grip strength (Jamar dynamometer) improved from 6–13 kp, and pain improved from 8 points to 0 points in visual analogue score (0–10 points). The patient reported that he would have the same procedure again if it necessary.

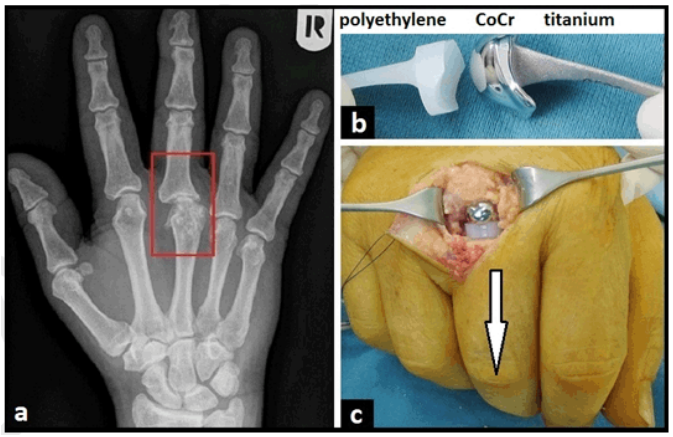


Figure 1: (a) Posteroanterior radiograph showing avascular osteonecrosis of the third metacarpal head accompanied with pronounced metacarpophalangeal joint osteoarthritis (rectangle), (b) Clinical photograph showing both components of SR™ MCP implant, and (c) Clinical photograph showing insertion of implant through the dorsal incision, note that there is a correct alignment of the third finger (arrow).

DISCUSSION

M. Mauclaire/Dieterich's disease is an uncommon condition mostly affecting the third metacarpal head. In literature, only case reports with no reliable conclusions regarding etiology and pathogenesis could be found. Any predispositions in systemic lupus erythematosus, juvenile dermatomyositis, gene mutations, long-term medication of glucocorticoids and intraosseous microinfarcts by repetitive microtrauma on the prominent third metacarpal head as the central pillar to load transmission are discussed [12–14]. Wright et al. [15] suggest a predisposition in vascular malformations of the epiphyseal vascular network that was found in 35% of specimens.

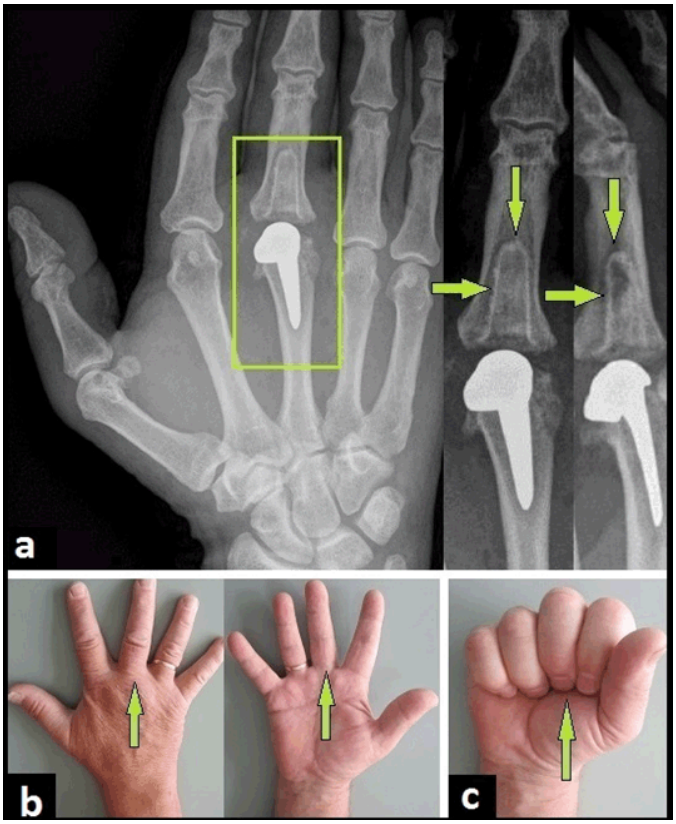


Figure 2: (a) Posteroanterior and lateral photographs showing correct positioning of the implant without any signs of loosening nor subsidence, note the sufficient cement coating around the distal UHMWPE component (arrows), (b) Clinical photographs demonstrating complete restoration of passive and active long finger extension, and (c) Clinical photograph demonstrating complete fist conclusion.

The preservation of motion in the metacarpophalangeal joints II–V has a top priority. A stable and functioning metacarpophalangeal joint is the key for satisfactory function of the overall finger. The stable active extrinsic motion-arc modulates synergistically the intrinsic function in the proximal interphalangeal (PIP) joint for a powerful extension and fist conclusion. On the other hand, the actions of the intrinsic muscles are necessary for stabilizing the metacarpophalangeal joint in flexion posture during PIP joint motion. Functional flexion postures averaged about 60 degree at the metacarpophalangeal and PIP joint and 40 degree at the distal interphalangeal (DIP) joint [16, 17]. A metacarpophalangeal joint arthrodesis should be avoided, and it is only considered when other surgical procedures have been failed [18]. Metacarpophalangeal joint resection arthroplasty can be one surgical option for low demand and/or rheumatoid patients [19].

In addition to the initial conservative treatment in patients with M. Mauclaire/ Dieterich's disease [20], joint-preserving surgical procedures and joint replacement can be applied. The core decompression is the method of choice when smaller intra-osseous findings are present [6]. For larger intra-osseous focal findings, curettage and filling of the necrotic cavity with autologous cancellous bone grafts is recommended [3, 21]. The subcapital flexion osteotomy (open wedge) of the metacarpal can be applied if the dorsal joint surface does not show cartilage lesions [22, 23]. For central or dorsal cartilage lesions the mosaicplasty is recommended [10]. Erne et al. [4] published satisfactory results with two cases following transplantation of a metatarsal head.

If the metacarpophalangeal joint is completely involved in osteoarthritis, the total joint replacement is inevitable. The unconstrained partial cemented metacarpophalangeal joint resurfacing SR™ MCP implant is one of the new generation type that is current in use [24]. The metacarpophalangeal joint is a condylar ball-and-socket joint with a convex surface on the metacarpal head and an incongruent (larger radius of curvature) concave surface on the proximal phalanx. One of the major complications of all unconstrained metacarpophalangeal joint implants is luxation tendency in the ulnopalmar direction. The SR™ MCP implant is designed to decrease this risk by having a greater arc of curvature on the dorsal aspect of the proximal component. In a biomechanical study, a higher intrinsic stability of this implant compared to un-affected human cadaver joints could be evaluated [25]. One disadvantage of implant is that cement removal is difficult in the necessity of revision and also there is a concern about the effect of heat polymerization [26]. Further studies with long-term results are needed to validate this concept.

CONCLUSION

M. Mauclaire/Dieterich's diesease as a rare condition mostly affecting the third metacarpal head. Motion preserving procedures at the metacarpophalangeal joints II-V are absolutely required to obtain function of the overall long finger. When distintictive osteoarthritis in metacarpophalangeal joints II-V is present, the use of an unconstrained resurfing metacarpophalangeal joint replacement is one surgical option that can be recommended. Arthrodesis or resection arthroplasty as a primary procedure should be avoided, and is to be considered as a salvage option when a joint replacement has failed.

****

Author Contribution

Ingo Schmidt – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

Copyright

© 2017 Ingo Schmidt. This article is distributed under the terms of Creative Commons Attribution License which permits unrestricted use, distribution and reproduction in any medium provided the original author(s) and original publisher are properly credited. Please see the copyright policy on the journal website for more information.

REFERENCES

1. Mauclaire P. Epiphysite des tetes metacarpiennes avec main un peu creuse. [Article in French]. Bulle et Mem Soc Nat Chir 1927;33:1377–8.
2. Dieterich H. Die subchondrale Herderkrankung am Metakarpale III. [Article in German]. Arch Klin Chir 1932;171:555–67.
3. Karlakki SL, Bindra RR. Idiopathic avascular necrosis of the metacarpal head. Clin Orthop Relat Res 2003 Jan;(406):103–8.
4. Erne HC, Lanz U, van Schoonhoven J, Prommersberger KJ. Aseptic osteonecrosis of the head of the metacarpal (Mauclaire's disease): Case report and review of the literature. [Article in German]. Handchir Mikrochir Plast Chir 2008 Jun;40(3):207–10.
5. Sagar P, Shailam R, Nimkin K. Avascular necrosis of the metacarpal head: A report of two cases and review of literature. Pediatr Radiol 2010 Dec;40(12):1895–901.
6. Myerthall SL, Graham B. Osteonecrosis of the base of the second metacarpal: A case report. J Hand Surg Am 1999 Jul;24(4):853–5.
7. Ares O, Seijas R, Conesa X, Pedemonte J. Avascular necrosis of the metacarpal head: Dieterich's disease. Acta Orthop Belg 2008 Oct;74(5):693–6.
8. Hu MH, Chen WC, Chang CH. Idiopathic osteonecrosis of the third metacarpal head. J Formos Med Assoc 2008 Jan;107(1):89–92.
9. Carstam N, Danielsson LG. Aseptic necrosis of the head of the fifth metacarpal. Acta Orthop Scand 1966;37(3):297–300.
10. Maes M, Hansen L, Cheyns P. Osteochondral mosaicplasty as a treatment method for bilateral avascular necrosis of the long finger metacarpal: Case report. J Hand Surg Am 2010 Aug;35(8):1264–8.
11. McGoldrick NP, McGoldrick FJ. Avascular necrosis of the metacarpal head: A case of Dietrich's disease and review of the literature. Am J Case Rep 2015 Jan 12;16:12–5.
12. Björkman A, Jörgsholm P, Burtscher IM. Osteonecrosis of the metacarpal head in a patient with a prothrombin 20210A gene mutation. Scand J Plast Reconstr Surg Hand Surg 2005;39(6):379–81.

13. Robinson AB, Rabinovich CE. Avascular necrosis of the metacarpals in juvenile dermatomyositis. J Clin Rheumatol 2010 Aug;16(5):233–6.
14. Thienpont E, Vandesande W, De Smet L. Dieterich's disease: Avascular necrosis of the metacarpal head: A case report. Acta Orthop Belg 2001 Apr;67(2):182–4.
15. Wright TC, Dell PC. Avascular necrosis and vascular anatomy of the metacarpals. J Hand Surg Am 1991 May;16(3):540–4.
16. Tomaino MM, Leit M. Finger metacarpophalangeal joint disease: The role of resection arthroplasty and arthrodesis. Hand Clin 2006 May;22(2):195–200.
17. Rongières M. Surgical treatment of degenerative osteoarthritis of the fingers. Chir Main 2013 Sep;32(4):193–8.
18. Beldner S, Polatsch DB. Arthrodesis of the Metacarpophalangeal and Interphalangeal Joints of the Hand: Current Concepts. J Am Acad Orthop Surg 2016 May;24(5):290–7.
19. Gruber AA. Long-term results of resection arthroplasty of the metacarpophalangeal joint in rheumatoid arthritis. [Article in German]. Handchir Mikrochir Plast Chir 2005 Feb;37(1):2–6.
20. Wijeratna MD, Hopkinson-Woolley JA. Conservative management of Dieterich disease: Case report. J Hand Surg Am 2012 Apr;37(4):807–10.
21. De Smet L. Avascular necrosis of the metacarpal head. J Hand Surg Br 1998 Aug;23(4):552–4.
22. Ohta S, Kakinoki R, Fujita S, Noguchi T. Open wedge flexion osteotomy of the metacarpal neck for the avascular necrosis of the third metacarpal head: Case report. Hand Surg 2012;17(2):251–3.
23. Wada M, Toh S, Iwaya D, Harata S. Flexion osteotomy of the metacarpal neck: A treatment method for avascular necrosis of the head of the third metacarpal: A case report. J Bone Joint Surg Am 2002 Feb;84-A(2):274–6.
24. Ibsen Sørenson A. Avanta SR-MCP arthroplasties, a prospective consecutive study for five years. J Hand Surg Eur 2011;36 E (Suppl 1):49.
25. Kung PL, Chou P, Linscheid RL, Berglund LJ, Cooney WP 3rd, An KN. Intrinsic stability of an unconstrained metacarpophalangeal joint implant. Clin Biomech (Bristol, Avon) 2003 Feb;18(2):119–25.
26. Linscheid RL. Implant arthroplasty of the hand: Retrospective and prospective considerations. J Hand Surg Am 2000 Sep;25(5):796–816.

ABOUT THE AUTHOR

Article citation: Schmidt I. The idiopathic avascular osteonecrosis of the third metacarpal head (M. Mauclaire/ Dieterich's disease). Int J Case Rep Images 2017;8(2):95–95.

Ingo Schmidt is a surgeon in the Department of Traumatology SRH Poliklinik, Waldklinikum Gera GmbH, Germany. From 1983 to 1989, he studied human medicine at the Friedrich-Schiller-University in Jena (Germany). From 1990 to 1999, Dr. Schmidt graduated his training for general surgery, traumatology, orthopaedics, and hand surgery at the University hospital in Jena. In 1994, he successfully defended his scientific work to gain the title as a medical doctor. He has published more than 20 scientific articles. His areas of interest include hip replacement, coverage of soft tissue defects, and hand surgery with special focus on total wrist replacement and arthroplasties of all other joints of the hand.

Access full text article on other devices

Access PDF of article on other devices

CASE REPORT PEER REVIEWED | OPEN ACCESS

Burkitt's lymphoma presenting as jejunojejunal intussusception in a child: A case report

Karimy Hamad Mehanna, José Ederaldo Queiroz Telles, Danielle Priscila Mauro

ABSTRACT

Burkitt's lymphoma represents 1–2% of all lymphoma and displays aggressive behavior, with B cells showing rapid growth. Its presentation as intestinal intussusception is unusual. We describe case of a seven-year-old boy with intussusception due to Burkitt's lymphoma. An emergency laparotomy revealed jejunojejunal intussusception as the cause of obstruction. The patient underwent enterectomy with restoration of the bowel continuity. Pathologist diagnosed Burkitt's lymphoma. He is receiving chemotherapy and is well one month postoperatively.

Keywords: Burkitt's lymphoma, Child, Intussusception, Non-Hodgkin lymphoma

How to cite this article

Mehanna KH, Telles JEQ, Mauro DP. Burkitt's lymphoma presenting as jejunojejunal intussusception in a child: A case report. Int J Case Rep Images 2017;8(2):96–100.

Karimy Hamad Mehanna[1], José Ederaldo Queiroz Telles[2], Danielle Priscila Mauro[3]

Affiliations: [1]MD, Department of Pediatric Surgery, Municipal Hospital and Maternity of São José dos Pinhais, State of Paraná, Brazil; [2]MD, PhD, Department of Pathology, Municipal Hospital and Maternity of São José dos Pinhais, Federal University of Paraná, State of Paraná, Brazil; [3]MD, First-year resident in pediatrics, Municipal Hospital and Maternity of São José dos Pinhais, State of Paraná, Brazil.

Corresponding Author: Karimy Hamad Mehanna, Department of Pediatric Surgery, Municipal Hospital and Maternity of São José dos Pinhais, State of Paraná, Brazil; E-mail: drakarimy@gmail.com

Received: 13 October 2016
Accepted: 01 November 2016
Published: 01 February 2017

Article ID: Z01201702CR10754KM

****

doi:10.5348/ijcri-201715-CR-10754

INTRODUCTION

Intussusception of the bowel can be described as the telescoping of a proximal segment of the intestine within the lumen of the adjacent segment. First reported in 1674, it is one of the most common causes of an acute abdomen in infancy. Burkitt's lymphoma is a subtype of non-Hodgkin lymphoma (NHL), showing malignant features and rapid growth. It was first described by Denis Burkitt in 1958. Commonly, presents as acute leukemia or malignant lesions in extra nodal sites. Intussusception due to Burkitt's lymphoma is rare presenting, with nonspecific symptoms. We present a case of jejunojejunal intussusception secondary to Burkitt's lymphoma in a child, and we would like to share the presentation, outcome, and our experience in the management of this child, with a review of literature.

CASE REPORT

A 7-year-old boy reported to the emergency with abdominal pain and vomiting for the last two days. A detailed history revealed an intermittent colicky abdominal pain for at least one year. No changes in bowel habits and no weight loss was reported.

On clinical examination, abdomen was mildly distended with palpable mass in the left flank and signs of peritoneal reaction in this affected site. Bowel sounds were reduced. All other aspects of the examination were normal. Laboratory investigations showed total leukocyte counts (TLC) of 12.55/mm^3 with neutrophilia and raised inflammatory markers (CRP 3.56 mg/dl). Chest X-ray was normal; abdominal plain X-ray showed dilated loops of bowel containing gas, but the findings were

nonspecific for intestinal obstruction or ileus (Figure 1). Ultrasonography (USG) showed target sign located at mesogastrium and left flank. The diameter of the lesion was 4 cm and the length was 6 cm (Figure 2). Additionally, a small amount of ascites was noted.

A decision for an emergency laparotomy was taken. Intraoperative findings confirmed jejunojejunal intussusception (Figure 3). The involved parts of the bowel were resected and end-to-end anastomosis was performed. During inspection of the bowel, we noted a mass within the jejunum, working as lead point of the intussusception (Figure 4). Cut sections of the specimen showed jejunal tumor (Figure 5). The histopathological diagnosis was Burkitt's lymphoma with clear surgical margins (Figure 6). On immunohistochemistry, CD-20 was positive (Figure 7).

Postoperatively the patient made an uneventful recovery and was referred to the oncology team for appropriate further management. He was subject to chest CT scan and bone-marrow biopsy. Both tests were normal. The disease was classificated in stage II (extra nodal site, disease restricted to abdomen). He is receiving vincristine, adriamycin, cyclophosphamide and prednisolone based chemotherapy on Department of Oncology (chemotherapy protocol of the French Society of Pediatric Oncology) and is well one month postoperatively.

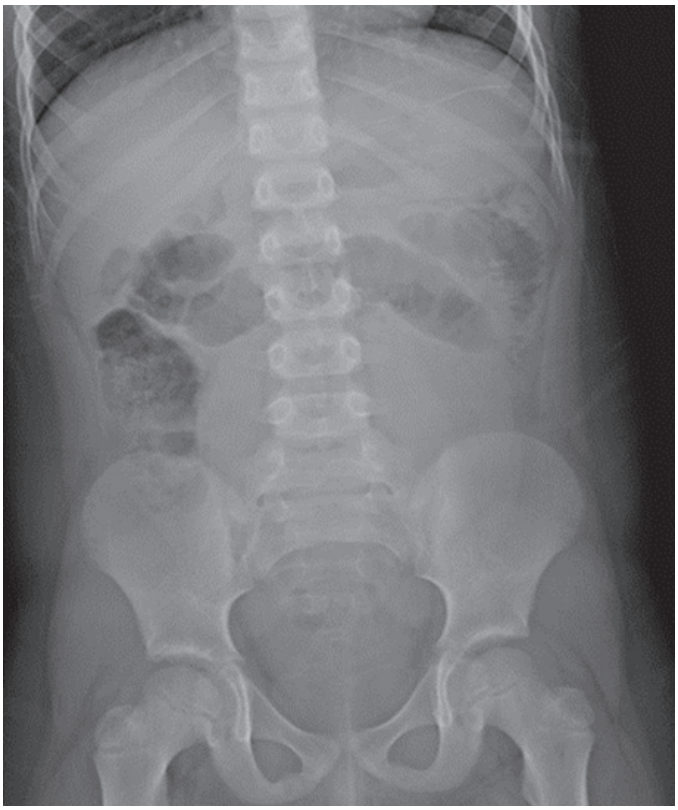


Figure 1: Abdominal plain X-ray showed dilated loops of bowel containing gas but the findings were nonspecific for intestinal obstruction or ileus.

DISCUSSION

Non-Hodgkin Lymphoma (NHL) is the third most frequent cancer of childhood and represents 1–4% of all gastrointestinal malignancies [1]. The peak age for gastrointestinal NHL in children is 5–15 years with a male sex preponderance 1.8-2.5 times that of female [2]. Small and large intestines are frequent affected and ileum is most commonly involved, in which the greatest concentration of gut-associated lymphoid tissue is present. Non-Hodgkin Lymphoma (NHL) is usually subdivided into three histological subtypes: (a) 65% B-cell NHL including both Burkitt's lymphoma and diffuse large B cell lymphoma – most commonly predict the abdomen as a primary site of presentation; (b) 20% lymphoblastic, and (c) 15% anaplastic large cell lymphoma. Burkitt's NHL is the most common, representing 40–50% of all NHL cases in childhood [3].

In upto 18% patients with primary abdominal Burkitt's lymphoma, intussusception is the first clinical sign. The incidence of NHL acting as lead point in intussusception is reported to be as high as 17%, and even higher (more than 50%) in children over 4–6 years of age [4]. Abdominal pain is present in 80% of cases, along with nausea, vomiting, changes in bowel habits and weight loss. Intestinal involvement by NHL was associated with an increased frequency of abdominal symptoms resulting in earlier laparotomies and earlier diagnosis [4].

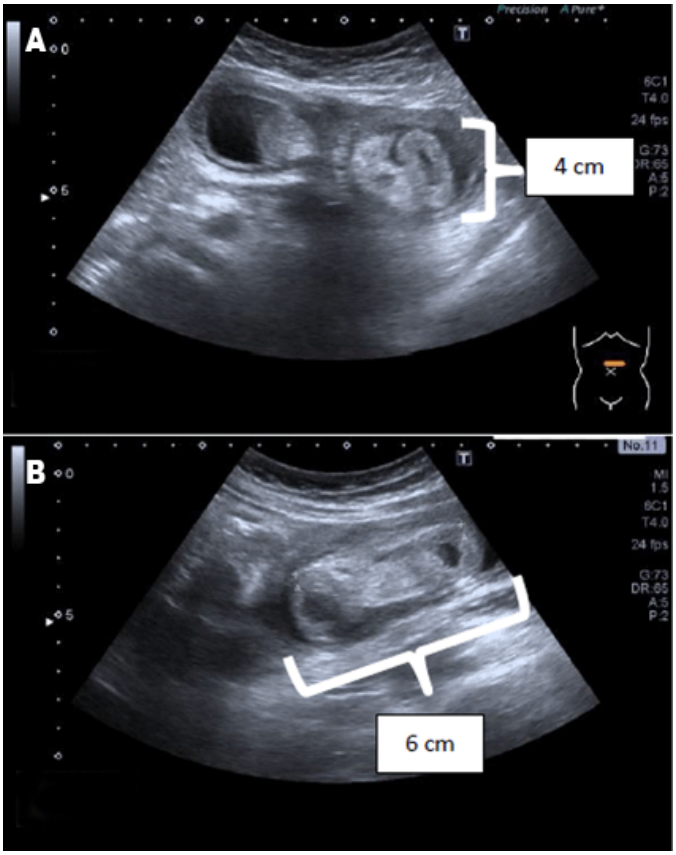


Figure 2: Ultrasonography showed target sign located at mesogastrium and left flank. (A) Diameter of the lesion was 4 cm, (B) Length of the lesion was 6 cm.

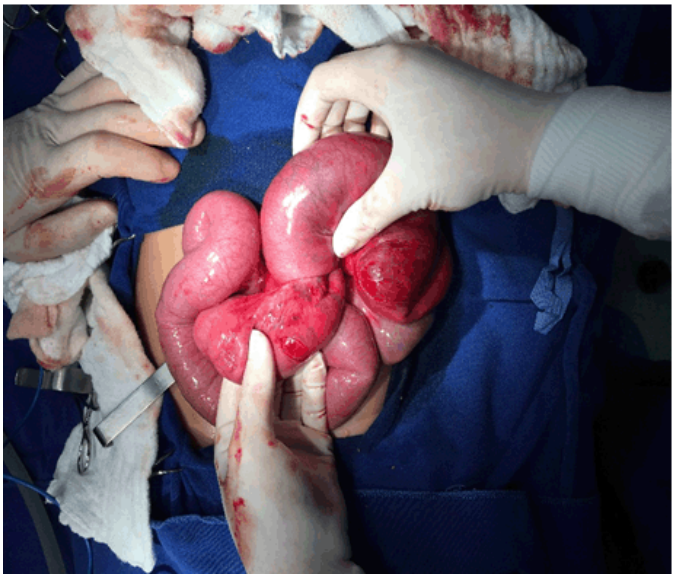


Figure 3: Small bowel intussusception: Jejunojejunal type of enteroenteric intussusception.

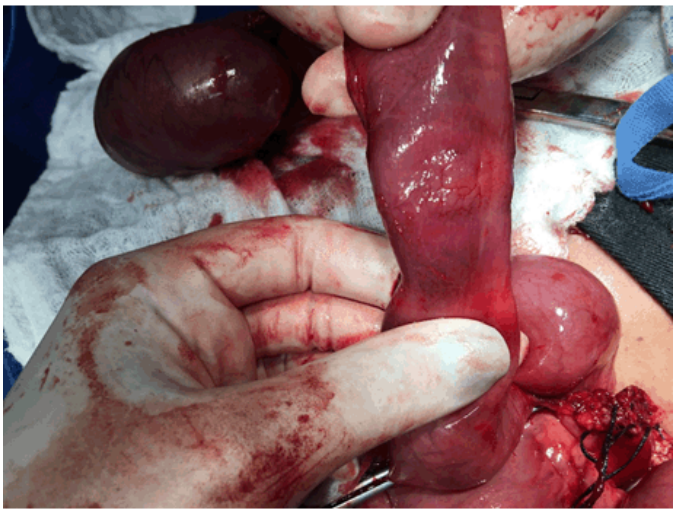


Figure 4: Jejunal tumor working as lead point of small bowel intussusception.

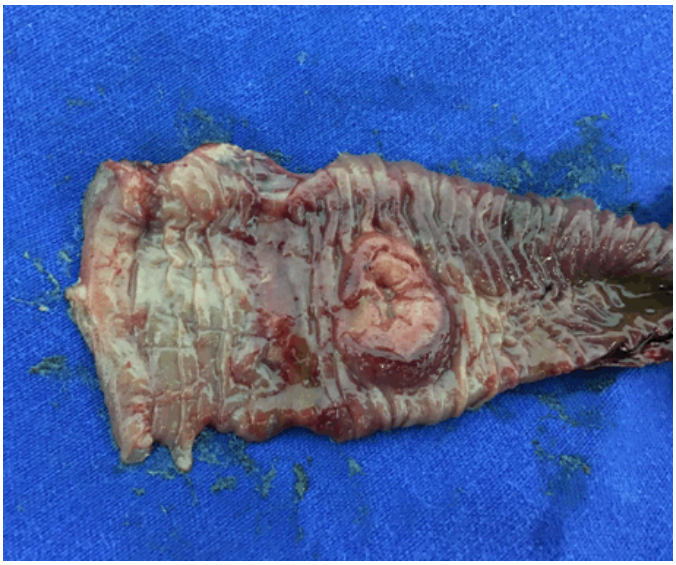


Figure 5: Cut section of the specimen showing jejunal tumor.

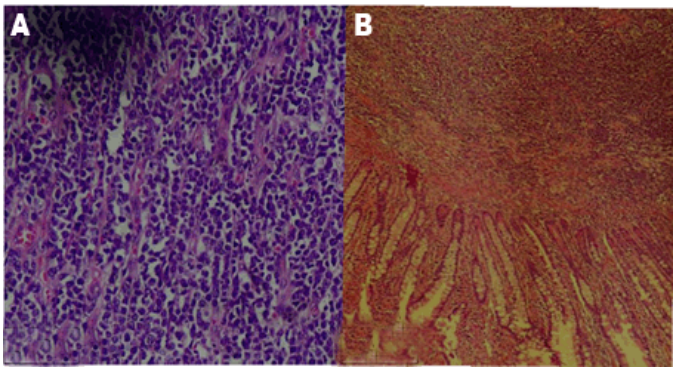


Figure 6: (A) Medium sized round malignant cells with frequent mitosis and apoptosis (H&E stain, x100), (B) Small bowel wall infiltration by diffuse malignant neoplasm (H&E stain, x100).

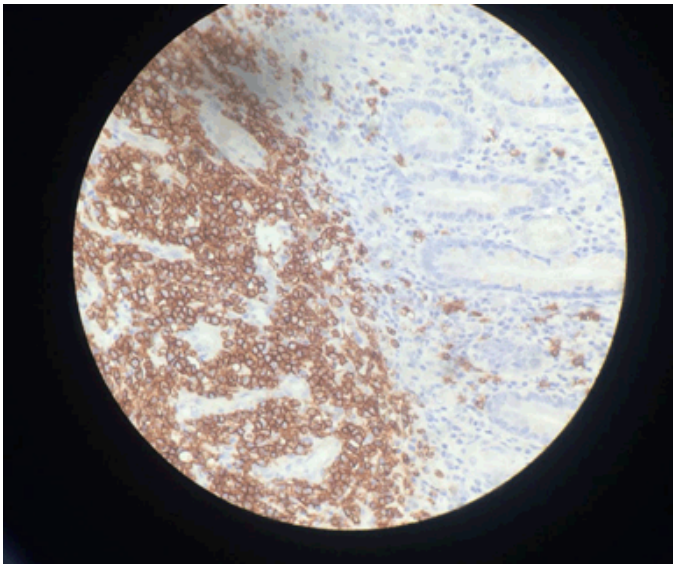


Figure 7: Malignant cells express intense CD20 membranous positivity (CD 20, x400).

Previous author's experiences with the combination of NHL with intussusception have not been satisfactory. In Ein et al. study, only 3 of 11 children were term survivors [5]. This study of over than 1200 infants and children in Toronto over a 40-year period revealed 11 lymphomas manifested as a leading point for the intussusception. Most patients were older than 4.5 years, were chronically ill, sometimes for several months, and had weight loss and abdominal mass, all of which pointed to a malignant process [5]. Puri et al. also reported that the only one death out of the entire series of 292 children with intussusception was a child with lymphoma [6].

The ideal treatment of gastrointestinal lymphoma must be individualized based on the type of disease and its location and a multidisciplinary approach with surgery and chemotherapy increases the chances of event free survival. Surgery plays a pivotal role in the management. In fact, LaQuaglia et al. in his study has concluded that bowel resection performed during emergency laparotomies for symptomatic, localized bowel involvement in patients

with NHL was associated with better prognosis [7]. Some reports demonstrate higher survival rate (58–89%) in patients having extensive surgical resection versus patients having only partial or incomplete resection (40–45%) at 2–5 years [3]. Complete resection of the tumor was shown to have the added advantage of avoiding bowel perforation, gastrointestinal hemorrhage or the tumor lysis syndrome after the initiation of chemotherapy [4]. Chemotherapy represents a cornerstone in the treatment of these patients and offers an excellent chance for long term disease free survival. Burkitt's lymphoma is very sensitive to chemotherapy. Therapy courses include the following drugs: cyclophosphamide, methotrexate, cytarabine, ifosfamide, etoposide, vincristine, vindesine, adriamycin, doxorubicin, dexamethasone. Rituximab is currently being studied in clinical trials, because it has shown good results in adult NHL. Although regimens are effective, they are toxic and up to 3% patients can die from treatment complications, with the acute tumor lysis syndrome being one of them. Burkitt's lymphoma patients who present with intussusception have a low stage disease (stage II), have a complete resection of their tumor, require shorter-duration, and less intense chemotherapy than patients diagnosed in other ways. Extent of disease at presentation and the resectability has been found to be the most important prognostic factor [4].

CONCLUSION

Intussusception should be considered in the differential diagnosis of all children who present with an acute abdomen, regardless of age. In the older age group of children, we need to keep a high index of suspicion for malignant lymphoma of the bowel. The clinical presentation is non-specific with abdominal pain, nausea and vomiting in most of the cases. Surgery is the gold standard in both diagnosis and treatment, ensuring the excision of the entire tumor with free margins. A multidisciplinary team with an oncologist assures efficient therapeutic management.

****

Author Contributions

Karimy Hamad Mehanna – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

José Ederaldo Queiroz Telles – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published

Danielle Priscila Mauro – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

Copyright

© 2017 Karimy Hamad Mehanna et al. This article is distributed under the terms of Creative Commons Attribution License which permits unrestricted use, distribution and reproduction in any medium provided the original author(s) and original publisher are properly credited. Please see the copyright policy on the journal website for more information.

REFERENCES

1. Ko SF, Lee TY, Ng SH, et al. Small bowel intussusception in symptomatic pediatric patients: Experiences with 19 surgically proven cases. World J Surg 2002 Apr;26(4):438–43.
2. Gupta A, Kumar S, Hai A, Agarwal S. Primary non hodgkin lymphoma of large bowel presenting as ileocolocolic intussusception in a child : A rare clinical presentation. The Internet Journal of Pediatrics and Neonatology 2008;10:1.
3. Tiwari C, Khedkar K, Jayaswal S, Shah H. Burkitt's lymphoma presenting as intussusception in four children. Pediatric Oncall Journal 2016;13:(3).
4. Shakya VC, Agrawal CS, Koirala R, et al. Intussusception due to non Hodgkin's lymphoma; different experiences in two children: Two case reports. Cases J 2009;2:6304.
5. Ein SH, Stephens CA, Shandling B, Filler RM. Intussusception due to lymphoma. Journal of Pediatric Surgery 1986;21(9):786–8.
6. Puri P, Guiney EJ. Small bowel tumors causing intussusception in childhood. Journal of Pediatric Surgery May 1986;21(5):461.
7. LaQuaglia MP, Stolar CJ, Krailo M, et al. The role of surgery in abdominal non-Hodgkin's lymphoma: Experience from the Childrens Cancer Study Group. J Pediatr Surg 1992 Feb;27(2):230–5.

Access full text article on other devices

Access PDF of article on other devices

CASE REPORT PEER REVIEWED | OPEN ACCESS

Diaphragmatic pacing as a treatment option for congenital central hypoventilation syndrome

Rodrigo A. S. Sardenberg, Riad N. Younes

ABSTRACT

The aim of this study is to present a 15-month-old child, case of congenital central hypoventilation syndrome successfully treated by diaphragmatic pacing. The diagnosis of this syndrome depends on the documentation of hypoventilation during sleep in the absence of primary neuromuscular, lung, cardiac, metabolic disease, or an identifiable brainstem lesion. While the cause of central congenital hypoventilation syndrome is not completely elucidated, the patients have mutations of the PHOX2B gene on chromosome 4. The diaphragmatic pacemaker currently represents an excellent treatment option, and the use of this device can provide reduction in upper airway infections and quality of life improvement.

Keywords: Diaphragm, Diaphragmatic paralysis, Pacemaker, Phrenic nerve

How to cite this article

Sardenberg RAS, Younes RN. Diaphragmatic pacing as a treatment option for congenital central hypoventilation syndrome. Int J Case Rep Images 2017;8(2):101–104.

Article ID: Z01201702CR10755RS

Rodrigo A. S. Sardenberg[1], Riad N. Younes[1]

Affiliations: [1]MD, Thoracic Surgery Department, Hospital Alemão Oswaldo Cruz Brazil.

Corresponding Author: Rodrigo Afonso Sardenberg, Rodrigo Claudio st, 477, apt 6, 01532-020 – São Paulo; E-mail: rodafs@uol.com.br

Received: 03 October 2016
Accepted: 26 October 2016
Published: 01 February 2017

****

doi:10.5348/ijcri-201716-CR-10755

INTRODUCTION

Classical congenital central hypoventilation syndrome (CCHS), also known as Ondine's Curse, is characterized by hypoventilation with normal respiration rates and shallow breathing during sleep with adequate ventilation during wakefulness. Severely affected individuals hypoventilate also when awake [1]. The CCHS represents an increasingly recognized group of conditions characterized by respiratory and autonomic nervous system dysregulation [2]. This rare disease was first reported in 1970 as a case report titled "Failure of Autonomic Control of Ventilation" [3], and came most visibly to the medical and public community with the American Thoracic Society (ATS) statement on CCHS in 1999 [2].

The diagnosis of CCHS is suspected due to absence of adequate ventilation during sleep, and others diseases were ruled out. While the cause of CCHS is not completely elucidated, those with CCHS have mutations of the PHOX2B gene, on chromosome 4. There are two types of disease presentation: the classic way, where the patient needs ventilatory support only during sleep (80% of cases), and the severe (20% of cases) when the patient requires ventilatory support 24 hours/day.

We report a CCHS patient with severe presentation type, successfully treated by diaphragmatic pacing.

CASE REPORT

A 15-month-old child, CCHS patient was referred as a candidate for diaphragmatic pacing with phrenic nerve stimulation, because he had chronic apnea and been dependent on mechanical ventilation since birth. At referral, his clinical condition was good. Blood tests and chest tomography were normal. The PHOX2B mutation was confirmed as 20/26.

The patient underwent general anesthesia without muscle blockers with single intubation. A staged bilateral mini-thoracotomy to access the pleural cavity was performed. The electrodes were placed underneath the phrenic nerves through careful dissection, and sutured to the pericardium by 4-0 prolene suture.

The receivers were placed in the subcutaneous tissue on the costal margin, and once they were connected to the electrodes, and they tested they showed good diaphragmatic function. The incisions were closed in layers and no chest tube was necessary. After an uneventfully recovery period, the diaphragm pacing was initiated four weeks after surgery.

Radiofrequency signals, generated by a battery-powered transmitter, were sent from an external antenna (Figure 1A), fixed to the implanted receivers (Figure 1B), which convert the radio signals into electrical impulses, causing diaphragmatic contraction.

To avoid fatigue, diaphragmatic pacing was initiated at a frequency of 15 Hz for 30 minutes during each waking hour in the first week, with increases of 30–45 minutes/week as tolerated by the patient. After 150 days hospitalization, the patient was discharged in good clinical condition, being submitted to 24 hours/day of continuous diaphragmatic pacing every day.

DISCUSSION

Congenital central hypoventilation syndrome is a rare disease— one for every 200,000 births, usually genetic in origin, resulting from a mutation in the gene PHOX2B on chromosome 4. This mutation causes a disorder in the central nervous system which leads to apnea, especially during REM sleep [2]. According to the gravity of the case, even when the individual is awake, able to maintain a satisfactory breathing on their health. As a result of hypoventilation, these individuals became hypoxemic and hypercarbic but lack the normal ventilation and arousal responses to the endogenous challenges during sleep, and the perception of asphyxia during wakefulness with and without exertion. When the disease is congenital, symptoms are present from birth, and the main symptoms are difficulty in maintaining the breathing frequency, difficulty in swallowing, cardiac arrhythmia, changes in temperature, eye disorders and gastroesophageal reflux. In 20% of patients, congenital megacolon (Hirschsprung's disease) is present, when so called these findings Haddad syndrome.

The diagnosis of such disease is made initially with the clinical state of the patient, followed by the completion of genetic testing, which identifies the type of gene mutation and PHOX2B, therefore, more appropriate monitoring of possible malfunctions associated with the specific type of mutation, and a genetic counseling for parents who wish to have other children.

Normally, other neuromuscular diseases, cardiac and neurological disorders should be investigated. There are two types of CCHS presentation: the classic way, where the patient needs ventilatory support only during sleep (80% of cases), and the severe (20% of cases) when the patient requires ventilatory support 24 hours/day.

The diaphragmatic pacemaker currently represents an excellent treatment option.

Available for use in the US for 40 years, and released in Brazil by ANVISA in 2009. It has been used by our group in 21 patients of various etiologies, all successfully. The youngest patient implanted in Brazil —the patient in this study— was 15 months old at the time of implantation and is progressing successfully.

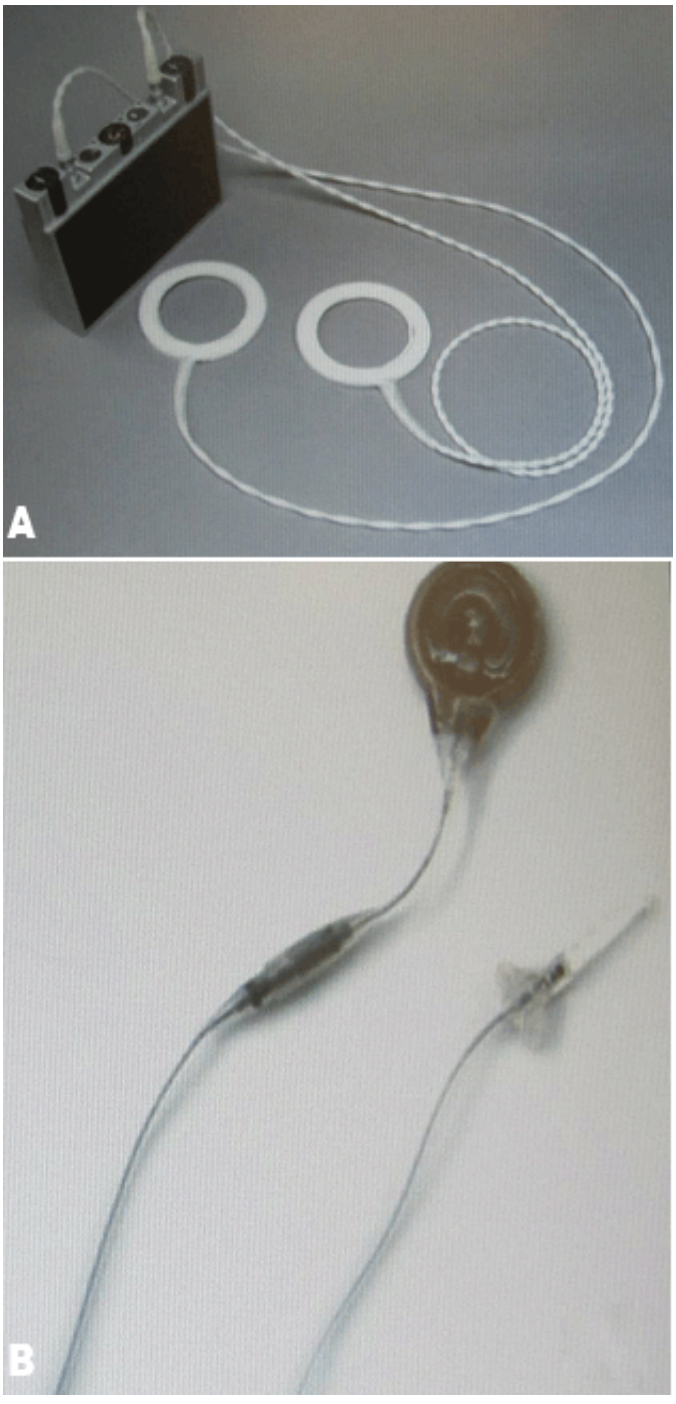


Figure 1: Device for phrenic nerve stimulation: (A) External parts and (B) Internal parts.

Figure 2: Patient carrying the device in a backpack.

Currently around the world, some patients are pacing for 30 years, and many for 20 years. The longest pacer patient in Brazil is pacing full time for three years [4]. diaphragmatic pacing can provide advantages to patients such as: reduction in lung infections; tracheostomy decannulation in some cases; ventilator weaning and better quality of life [4].

An upper airway evaluation is another assessment that can be helpful, specially in those where tracheostomy decannulation is being considered. Increasing the size of the upper airway with tonsillectomy and/or adenoidectomy may help minimize upper airway obstruction [5]. If the patient can sustain adequate ventilation with a small tracheostomy, the cannula may be removed [6].

Recently, a new drug treatment (desogestrel) for CCHS patients— in order to improve CO_2 chemosensitivity— was reported. One of the two patients described that without setting up the non-invasive ventilation, in this context, the benefit of desogestrel is currently conjectural [7]. Successful diaphragm pacing requires proximity to a medical team willing to maintain this system. Therefore, diaphragm pacing is an attractive alternative mode of mechanically assisted ventilation for many patients with CCHS.

Patients can lead a much more normal life by being ventilator-free, enabling them to participate in daily activities, thus improving quality of life.

CONCLUSION

Diaphragm pacing is an attractive alternative mode of mechanically assisted ventilation for many patients with congenital central hypoventilation syndrome. Patients can lead a much more normal life by being ventilator free, enabling them to participate in daily activities, thus improving quality of life.

****

Author Contributions

Rodrigo A. S. Sardenberg – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published
Riad N. Younes – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

Copyright

© 2017 Rodrigo A. S. Sardenberg et al. This article is distributed under the terms of Creative Commons Attribution License which permits unrestricted use, distribution and reproduction in any medium provided the original author(s) and original publisher are properly credited. Please see the copyright policy on the journal website for more information.

REFERENCES

1. Weese-Mayer DE, Rand CM, Berry-Kravis EM, et al. Congenital central hypoventilation syndrome from past to future: Model for translational and transitional autonomic medicine. Pediatr Pulmonol 2009 Jun;44(6):521–35.
2. Weese-Mayer DE, Shannon DC, Keens TG, Silvestri JM. Idiopathic congenital central hypoventilation syndrome: Diagnosis and management. American Thoracic Society. Am J Respir Crit Care Med 1999 Jul;160(1):368–73.

3. Mellins RB, Balfour HH Jr, Turino GM, Winters RW. Failure of automatic control of ventilation (Ondine's curse). Report of an infant born with this syndrome and review of the literature. Medicine (Baltimore) 1970 Nov;49(6):487–504.
4. Sardenberg RA, Secaf LB, Pinotti AC, Taricco MA, Brock RS, Younes RN. Diaphragmatic pacing: unusual indication with successful application. [Article in English, Portuguese]. J Bras Pneumol 2011 Sep-Oct;37(5):697–9.
5. Chen ML, Tablizo MA, Kun S, Keens TG. Diaphragm pacers as a treatment for congenital central hypoventilation syndrome. Expert Rev Med Devices 2005 Sep;2(5):577–85.
6. Kun SS, McComb JG, Shaul DB, Atkinson JB, Keens TG. 23 years of diaphragmatic pacing in children. Am J Respir Critic Care Med 2004 Jul;172(2):200–5.
7. Straus C, Trang H, Becquemin MH, Touraine P, Similowski T. Chemosensitivity recovery in Ondine's curse syndrome under treatment with desogestrel. Respir Physiol Neurobiol 2010 Apr 30;171(2):171–4.

Access full text article on other devices

Access PDF of article on other devices

CASE REPORT PEER REVIEWED | OPEN ACCESS

Streptococcus pseudoporcinus subacute mitral valve endocarditis: A case report

Peir-Yu Fang, Sandeep A. Gandhi

ABSTRACT

This is a case of a patient with subacute *Streptococcus pseudoporcinus* endocarditis, who presented with subjective fever, weight loss, and mental status changes. Multiple blood cultures were positive for *Streptococcus pseudoporcinus* and echocardiogram revealed mitral valve vegetation. Antibiotics cleared his blood, but the patient underwent a mitral valve replacement. This article illustrates a potentially emerging infection by a relatively new pathogen.

Keywords: Indolent infection, Mitral valve endocarditis, Non-female genital source, *Streptococcus pseudoporcinus* bacteremia, Subacute endocarditis

How to cite this article

Peir-Yu F, Gandhi SA. *Streptococcus pseudoporcinus* subacute mitral valve endocarditis: A case report. Int J Case Rep Images 2017;8(2):105–107.

Article ID: Z01201702CR10756PF

Peir-Yu Fang[1], Sandeep A. Gandhi[2]

Affiliations: [1]DO, MS, Third-year resident in Family Medicine at Peconic Bay Medical Center-Northwell Health; Riverhead, NY, USA; [2]MD FACP FIDSA, Infectious Disease Consultant, Department of Medicine at Peconic Bay Medical Center-Northwell Health; Riverhead, NY, USA.

Corresponding Author: Sandeep A. Gandhi, Infectious Disease Consultant, Department of Medicine at Peconic Bay Medical Center-Northwell Health; Riverhead, NY, USA; E-mail: sansu1994@hotmail.com

Received: 04 October 2016
Accepted: 03 November 2016
Published: 01 February 2017

****

doi:10.5348/ijcri-201717-CR-10756

INTRODUCTION

Streptococcus pseudoporcinus is a β-hemolytic *Streptococcus* first described in 2006. However, it was misidentified as *Streptococcus porcinus* prior to its formal recognition. Most reported cases of *Streptococcus pseudoporcinus* were from isolates in female genitourinary tract. The similarities between the biochemical characteristics of *Streptococcus pseudoporcinus* and *Streptococcus agalactiae* pose challenges to distinction of the species under conventional testing methods [1–4].

CASE REPORT

A 77-year-old male with a history of chronic obstructive pulmonary disease, mitral valve prolapse, Raynaud's phenomenon, left knee replacement, a pulmonary embolism two years ago, and chronic back pain, who underwent spinal nerve radiofrequency ablations on October 13 and on November 10, 2015. He presented to our hospital with a weight loss of 15 pounds over a period of three months, a subjective fever in the previous one month, and altered mental status for one day prior to admission. The review of systems was negative for chills, sweats, flu-like symptoms, headache, vision changes, sore throat, earache, neck pain, cough, shortness of breath, abdominal pain, nausea, vomiting, diarrhea, constipation, back or joint pain, rash, or genitourinary complaints. No injuries were reported. There was no history of antibiotic use in the past 90 days or international travel. Home medications included vitamin D 5000 units daily and acetaminophen extra strength 500 mg, two tablets every six hours as needed. He had quit smoking six months previously. There was no family history of recurrent infections. His temperature

was 97.8°F, pulse 72 beats per minute, respirations 18 breaths per minute, blood pressure 102 mmHg/68 mmHg, and pulse oximetry was 97% on two liters oxygen. Patient weighs 75 kg. On physical examination, he was alert and appropriately verbally responsive. His neck was supple and no cervical lymphadenopathy was palpable. The heart examination did not reveal any murmurs, rubs, or gallops and the lungs were clear to auscultation. There was no splenomegaly or spinal tenderness. The extremities were without edema, splinter hemorrhages, Osler's nodes, or Janeway lesions.

Initial labs were: white blood cell count 8,600/uL, with neutrophilia of 83.8%, hemoglobin 10.7 g/dL, and platelet 161,000/uL, sodium 129 mmol/L and creatinine 1.0 mg/dL. Erythrocyte sediment rate 105 mm/hr and C-reactive protein 8.04 mg/dL. Urinalysis was positive for large blood and 8–12 red blood cells per high powered field. Chest X-ray and CT chest without contrast were both negative for acute airspace disease and a pleural effusion. A CT scan of brain revealed severe chronic microvascular disease with age related volume loss. Computed tomography scan of lumbar spine without contrast did not show any fluid collection, but multilevel degenerative disk disease with moderate spinal canal stenosis from L1 to S1 was noted.

The patient was admitted to the hospital and was started on piperacillin/tazobactam and received a one gram dose of intravenous vancomycin in the emergency room. After admission, patient did not have any documented fevers. Blood cultures obtained prior to initiation of antibiotics showed *Streptococcus pseudoporcinus* with confirmation tests repeated three times. Subsequently, the antibiotic was switched to ceftriaxone. A repeat blood culture was negative. Echocardiogram showed a prolapsed mitral valve and a large mobile mass on the posterior outflow valve leaflet. The mass was seen prolapsing through the coopting valve (Figure 1). Doppler showed severe mitral valve regurgitation. The left ventricular ejection fraction was 60% with normal wall motion. Patient was transferred to another hospital for valve replacement.

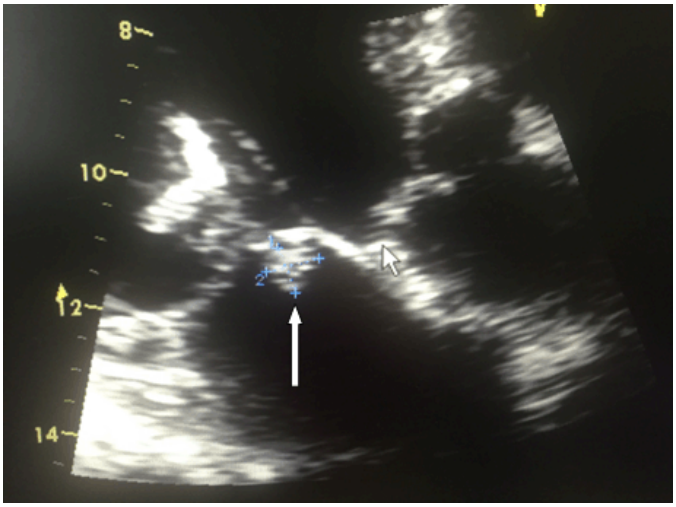


Figure 1: Echocardiogram from the patient showing the vegetation in the outflow side of mitral valve (long arrow).

DISCUSSION

In the microbiology lab, when the patient's blood cultures became positive for gram-positive cocci in chains, a catalase test was performed. It was negative. Subsequently, the Vitek system confirmed the organism as *Streptococcus pseudoporcinus*. *Streptococcus pseudoporcinus* is a β-hemolytic *Streptococcus* with a wide-zone hemolysis first identified in 2006 [1]. Prior to 2006, *Streptococcus pseudoporcinus* was grouped under *Streptococcus porcinus*, which has been associated with rare cases of septicemia [2]. Most of the reported cases of *Streptococcus pseudoporcinus* infection were from isolates in female genitourinary tract [1–3]. *Streptococcus porcinus* and *Streptococcus pseudoporcinus* may be misidentified as Group B *Streptococcus* because of serological cross-reactivity [1]. In a 14-month prospective observational study done in Thailand, no isolates of *Streptococcus pseudoporcinus* were recovered from blood or sterile sites of any patients during their study period [4]. There was one case of a thumb infection due to *Streptococcus pseudoporcinus* reported in 2009 in Washington State from injury sustained by a car door jamb [5]. In this report, the authors believed that the patient acquired the infection from his wife's vaginal tract. To our knowledge, there has been no report of *Streptococcus pseudoporcinus* subacute endocarditis. Four out of five reported cases of bacteremia due to *Streptococcus pseudoporcinus* identified in a study were female while the gender of the remainder case was unidentified [6].

Streptococcus porcinus and *Streptococcus pseudoporcinus* are hippurate hydrolysis negative, while *Streptococcus agalactiae* is positive. *Streptococcus porcinus* is about 100% Voges-Proskauer positive while only about half of *Streptococcus pseudoporcinus* strains are Voges-Proskauer positive [5, 7]. *Streptococcus porcinus* and *Streptococcus pseudoporcinus* can be differentiated from *Streptococcus agalactiae* by fermentation of mannitol and sorbitol. However, because of the similarities in the biochemical characteristics, 16s rRNA gene sequencing is necessary to differentiate *Streptococcus pseudoporcinus* from *Streptococcus porcinus* [1–7]. We report a rare case of *Streptococcus pseudoporcinus bacteremia* with secondary endocarditis from a non-female genitourinary source.

We conducted a search of the English literature via OVID and PubMed; however, it did not yield any case reports of subacute *Streptococcus pseudoporcinus* endocarditis to date.

CONCLUSION

Streptococcus pseudoporcinus is mostly associated with female genitourinary infections. No known association of *Streptococcus pseudoporcinus* with endocarditis has been reported previously. We have reported a rare case of *Streptococcus pseudoporcinus bacteremia* associated with endocarditis from a non-genitourinary source. This organism may be an emerging pathogen in the etiology of mitral valve endocarditis.

****

Acknowledgements
We are thankful to Dr. Rameshchandra Dabhi, at Peconic Bay Medical Center-Northwell Health for the echocardiogram image.

Author Contributions
Peir-Yu Fang – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published
Sandeep A. Gandhi – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published

Guarantor
The corresponding author is the guarantor of submission.

Conflict of Interest
Authors declare no conflict of interest.

Copyright
© 2017 Peir-Yu Fang et al. This article is distributed under the terms of Creative Commons Attribution License which permits unrestricted use, distribution and reproduction in any medium provided the original author(s) and original publisher are properly credited. Please see the copyright policy on the journal website for more information.

REFERENCES

1. Bekal S, Gaudreau C, Laurence RA, Simoneau E, Raynal L. Streptococcus pseudoporcinus sp. nov., a novel species isolated from the genitourinary tract of women. J Clin Microbiol 2006 Jul;44(7):2584–6.
2. Gaudreau C, Simoneau E, Labrecque O, et al. Epidemiological, biochemical and antimicrobial susceptibility characteristics of Streptococcus pseudoporcinus isolated in Quebec, Canada, from 1997 to 2006. J Med Microbiol 2007 Dec;56(Pt 12):1620–4.
3. Stoner KA, Rabe LK, Austin MN, Meyn LA, Hillier SL. Incidence and epidemiology of Streptococcus pseudoporcinus in the genital tract. J Clin Microbiol 2011 Mar;49(3):883–6.
4. Suwantarat N, Grundy M, Rubin M, et al. Recognition of *Streptococcus pseudoporcinus* colonization in women as a consequence of using matrix-assisted laser desorption ionization-time of flight mass spectrometry for group B Streptococcus identification. J Clin Microbiol 2015 Dec;53(12):3926–30.
5. Mahlen SD, Clarridge JE 3rd. Thumb infection caused by Streptococcus pseudoporcinus. J Clin Microbiol 2009 Sep;47(9):3041–2.
6. Shewmaker PL, Steigerwalt AG, Whitney AM, et al. Evaluation of methods for identification and determination of the taxonomic status of strains belonging to the Streptococcus porcinus-Streptococcus pseudoporcinus complex isolated from animal, human, and dairy sources. J Clin Microbiol 2012 Nov;50(11):3591–7.
7. Ocque R, Pasculle W. Clinical Microbiology: Case 648 – Vaginal Culture of a Pregnant Woman. [Available at: http://path.upmc.edu/cases/case648.html]

Access full text article on other devices

Access PDF of article on other devices

CASE REPORT PEER REVIEWED | OPEN ACCESS

A case of uncombable hair syndrome and global developmental delays

Hana Kathryn Cobb, Alvin Yuhico Tiu

ABSTRACT

Introduction: Uncombable hair syndrome, also known as spun-glass hair and pili trianguli et canaliculi, is a rare genetic disorder where a structural anomaly of the hair shaft causes silvery-blonde, disorganized and unruly hair. The hair is usually normal in infancy, becomes uncombable during the first year of life, and improves during puberty or later in life. All scalp hair is affected. However, other body hair is unaffected. Uncombable hair syndrome is usually isolated and not typically associated with developmental delays or disorders of other organs. Autosomal dominant and sporadic inheritance patterns have been reported. A genetic mutation or causal gene has not been identified. Case Report: We report a case of 17-month-old girl with uncombable hair and global developmental delays. She was born with straight brown hair, However by four months of age her hair became silvery, stiff, and unruly. Extensive evaluation by specialists failed to identify a reason for her developmental delays. Both parents were healthy and had normal hair. The maternal grandmother had blonde, coarse and slightly unruly hair that was worse when she was younger and improved during puberty. Microscopic hair evaluation of the child's hair showed triangular cross sections (pili trianguli) and longitudinal canal-like grooves (pili canaliculi) confirming the diagnosis of uncombable hair syndrome. Conclusion: Uncombable hair syndrome is a rare genetic disorder that must be suspected clinically. Scanning electron microscopy can confirm the diagnosis, differentiate uncombable hair syndrome from other hair shaft anomalies and accurately determine the inheritance pattern within a family.

Keywords: Congenital hair anomalies, Genodermatoses, Pili trianguli et canaliculi, Spun-glass hair, Uncombable hair syndrome

Hana Kathryn Cobb[1], Alvin Yuhico Tiu[2]

Affiliations: [1]BS, Fourth Year Medical Student, University of Washington School of Medicine, Seattle, Washington, United States; [2]MD, Attending Physician, Family Medicine, VA Puget Sound Health Care System, Seattle, Washington, United States.

Corresponding Author: Hana Kathryn Cobb, 1514 Bellevue Avenue #502, Seattle, Washington, 98122; E-mail: hanacobbuwsom@gmail. com

Received: 19 August 2016
Accepted: 21 October 2016
Published: 01 February 2017

How to cite this article

Cobb HK, Tiu AY. A case of uncombable hair syndrome and global developmental delays. Int J Case Rep Images 2017;8(2):108–111.

Article ID: Z01201702CR10757HC

****

doi:10. 5348/ijcri-201718-CR-10757

INTRODUCTION

Uncombable hair syndrome, also known as spun-glass hair and pili trianguli et canaliculi, is a rare genetic disorder where a structural anomaly of the hair shaft causes the hair to be silvery-blonde, disorganized and stand out from the scalp [1–3]. The hair is usually normal in infancy, becomes uncombable during the first year of life, and

improves in late childhood or during puberty [1–4]. The stiffness and unruly nature of the hair is due to premature keratinization of the inner root sheath [1]. The diagnosis must be suspected clinically and can be confirmed with microscopy. Scanning electron microscopy shows both characteristic features of uncombable hair syndrome: triangular cross sections (pili trianguli) and canal-like longitudinal grooves (pili canaliculi) [1–5]. Uncombable hair syndrome is usually an isolated disorder and not associated with developmental delays, diseases of other organs, or other genetic syndromes [1, 2]. Autosomal dominant and sporadic inheritance patterns have been reported. A genetic mutation or causal gene has yet to be identified [1, 2, 4, 6].

CASE REPORT

A 17-month-old baby girl was presented to the dermatology and genetics clinics with unusual hair. She was born at 36 weeks gestation by spontaneous vaginal delivery following an uneventful pregnancy. She received phototherapy for perinatal jaundice. She was born with straight dark brown hair; however, by four months of age her hair became progressively silvery-blonde, dry, stiff and unruly. Her parents were unable to comb her hair flat and different treatments and shampoos were not effective. She had low muscle tone and global developmental delays. She did not walk and had just started to crawl. She had difficulty swallowing and sometimes rocked back and forth to soothe herself when she was upset. Both parents were healthy, had no other children, and had normal hair. The maternal grandmother had blonde, coarse and slightly unruly hair that was worse when she was younger and improved during puberty. No other family members had unusual hair.

On examination the patient's scalp hair was shiny, silvery-blonde, coarse and stuck out from her head (Figure 1). Her eyebrows, eyelashes, fingernails, toenails and the remainder of her body hair were normal. The quality and appearance of her skin was normal. On neurological assessment she was hypotonic with decreased deep tendon reflexes. Her fine motors skills, gross motor skills and speech were delayed six months. A swallow study showed aspiration of thin liquids. Routine vision screening, hearing screening, EEG and brain MRI scan were normal. Genetic and metabolic testing including array comparative genomic hybridization, newborn metabolic screening, Prader–Willi syndrome testing, urine organic acids, plasma amino acids, and transferrin isoelectric focusing for congenital disorders of glycosylation were normal. Serum ammonia, lactate, copper, ceruloplasmin and creatine kinase were also normal. The extensive medical testing and evaluation failed to identify a unifying cause or genetic syndrome to explain her hypotonia and developmental delays. Uncombable hair syndrome was suspected based on the appearance of her scalp hair and the family history. Scanning electron microscopy of her hair showed triangular cross sections (pili trianguli) and longitudinal canal-like grooves (pili canaliculi) consistent with uncombable hair syndrome. The patient's family was advised to gently brush her hair and avoid harsh chemicals in the hair. They were advised that the stiffness and unruly nature of her hair should improve during puberty or later in life.

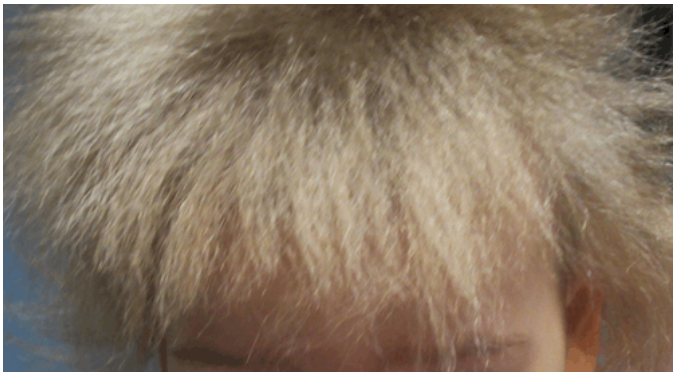


Figure 1: Scalp hair in uncombable hair syndrome.

DISCUSSION

Uncombable hair syndrome, also known as spun-glass hair and pili trianguli et canaliculi, is a rare genetic hair disorder. It was likely first described in the book 'Les Velus' (the Hirsute) where an affected individual was described as having mop hair [7]. The hair is generally normal in infancy, becomes unruly and uncombable between three and twelve months of life, and improves during puberty or later in life [1–5]. All scalp hair is affected; however, the eyelashes, eyebrows and remaining body hair are generally unaffected. The hair quantity and tensile strength is normal [1–3]. A structural anomaly of the hair shaft causes the hair to be silvery-blonde, disorganized, coarse and stand out from the scalp. The stiffness and

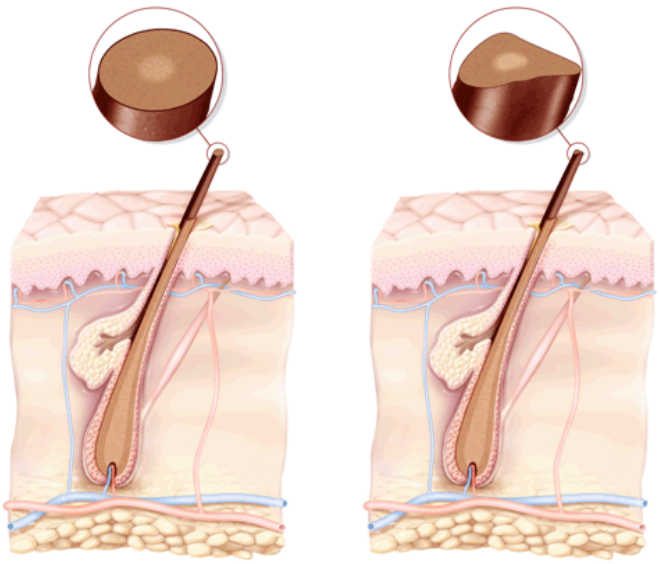


Figure 2: The shaft of normal hair (left) and hair in uncombable hair syndrome (right).

unruly nature of the hair is due to the triangular, or kidney-shaped, hair shaft when viewed in cross section (Figure 2). Premature keratinization of the inner root sheath of the hair alters the shape of the hair shaft [3]. The silvery or shiny appearance of the hair is due to the way the unique hair shaft reflects light [3, 7].

Treatment for uncombable hair syndrome is largely ineffective and generally limited to gentle handling, soft brushes, avoidance of harsh chemical treatments and creative styling [3, 5]. There is a report of a patient who had increased hair growth and improved combability of their hair following four months of oral biotin supplementation [8]. Hair conditioners with zinc pyrithione may lead to some improvement due to a moisturizing effect [3]. The condition usually spontaneously improves in late childhood or during puberty [2, 3, 5].

The diagnosis of uncombable hair syndrome must be suspected clinically and is confirmed with microscopy [1, 7]. The differential diagnosis is primarily the other structural hair anomalies including monilethrix, pili annulati, pili torti (twisting hair), pseudomonilethrix, trichorrhexis invaginata (bamboo hair), trichorrhexis nodosa (broom stick hair), and wooly hair [2, 5, 6]. Most hair shaft defects can be readily seen with light microscopy. Light microscopy of an uncombable hair syndrome hair shaft may show a band or darkening on one edge caused by the shadow as light passes over the longitudinally grooved hair. Gently moving the microscope's micrometer may allow the observer to see the canal along the length of the hair [2, 3]. However, often the hair appears normal under light microscopy. The diagnosis of uncombable hair syndrome can be reliably confirmed with scanning electron microscopy where the two classic hair shaft alterations are clearly seen: triangular cross sections (pili trianguli) and longitudinal canal-like grooves (pili canaliculi) [1–3, 5]. If scanning electron microscopy is not available, light microscopic examination of hairs embedded in paraffin show the triangular hair cross sections [3, 5].

Uncombable hair syndrome is usually an isolated condition and not typically associated with other genetic syndromes, disorders of other organs, developmental delays or neuropsychological problems. Genetic counseling for uncombable hair syndrome is not indicated unless features of other genetic syndromes are present [1, 2, 9]. Affected individuals often have a negative family history, implying sporadic inheritance, though the characteristic hair shaft anomaly may be seen in clinically unaffected family members when hair is evaluated with scanning electron microscopy. In one report, two siblings were clinically affected and both parents had normal hair. Scanning electron microscopic evaluation of the affected siblings and their father showed the classic hair morphology (pili trianguli et canaliculi) indicating autosomal dominant inheritance in the family [2, 4]. Uncombable hair syndrome is not clinically detectable unless at least 50% of the hairs are affected [1, 3]. Autosomal dominant inheritance has been reported with both complete and incomplete penetrance [3, 9]. Families thought to have incomplete penetrance may actually show complete penetrance when clinically unaffected family members are evaluated with scanning electron microscopy. A causal gene, locus or mutation has yet to be identified [3, 5, 6].

CONCLUSION

Uncombable hair syndrome is a rare genetic structural hair shaft anomaly with autosomal dominant and sporadic inheritance patterns reported. Treatment is usually not effective and the condition often spontaneously resolves during puberty. Scanning electron microscopy is useful to differentiate uncombable hair syndrome from the other hair shaft anomalies and to accurately determine the inheritance pattern within a family. Potential future investigations to consider include attempts to identify a causative gene and determine possible associations of uncombable hair syndrome with other medical conditions or genetic syndromes.

****

Author Contributions

Hana Kathryn Cobb – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Alvin Yuhico Tiu – Substantial contributions to conception and design, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

Copyright

© 2017 Hana Kathryn Cobb et al. This article is distributed under the terms of Creative Commons Attribution License which permits unrestricted use, distribution and reproduction in any medium provided the original author(s) and original publisher are properly credited. Please see the copyright policy on the journal website for more information.

REFERENCES

1. Resta M. Pili trianguli et canaliculi (uncombable hair syndrome). European Journal of Pediatric Dermatology 2015;25(2):116–7.

2. Pereira G, Xavier L, Valente N, Machado-Pinto J. Do you know this syndrome? Anais Brasileiros de Dermatologia 2006;81(2):1–6.
3. Trueb R. Uncombable hair syndrome. Orphanet Encyclopedia 2003. [Available at: http://www.orpha.net/data/patho/GB/uk-uncombable.pdf]
4. Hebert AA, Charrow J, Esterly NB, Fretzin DF. Uncombable hair (pili trianguli et canaliculi): Evidence for dominant inheritance with complete penetrance based on scanning electron microscopy. Am J Med Genet 1987 Sep;28(1):185–93.
5. Sybert V. Genetic Skin Disorders, 2ed. New York: Oxford University Press; 2010. p. 211–12.
6. Rimoin D, Connor J, Pyeritz R, Korf B. Emery and Rimoin's Principles and Practice of Medical Genetics, 5ed. Philadelphia: Elsevier Ltd; 2007. p. 3536–40.
7. Zanca A, Zanca A. Ancient observations of "uncombable hair syndrome". Int J Dermatol 1993 Oct;32(10):707.
8. Shelley WB, Shelley ED. Uncombable hair syndrome: Observations on response to biotin and occurrence in siblings with ectodermal dysplasia. J Am Acad Dermatol 1985 Jul;13(1):97–102.
9. Calderon P, Otberg N, Shapiro J. Uncombable hair syndrome. J Am Acad Dermatol 2009 Sep;61(3):512–5.

Access full text article on other devices

Access PDF of article on other devices

CASE REPORT PEER REVIEWED | OPEN ACCESS

Successive spontaneous abortions caused by a whole-arm translocation between chromosome 10 homologs

Hana Kathryn Cobb, Dana Knutzen, Alvin Yuhico Tiu

ABSTRACT

Introduction: Whole-arm translocations between homologous chromosomes are rare genetic events that generally involve Robertsonian translocations between acrocentric chromosomes. These inter-homologous translocations only produce unbalanced zygotes, therefore never result in viable pregnancies. The only reports of whole-arm translocations between non-acrocentric homologs involve chromosomes 2, 4, 7 and 9. Case Report: We report a 21-year-old healthy female with isochromosomes for the short and long arm of chromosome 10 who was ascertained due to successive spontaneous abortions. Genetic testing of decidual tissue and the patient's peripheral blood revealed a whole-arm translocation between the chromosome 10 homologs: 46, XX, t(10;10)(p10;p10). The result of this translocation was two isochromosomes: i(10p) and i(10q). Conclusion: This is the first reported case of a whole-arm translocation between chromosome 10 homologs and is a unique case of successive spontaneous abortions. Our case expands the limited spectrum of whole-arm translocations between non-acrocentric homologous chromosomes.

Hana Kathryn Cobb[1], Dana Knutzen[2], Alvin Yuhico Tiu[3]

Affiliations: [1]BS, Fourth Year Medical Student, University of Washington School of Medicine, Seattle, Washington, United States; [2]CGC, Certified Genetic Counselor, CHI Franciscan Health, Tacoma, Washington, United States; [3]MD, Attending Physician, Internal Medicine, VA Puget Sound Health Care System, Seattle, Washington, United States.

Corresponding Author: Hana Kathryn Cobb, 1514 Bellevue Avenue #502, Seattle, Washington, 98122; E-mail: hanacobbuwsom@gmail.com

Received: 02 September 2016
Accepted: 21 October 2016
Published: 01 February 2017

Keywords: Isochromosome, Spontaneous abortion, Whole-arm translocation

How to cite this article

Cobb HK, Knutzen D, Tiu AY. Successive spontaneous abortions caused by a whole-arm translocation between chromosome 10 homologs. Int J Case Rep Images 2017;8(2):112–115.

Article ID: Z01201702CR10758HC

****

doi:10.5348/ijcri-201719-CR-10758

INTRODUCTION

Whole-arm translocations between homologous chromosomes are rare genetic events that generally involve Robertsonian translocations between acrocentric chromosomes. These inter-homologous translocations can only produce unbalanced zygotes, and never result in viable pregnancies [1–5]. The only reports of whole-arm translocations between nonacrocentric homologs involve chromosomes 2, 4, 7 and 9 [1–7]. We report the first case of a whole-arm translocation between chromosome 10 homologs.

CASE REPORT

A 21-year-old G2P0020 phenotypically normal female was referred for genetic counseling following two spontaneous abortions and an abnormal chromosome analysis. A pedigree is shown in Figure 1. She smokes five cigarettes per day and takes no prescription

medications. She had a deep venous thrombosis as a teenager thought to be secondary to oral contraceptives and tested negative for methylene tetrahydrofolate reductase (MTHFR) deficiency. Genetic testing showed that she was homozygous normal at the MTHFR 677C locus indicating no increased risk for cardiovascular events or arterial thrombosis. Her mother is healthy and had one spontaneous abortion. Her father has a history of systemic lupus erythematous, deep venous thrombosis, pulmonary embolus, MTHFR deficiency, and protein C and S deficiencies. The proband's spouse is healthy and has a five-year-old daughter with a cleft lip from a prior relationship. He was the father for both of the proband's pregnancy losses. His mother is healthy and had three spontaneous abortions. His father died from pancreatic cancer in between late 40s and early 50s. The family history is otherwise negative for stillbirths, neonatal deaths, birth defects or other spontaneous abortions.

The proband's pregnancies both resulted in first trimester spontaneous abortions followed by dilation and curettage. The first pregnancy loss was not further evaluated. Tissue obtained during the second dilation and curettage was sent for chromosome analysis (Figure 2). Twenty cells were counted and all five metaphase cells analyzed and karyotyped revealed a female chromosome complement with a whole-arm translocation between the chromosome 10 homologs: 46,XX,t(10;10)(p10;p10). The result of this translocation was a 10p isochromosome and a 10q isochromosome: i(10p) and i(10q). The tissue appeared to be all maternal decidua and therefore the result was thought to represent the maternal rather than the fetal karyotype. The proband's peripheral blood was subsequently sent for chromosome analysis (Figure 3). Twenty cells were counted and all eight metaphase cells that were analyzed and karyotyped revealed a female chromosome complement with a balanced whole-arm translocation between the chromosome 10 homologs similar to the i(10p) and i(10q) isochromosomes seen in the analysis of placental tissue. The i(10p) and i(10q) isochromosomes are further illustrated when displayed next to a chromosome 10 idiogram (Figure 4).

A reproductive endocrinology and infertility specialist evaluated the proband. She has regular monthly menses, transvaginal ultrasound showed a normal uterus and ovaries, lab evaluation of her hypothalamic-pituitary-ovarian axis was normal (thyroid stimulating hormone, follicle-stimulating hormone, luteinizing hormone and anti-mullerian hormone), and she had good ovarian reserve. Her evaluation indicated that she was capable of carrying a pregnancy, however, due to her unique genetic

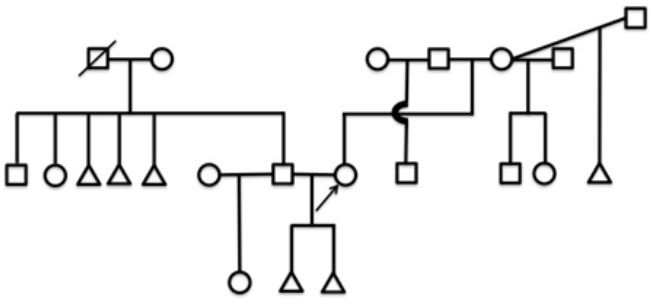


Figure 1: Pedigree of the patient.

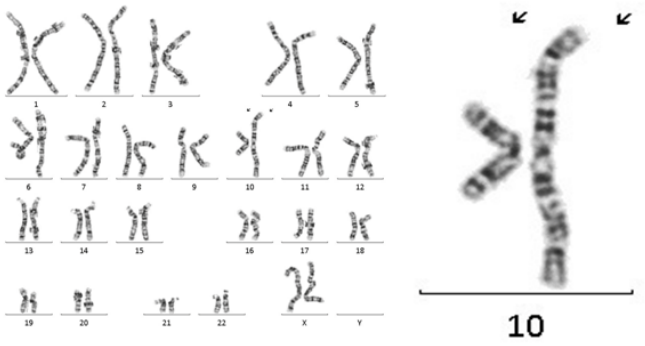


Figure 2: G-band analysis at the 400-band level of placental tissue (decidua). The arrows indicate the i(10p) (left) and i(10q) (right) isochromosomes.

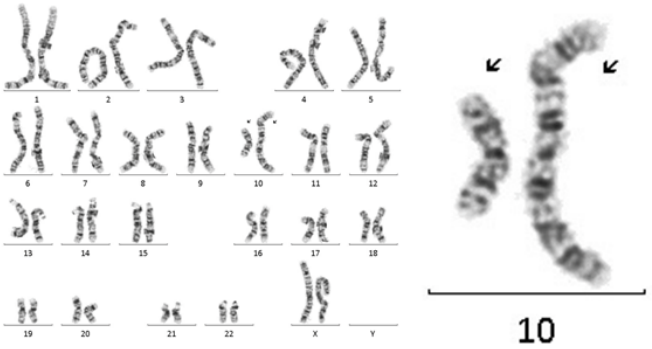


Figure 3: G-band analysis at the 550-band level of maternal peripheral blood (lymphocytes). The arrows indicate the i (10p) (left) and the i(10q) (right) isochromosomes.

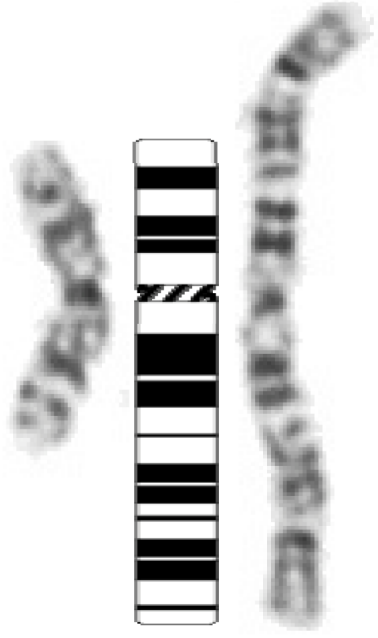


Figure 4: (A) Isochromosomes i(10p) and (c) i(10q) to the left and right of a (B) 550-band level chromosome ideogram.

composition she was told that she would be unable to conceive with her own oocytes. Reproductive and family building options were discussed: (1) donor oocyte IVF using spouse's sperm and donor eggs, (2) embryo adoption of a donated embryo, and (3) child adoption.

DISCUSSION

Balanced chromosomal rearrangements are relatively common in humans. Balanced translocation carriers are usually phenotypically normal and are at high risk for recurrent spontaneous abortions [1]. Pregnancies of balanced translocation carriers have four potential outcomes: (1) a child with normal chromosomes, (2) a child with the same balanced translocation as the affected parent, (3) a child with a chromosomal imbalance which may cause an abnormal phenotype, and (4) spontaneous abortion. Translocations generally occur between non-homologous chromosomes. Whole-arm translocations between homologous chromosomes are rare genetic events that usually involve Robertsonian translocations between acrocentric chromosomes [1, 6].

Ohama [6] discussed in detail various mechanisms that could lead to whole-arm translocations between nonacrocentric homologous chromosomes. Potential mechanisms for whole-arm translocation between chromosome 10 homologs include:

(a) crossover within a small pericentric inversion loop between homologs 10 during meiosis I,

(b) transverse division of the chromosome 10 homolog centromeres during meiosis II or mitosis and formation of isochromosomes 10, and

(c) a meiotic or mitotic breakage and exchange at or near the centromeres of the chromosomes 10.

All mechanisms except for (c) during mitosis result in unbalanced and nonviable zygotes due to the segregation of i(10p) and i(10q) into different daughter cells. Mechanism (c) is likely the event that led to our proband's genetic composition as it is the only one that would produce a balanced translocation zygote. If the translocation occurred between meiosis II and mitosis, it would result in a non-mosaic whole-arm translocation. If the translocation arose during early mitotic division, the result would be mosaicism [1, 5, 6].

In our case, no normal cell lines were found in the two cell types analyzed, and therefore only abnormal maternal gametes are predicted. However, mosaicism must be considered given that a limited number of cells in only two cell types were analyzed. There are reports of mosaicism involving whole-arm homologous translocations in chromosome 2 [1, 6]. In male translocation carriers, gonadal mosaicism and chromosomally balanced sperm can be identified by FISH analysis. In female translocation carriers, identification of gonadal mosaicism and chromosomally balanced ovum requires invasive methods: ovarian biopsy or in vitro fertilization (IVF) with ovum collection [1]. Potential future investigations for this patient include analysis of more decidual and peripheral blood cells, analysis of skin fibroblasts, and ovarian biopsy to look for evidence of mosaicism. Additional testing could include segregation analysis of the patient and her parents to determine the parental origin of the two isochromosomes. However, the proband's father is deceased and mother is not available for genetic testing.

CONCLUSION

This is the first reported case of a whole-arm translocation between chromosome 10 homologs and is a unique case of successive spontaneous abortions. Our case expands the limited spectrum of whole-arm translocations between nonacrocentric homologous chromosomes. Carriers of these unique chromosomal aberrations are likely to experience recurrent spontaneous abortions and will not be able to produce viable offspring. Mosaicism must be considered in cases of whole-arm translocations. These patients should receive genetic and infertility counseling to better understand their condition and their reproductive options.

****

Author Contributions

Hana Kathryn Cobb – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Alvin Yuhico Tiu – Substantial contributions to conception and design, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Dana Knutzen – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

Copyright

© 2017 Hana Kathryn Cobb et al. This article is distributed under the terms of Creative Commons Attribution License which permits unrestricted use, distribution and reproduction in any medium provided the original author(s) and original publisher are properly credited. Please see the copyright policy on the journal website for more information.

REFERENCES

1. Almeida C, Dória S, Moreira M, Pinto J, Barros A. Normal sperm in a 2;2 homologous male translocation carrier. J Assist Reprod Genet 2012 Jul;29(7):665–8.
2. Bernasconi F, Karagüzel A, Celep F, Normal phenotype with maternal isodisomy in a female with two isochromosomes: I(2p) and i(2q). Am J Hum Genet 1996 Nov;59(5):1114–8.
3. Eggerding FA, Schonberg SA, Chehab FF, Norton ME, Cox VA, Epstein CJ. Uniparental isodisomy for paternal 7p and maternal 7q in a child with growth retardation. Am J Hum Genet 1994 Aug;55(2):253–65.
4. Lindenbaum R, Woods C, Norbury C, Powey S, Rysteck G. An individual with maternal disomy of chromosomes 4 and iso(4p), iso(4q). American Journal of Human Genetics. suppl 1991;49:A285.
5. Niikawa N, Ishikawa M. Whole-arm translocation between homologous chromosomes 7 in a woman with successive spontaneous abortions. Hum Genet 1983;63(1):85–6.
6. Ohama K, Kusumi I, Takahara H, Kajii T. Successive spontaneous abortions including one with whole-arm translocation between chromosomes 2. Hum Genet 1978 Jan 19;40(2):221–5.
7. Björck EJ, Anderlid BM, Blennow E. Maternal isodisomy of chromosome 9 with no impact on the phenotype in a woman with two isochromosomes: I(9p) and i(9q). Am J Med Genet 1999 Nov 5;87(1):49–52.

Access full text article on other devices

Access PDF of article on other devices

CASE REPORT PEER REVIEWED | OPEN ACCESS

Liver disease masquerading as primary cardiopulmonary disease: Hepatopulmonary syndrome as a result of idiopathic cirrhosis

Bartholameuz Nuwan Aravinda, Ratnatilaka Asoka, Sadikeen Aflah

ABSTRACT

Introduction: Liver disease and portal hypertension can be associated with pulmonary vascular complications including hepatopulmonary syndrome and portopulmonary hypertension. Hepatopulmonary syndrome is characterized by arterial hypoxemia and pulmonary vascular dilatations. Exertional dyspnea, platypnea-orthodeoxia, cyanosis and digital clubbing are commonly found symptoms and signs in hepatopulmonary syndrome. Case Report: Here we discuss a patient with chronic liver disease whose initial presentation was hepatopulmonary syndrome with progressive exertional dyspnea, cyanosis and clubbing. Diagnosis of hepatopulmonary syndrome was made through a constellation of findings in blood gas analysis, contrast echocardiography with biochemical and ultrasound evidence of liver disease applied to standard criteria; having excluded other possible causes. Conclusion: Hepatopulmonary syndrome should be suspected in any patient with liver disease and hypoxia. It should also be formulated in the differential diagnosis of a patient with otherwise unexplained exertional dyspnea and cyanosis with digital clubbing.

Bartholameuz Nuwan Aravinda[1], Ratnatilaka Asoka[2], Sadikeen Aflah[3]

Affiliations: [1]MBBS, Registrar-Medicine, National Hospital, Colombo, Sri Lanka; [2]MBBS, MD, Consultant Physician, National Hospital, Colombo, Sri Lanka; [3]MBBS, MD, Consultant Chest Physician, Central Chest Clinic, Colombo, Sri Lanka.

Corresponding Author: Nuwan Aravinda Bartholameuz Ward 42/56A, National Hospital, Colombo 10, Sri Lanka, City- Colombo, Colombo 01000; E-mail: nbaravinda@gmail.com

Received: 04 September 2016
Accepted: 01 November 2016
Published: 01 February 2017

Keywords: Cirrhosis, Cyanosis, Digital clubbing, Hepatopulmonary syndrome

How to cite this article

Bartholameuz NA, Ratnatilaka A, Sadikeen A. Liver disease masquerading as primary cardiopulmonary disease: Hepatopulmonary syndrome as a result of idiopathic cirrhosis. Int J Case Rep Images 2017;8(2):116–119.

Article ID: Z01201702CR10759BA

****

doi:10.5348/ijcri-201720-CR-10759

INTRODUCTION

Cyanosis indicates the presence of deoxygenated haemoglobin of ≥5 g/dL in arterial blood. Causes of cyanosis include intracardiac right-left shunts, lung diseases and pulmonary arteriovenous malformation (AVM). Clubbing is due to fibrovascular proliferation in nail beds, mediated by platelet derived growth factor released from megakaryocytes or platelet emboli which do not reach nail bed unless there is pulmonary capillary damage or intracardiac shunts [1]. Here we discuss a rare cause of dyspnea, cyanosis and clubbing associated with cirrhosis which mimicked primary cardiopulmonary disease.

CASE REPORT

A 56-year-old female was admitted with a history of progressive shortness of breath on exertion for 6 months duration. She had severe central cyanosis with gross clubbing. Mild icterus, palmar erythema and prominent

peripheral pulses were also noted. Respiratory rate was 24/min. Systemic examination was normal. Arterial oxygen saturation (SpO_2) lying supine was 80% and on standing was 76%. Investigations revealed leucopenia ($4.31x10^3/\mu L$), thrombocytopenia ($31x10^3/\mu L$), and hemoglobin 15.6 g/dL with macrocytosis in peripheral blood smear. Arterial blood gas (ABG) analysis in supine position showed pH 7.43, PCO_2 28.6 mmHg, PaO_2 56 mmHg, HCO_3^- 19.4 mmol/L and Alveolar-arterial oxygen gradient of 58 mmHg. PaO_2 dropped to 50.2 mmHg upon standing.

Total serum protein was normal (67 g/dL) with reversed serum albumin: globulin ratio (0.6). Serum bilirubin was elevated (42.8 µmol/L). PT was 17.4 s (INR 1.26) and APTT was 46 s (26 s–40 s). Liver enzymes were normal (AST 40 U/L, ALT 22 U/L, and ALP 107 U/L). Abdominal ultrasound detected coarse echo texture in a normal size liver, splenomegaly (12.5 cm; normal <12 cm), portal vein diameter of 1.4 cm (<1.3 cm) without ascites or pleural effusion. Esophagogastroduodenoscopy showed mild portal hypertensive gastropathy and no varices. Hepatitis B and C serology were negative. Serum ferritin was 172 ng/mL. Serum ceruloplasmin levels were normal and there were no Kayser-Fleischer rings. ANA and anti-mitochondrial antibodies were negative. Lipid profile was normal. TSH, Free T4, and HbA1c were normal.

High resolution computed tomography of the chest (HRCT) revealed normal lung parenchyma (Figure 1). Contrast-enhanced computed tomography (CECT) of the chest and pulmonary angiogram detected pulmonary venous congestion and dilated venous collaterals without evidence of pulmonary hypertension or AVM. Gross splenorenal collaterals were noted (Figure 2). No evidence of portal vein thrombosis was found.

Transthoracic echocardiogram was normal. Transesophageal echocardiography detected a small patent foramen ovale (PFO) but bubble contrast study excluded functional PFO. There was indirect evidence of pulmonary capillary dilatation by detecting micro-bubbles in left atrium after three cardiac cycles following the appearance of bubbles in the right atrium.

Her lung function tests were normal (FEV1 94.1%; FVC 99.5%; FEV1: FVC 94.6%, VC 88.1%) except for low DLCO (44%) reflecting a diffusion-perfusion defect due to pulmonary vascular dilatation and hyperdynamic circulation.

This is a case with hypoxia, clubbing, cirrhosis and portal hypertension with evidence of intra-pulmonary vascular dilatation (IPVD) confirming the diagnosis of hepatopulmonary syndrome (HPS).

DISCUSSION

In this case, cyanosis was due to dilatation of pulmonary vessels leading to ventilation perfusion mismatch causing hypoxia. Aetiology of pulmonary vascular dilatation in cirrhosis is thought to relate to an

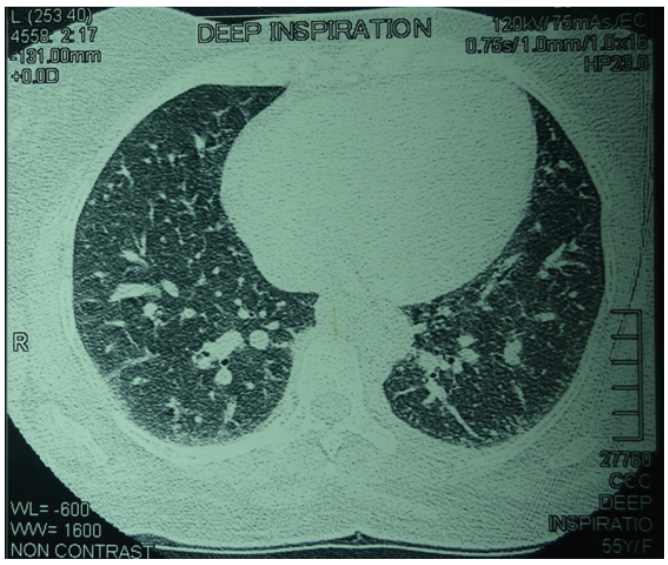


Figure 1: High resolution computed tomography of the chest showing normal lung parenchyma and dilated pulmonary vasculature extending up to pleural surface.

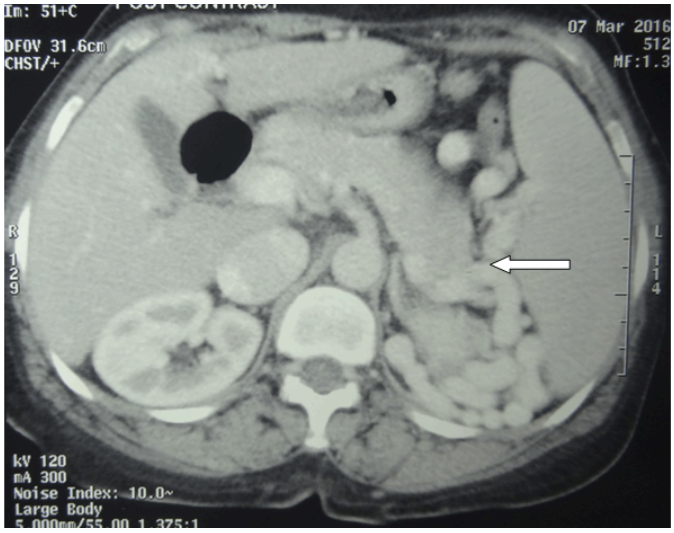


Figure 2: Contrast-enhanced computed tomography showing gross splenorenal collaterals (arrow).

increase in pulmonary NO by means of both endothelial and inducible NO synthase (eNOS and iNOS) [2–4].

An increased hepatic production of vasoconstrictor Endothelin-1 (ET-1) stimulates the production of ETB receptors in pulmonary microcirculation. ETA causes vasoconstriction while ETB causes vasodilatation through increase in eNOS activity [2–4].

Portal hypertension weakens intestinal mucosal barrier due to impaired drainage. It allows increased enteral translocation of bacteria and endotoxins which stimulate release of vasoactive substances like TNF-α. This leads to increased pulmonary sequestration of macrophages and local production of pro-inflammatory mediators causing an increase in iNOS activity and NO production [2–4].

Normal pulmonary functions and HRCT of the chest excluded chronic parenchymal lung disease.

Transesophageal echocardiography with bubble contrast study could not reveal any functional intra-cardiac shunts but indicated presence of pulmonary vascular dilatation in the absence of AVM. These micro-bubbles are larger than normal pulmonary capillary diameter (8–15 μm) and cannot pass through normal pulmonary capillaries [2]. Evidence of pulmonary vascular dilatation was also apparent in HRCT and CT pulmonary angiogram. The dilatation of small peripheral pulmonary vessels is the hallmark of HPS [5, 6].

This patient has orthodeoxia which is defined as a fall in PaO_2 ≥ 5% when upright, or 4 mmHg [2]. PaO_2 decreases in upright position as blood flow increases through already dilated vessels in basal segments of lungs, due to gravity. This increases ventilation-perfusion mismatch and hypoxia worsens [7].

Triad of liver disease (portal hypertension and/or cirrhosis), IPVD (positive findings in contrast echocardiography or abnormal uptake in the brain (>6%) with radioactive lung perfusion scanning) and arterial hypoxemia (PaO_2 <80 mmHg or alveolar arterial oxygen gradient > 15 mmHg while breathing ambient air) is diagnostic of HPS [2]. SpO_2 improved to 96% (PaO_2 99.9 mmHg) with oxygen (4 L/min via face mask). MELD (Model for end-stage liver disease) score was 12. Since only proven treatment for HPS is liver transplantation [3], patient was referred to transplant surgeon for further management.

Domiciliary oxygen was arranged as supportive therapy. The patient was a teetotaller and aetiology of cirrhosis and portal hypertension was not identified. Time taken from initial presentation to diagnosis approximates twenty months emphasized the importance of increased awareness of HPS among clinicians across different subspecialties [8].

CONCLUSION

Hepatopulmonary syndrome (HPS) should be suspected in any patient with established liver disease and hypoxia. It can also be the initial presentation of liver disease mimicking primary pulmonary disease such as interstitial lung disease or primary cyanotic heart disease with secondary cardiogenic cirrhosis. Identifying primary underlying pathology helps institution of appropriate care to patients. Therefore, HPS should also be entertained in differential diagnosis of a patient presenting with exertional dyspnea, cyanosis and digital clubbing beyond traditional cardiopulmonary causes.

****

Author Contributions

Bartholameuz Nuwan Aravinda – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published
Ratnatilaka Asoka – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published
Sadikeen Aflah – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Revising it critically for important intellectual content Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

Copyright

© 2017 Bartholameuz Nuwan Aravinda et al. This article is distributed under the terms of Creative Commons Attribution License which permits unrestricted use, distribution and reproduction in any medium provided the original author(s) and original publisher are properly credited. Please see the copyright policy on the journal website for more information.

REFERENCES

1. Spicknall KE, Zirwas MJ, English JC 3rd. Clubbing: An update on diagnosis, differential diagnosis, pathophysiology, and clinical relevance. J Am Acad Dermatol 2005 Jun;52(6):1020–8.
2. Rodríguez-Roisin R, Krowka MJ. Hepatopulmonary syndrome: A liver-induced lung vascular disorder. N Engl J Med 2008 May 29;358(22):2378–87.
3. Tumgor G. Cirrhosis and hepatopulmonary syndrome. World J Gastroenterol 2014 Mar 14;20(10):2586–94.
4. Porres-Aguilar M, Altamirano JT, Torre-Delgadillo A, Charlton MR, Duarte-Rojo A. Portopulmonary hypertension and hepatopulmonary syndrome: A clinician-oriented overview. Eur Respir Rev 2012 Sep 1;21(125):223–33.
5. McAdams HP, Erasmus J, Crockett R, Mitchell J, Godwin JD, McDermott VG. The hepatopulmonary syndrome: Radiologic findings in 10 patients. AJR Am J Roentgenol 1996 Jun;166(6):1379–85.
6. Chawla A, Gaikwad V, Dubey N, Bosco J. CT Pulmonary Angiography Features of a Hepatopulmonary Syndrome. Korean J Radiol 2015 Jul-Aug;16(4):951–2.
7. Naeije R. Hepatopulmonary syndrome and portopulmonary hypertension. Swiss Med Wkly 2003 Mar 22;133(11-12):163–9.
8. Dhoble C, Saoji N, Jeswani J, Rios R. A 45-Year-Old Undiagnosed Cirrhotic Patient with Hepatopulmonary Syndrome as First Presentation: A Case Report. Am J Case Rep 2015 Oct 22;16:751–5.

Access full text article on other devices

Access PDF of article on other devices

CASE REPORT PEER REVIEWED | OPEN ACCESS

A successfully-treated case of penetrating facial trauma

Tetsuya Yumoto, Atsuyoshi Iida, Kohei Tsukahara, Hiromichi Naito, Michihisa Terado, Keiji Sato, Isao Date, Atsunori Nakao

ABSTRACT

Introduction: Penetrating intracranial injury caused by a metal bar is rare and often causes severe damage without fast treatment. Case Report: A 59-year-old male fell from a height of four feet onto upward-pointed metal bars and was transferred to our emergency department after suffering an accidental penetration of a metal bar through his face. Computed tomography (CT) scans were conducted; however, the image resolution was unsatisfactory and the amount of brain damage could not be evaluated due to serious artifacts associated with the metal bar. After removing the foreign body, follow-up CT scn revealed increased hemorrhage in the frontal lobe. Emergency craniotomy and removal of the hematoma followed. Conclusion: Artifacts from a penetrating metal bar on CT scan often hide actual brain damage along the trajectory of penetration. Emergent surgical intervention and early follow-up CT scan is necessary for any chance to save the patient's life. In these cases, proper examination, radiological tests, and early decision-making using a multidisciplinary strategy provide optimal outcomes.

Keywords: Artifacts, Intracranial stab injury, Metal bar, Transorbital brain injury

How to cite this article

Yumoto T, Iida A, Tsukahara K, Naito H, Terado M, Sato K, Date I, Nakao A. A successfully-treated case of penetrating facial trauma. Int J Case Rep Images 2017;8(2):120–123.

Article ID: Z01201702CR10760TY

****

doi:10.5348/ijcri-201721-CR-10760

Tetsuya Yumoto[1], Atsuyoshi Iida[1], Kohei Tsukahara[1], Hiromichi Naito[1], Michihisa Terado[1], Keiji Sato[1], Isao Date[2], Atsunori Nakao[1]

Affiliations: [1]Department of Emergency and Critical Care Medicine, Okayama University Graduate School of Medicine, Dentistry, and Pharmaceutical Sciences; [2]Department of Neurological Surgery, Okayama University Graduate School of Medicine, Dentistry, and Pharmaceutical Science.

Corresponding Author: Atsunori Nakao, MD, PhD, Department of Emergency and Critical Care Medicine, Okayama University Graduate School of Medicine, Dentistry, and Pharmaceutical Sciences, 2-5-1 Shikata-cho, Kita-ku, Okayama, 700-8558, Japan; E-mail: qq-nakao@okayama-u.ac.jp

Received: 04 August 2016
Accepted: 15 October 2016
Published: 01 February 2017

INTRODUCTION

Penetrating intracranial stab injuries caused by metal bars are very rare among ordinary people and often cause severe damage without swift treatment [1]. Penetrating trauma to the face presents a broad spectrum of injuries and treatment challenges due to potential injuries that could occur when the penetrating object is removed.

Immediate complications of transfacial penetrating trauma include cerebral contusion, intracerebral hematoma, pneumocephalus, intraventricular hemorrhage, cranial nerve damage, severe permanent neurological damage, and brain stem and cerebrovascular injury [2]. Delayed complications include cerebrospinal fluid fistula, pneumocephalus, orbital cellulitis, carotid-cavernous sinus fistula, central nervous system

infections, traumatic aneurysm, and delayed intracranial hemorrhage [3].

Computed tomography (CT) scan is usually the first-line radiologic assessment in the emergency room for patients with head injuries. However, artifacts caused by the penetrating object on CT scan often hide the extent of actual brain damage along the trajectory of penetration and treatment of the brain insult may be delayed. Infections can easily complicate penetrating craniocerebral injuries, subdural empyemas, or brain abscesses. Treatment strategies for facial penetrating injury, including prevention and proper management of infectious complications, should be decided with input from multidisciplinary experts such as neurosurgeons, otolaryngologists, and ophthalmologists. This case report may provide guidelines for effective treatment of an intracranial penetrating injury, which may help emergency physicians.

CASE REPORT

A 59-year-old male with a penetrating injury to his face was presented to our department. The patient had fallen from a height of approximately four feet and impaled himself on a metal bar (1 cm in diameter). The iron bar, which had been cut, was still embedded in the right side of his face, approximately 3 cm below his left eye, penetrating his maxillofacial region (Figure 1). The patient did not experience any loss of consciousness and paramedics reported no significant blood loss at the scene. On arrival, his Glasgow Coma Scale score was E4V5M6, the airway was clear, and cervical spine injury was excluded. His vital signs included a heart rate 78 beats/min, blood pressure 142/84 mmHg, respiratory rate 18 breaths/min, and axillary temperature 36.3°C. The pupils were equal in size, round, and reactive to light. His vision was normal bilaterally. No motor or sensory deficits were present on the neurological examination. Computed tomography scan of the brain showed marked artifacts as bright and dark streaks over the trajectory of the metal bar without obvious brain contusion hemorrhage. Therefore, CT scan was again performed after the foreign metallic body was carefully removed with X-ray guided. Right mandibular and sphenoidal bone fracture and intraparenchymal hematoma in the frontal lobe with pneumocephalus caused by direct compaction of the metal bar were noted (Figure 2). Following imaging, the patient became drowsy and was emergently transported to the operating room. Under general anesthesia, emergent craniotomy and hematoma removal was performed 90 minutes after removal of the bar. Broad-spectrum antibiotic treatment with ceftriaxone was started. The patient recovered well from the surgery and the wounds healed without incident or cerebrospinal fluid leak. Ophthalmic examination revealed normal vision 10 days postoperatively. No facial paralysis was noted.

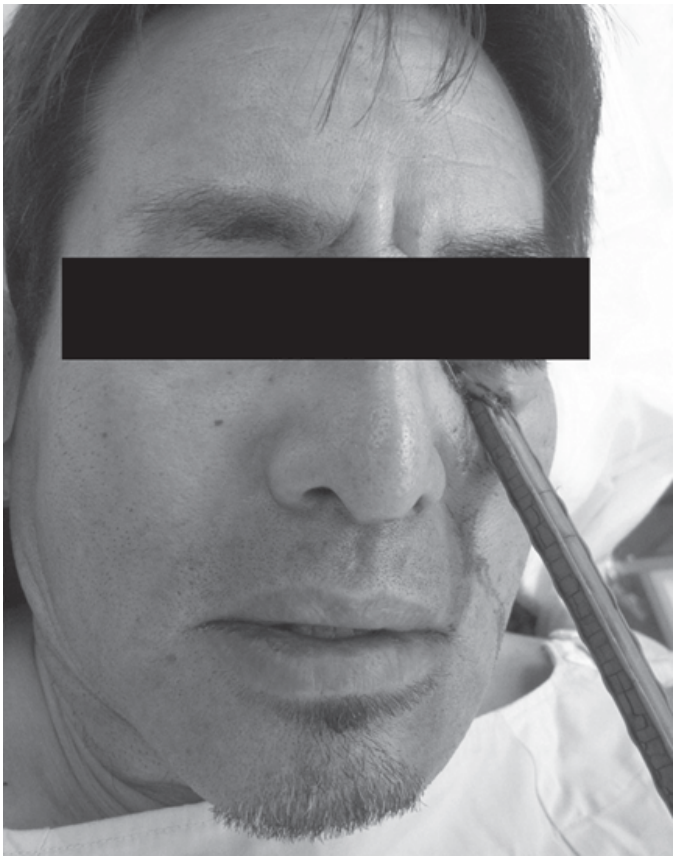


Figure 1: Image of face with the metal bar in situ.

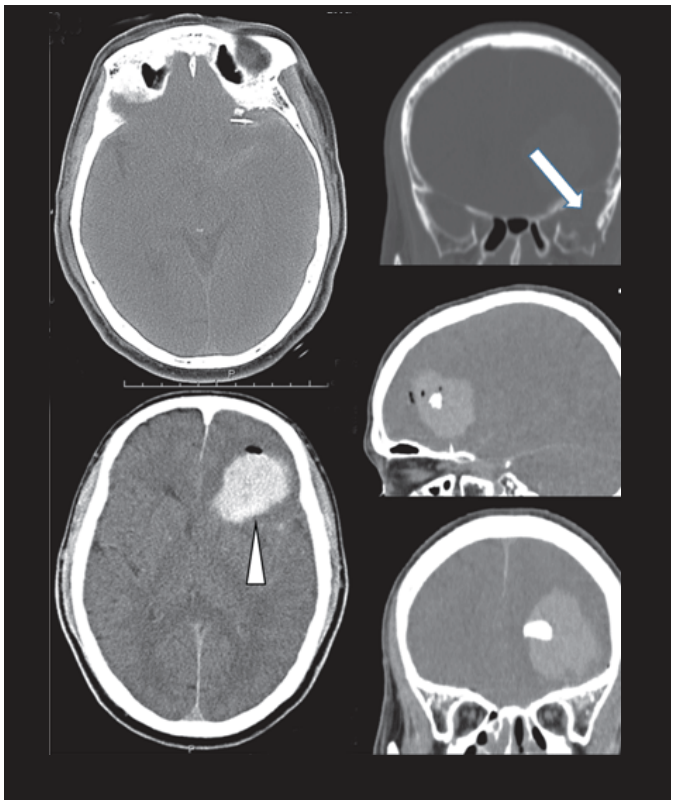


Figure 2: Computed tomography demonstrated right mandibular and sphenoidal bone fracture (white arrow) and intraparenchymal hematoma in the frontal lobe with pneumocephalus (arrowhead).

DISCUSSION

Penetrating craniofacial injury is rare, accounting for 0.4% of all head injuries [4]. Most penetrating brain injuries in civilian emergency practice are missile injuries. Most non-missile penetrating brain injuries are caused by knives, pens, and chopsticks.

The face has protective reflexes that help divert it from oncoming objects. The face has a smaller surface area than the trunk or extremities. Furthermore, the structure of the face and cranium are suited to absorb shocks, owing to the presence of resistant pillars, buttresses, and the presence of pneumatized cavities. Therefore, in general, penetrating facial injuries result in less morbidity to the patient. A penetrating facial injury may not seem serious at first glance, but there may be deeper intracranial injury or injury to the orbit or dura that can be overlooked. The concentrated force of the small area at the tip of the metal bar may enable penetration into the cranial bone. The mechanism of vascular and neuronal injuries caused by cranial stab wounds may differ from that caused by other types of head trauma. Unlike high-energy accidents, no diffuse shearing injury to the brain occurs. Therefore, a patient with intracranial stab wounds usually has a better prognosis than a patient with penetrating injuries caused by an object with a higher kinetic energy [5].

Radiology of the cranium is helpful in evaluating the intracranial course of metallic materials, but the radiodensity of wood is almost the same as that of the brain and soft tissue, so it is difficult to detect.

Although it may be limited for evaluating plastic or wood, head CT scan is the most valuable test for first line evaluation of foreign objects and assessing the extent of injury. However, in many cases, optimal evaluation cannot be performed on cerebral CT scan due to the presence of artifacts caused by the object.

Angiography is advocated by some authors for possible cerebral vascular injuries in patients after penetrating head injuries to rule out unexpected vascular lesions. Arteriovenous fistulae and traumatic aneurysms are the most important findings, since they are seldom clinically evident. Delaying carotid angiography until the start of the second week has previously been proposed to allow for better visualization of these complications. However, traumatic aneurysms can burst at any time post injury, and the mortality from a second hemorrhage is unacceptably high [6, 7].

Management of patients with penetrating brain injuries should follow fundamental surgical principles, including removing the object under direct vision in order to reduce further brain tissue damage by the foreign body catching on the bone fragments. The most appropriate management in the pre-hospital environment is to leave the penetrating object in situ and move the patient to the trauma center. Key strategies for emergency physicians and medical technicians are to stabilize the object to prevent movement and conduct serial neurological examinations. Having a systematic method to evaluate and manage such injuries is important. Following removal of the foreign body, thorough debridement with removal of all involved skull bone and retained fragments, evacuation of the hematoma followed by careful hemostasis along the trajectory, and meticulous dural closure to reduce the chance of cerebrospinal fluid fistula are mandatory. Prevention of infection needs to be addressed. Patients should be given tetanus shots and preoperative and postoperative antibiotics, if indicated. Some reports suggest that although penetrating craniofacial injuries with a wooden bar, which can affect extensively by broken fragments, usually require surgical intervention, injuries with a metal bar can be managed without surgical intervention with good outcomes [8].

It is imperative to leave the foreign body in place until adequate preoperative planning has been completed and subsequent removal should only be attempted under direct visualization in an operating room. Premature removal without adequate preparation can put the patient at risk for further injury or fatal bleeding if the object has caused a tamponade of the bleeding [9]. Our patient presented with corresponding brain hemorrhages with a great mass obscured on the follow-up CT scan, indicating that bleeding occurred after removal of the object. Based on this experience, we recommend immediate surgical decompression with early follow-up CT to ascertain the extent of brain damage and monitor patients for possible delayed events.

Prognosis depends on the affected brain area and the involvement of major vessel injuries. Complications and sequelae after penetrating brain injury are including life-threatening hemorrhage, local trauma to the brain and its vasculature, brain abscesses, meningitis, *cerebrospinal fluid l*eakage, and neurological deficits. Epilepsy, behavioral changes, and psychological issues could be attributed to frontal lobe injury [10]. In our patient, proper assessment of the injury and management decisions were made with input from a multidisciplinary team of clinicians in neurosurgery, ophthalmology, otolaryngology and plastic surgery.

CONCLUSION

Transorbital brain injury caused by a metal bar is an uncommon but potentially fatal event unless managed appropriately. Computed tomography scan of the brain showed marked artifacts over the trajectory of the metal bar. Emergent surgical intervention and early follow-up computed tomography scans are necessary to have any chance to save the patient's life.

****

Author Contributions

Tetsuya Yumoto – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising

it critically for important intellectual content, Final approval of the version to be published
Atsuyoshi Iida – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published
Kohei Tsukahara – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published
Hiromichi Naito – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published
Michihisa Terado – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published
Keiji Sato – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published
Isao Date – Substantial contributions to conception and design, Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published
Atsunori Nakao – Substantial contributions to conception and design, Acquisition of data, or analysis and interpretation of data, Drafting the article or revising it critically for important intellectual content, Final approval of the version to be published

Guarantor
The corresponding author is the guarantor of submission.

Conflict of Interest
Authors declare no conflict of interest.

Copyright
© 2017 Tetsuya Yumoto et al. This article is distributed under the terms of Creative Commons Attribution License which permits unrestricted use, distribution and reproduction in any medium provided the original author(s) and original publisher are properly credited. Please see the copyright policy on the journal website for more information.

REFERENCES

1. Lin HL, Lee HC, Cho DY. Management of transorbital brain injury. J Chin Med Assoc 2007 Jan;70(1):36–8.
2. Miscusi M, Arangio P, De Martino L, De-Giorgio F, Cascone P, Raco A. An unusual case of orbito-frontal rod fence stab injury with a good outcome. BMC Surg 2013 Aug 13;13:31.
3. Kitakami A, Kirikae M, Kuroda K, Ogawa A. Transorbital-transpetrosal penetrating cerebellar injury: Case report. Neurol Med Chir (Tokyo) 1999 Feb;39(2):150–2.
4. Gennarelli TA, Champion HR, Sacco WJ, Copes WS, Alves WM. Mortality of patients with head injury and extracranial injury treated in trauma centers. J Trauma 1989 Sep;29(9):1193–201; discussion 1201–2.
5. Caldicott DG, Pearce A, Price R, Croser D, Brophy B. Not just another 'head lac'...low-velocity, penetrating intra-cranial injuries: A case report and review of the literature. Injury 2004 Oct;35(10):1044–54.
6. du Trevou MD, van Dellen JR. Penetrating stab wounds to the brain: The timing of angiography in patients presenting with the weapon already removed. Neurosurgery 1992 Nov;31(5):905–11; discussion 911–2.
7. Holmes B, Harbaugh RE. Traumatic intracranial aneurysms: A contemporary review. J Trauma 1993 Dec;35(6):855–60.
8. Park SH, Cho KH, Shin YS, et al. Penetrating craniofacial injuries in children with wooden and metal chopsticks. Pediatr Neurosurg 2006;42(3):138–46.
9. Schreckinger M, Orringer D, Thompson BG, La Marca F, Sagher O. Transorbital penetrating injury: Case series, review of the literature, and proposed management algorithm. J Neurosurg 2011 Jan;114(1):53–61.
10. Bodwal J, Sreenivas M, Aggrawal A. Intracranial penetrating injury by screw driver: A case report and review of literature. J Forensic Leg Med 2013 Nov;20(8):972–5.

Access full text article on other devices

Access PDF of article on other devices

CASE REPORT PEER REVIEWED | OPEN ACCESS

Chronic renal failure secondary to diabetes mellitus

Mustafa Z. Mahmoud, Omer A. Mahmoud, Maram A. Fagiri

ABSTRACT

Introduction: Diabetes mellitus is the most common cause of renal failure. Even when diabetes is controlled, the disease can lead to chronic renal failure (CRF). A patient with chronic renal failure always undergoes either dialysis or renal transplantation, which both are very expensive financially. Testing in patients with CRF typically includes a complete blood count (CBC), basic metabolic panel, and urinalysis, with calculation of renal function. Renal ultrasonography is the initial imaging modality in the diagnosis of CRF, where features of atrophied, echogenic kidneys with poor corticomedullary differentiation always observed. The aim of this case report is to focus on the role of ultrasound imaging in the workup of chronic renal failure. Case Report: A 48-year-old male, with 22 years history of type 2 diabetes mellitus complains of CRF primarily due to diabetic nephropathy, was admitted to the hospital for dialysis. The patient had been undergoing hemodialysis three times per week. On physical examination he was in a fair condition. Laboratory investigations revealed an increased level of creatinine 6.9 mg/dl (normal value <1.5 mg/dl) and blood urea nitrogen (BUN) 49 mg/dl (normal value 10–20 mg/dl) were noted. Normal levels for sodium 140 mg/dl (normal value 136–145 mg/dl) was detected, but there was an increased level of potassium 7 mg/dl (normal value 3.5–5 mg/dl), calcium 11.9 mg/dl (normal value 9–10.5 mg/dl), and phosphorus 5.8 mg/dl (normal value 3–4.5 mg/dl). Abdominal ultrasound scanning presented sonographic features compatible with CRF as bilateral renal atrophy, poor corticomedullary differentiation, and increased renal echogenicity. Conclusion: Morphological parameters as bilateral renal size, parenchymal thickness, and renal echogenicity can influence further diagnostic and therapeutic interventions of CRF.

Keywords: Blood urea nitrogen, Chronic renal failure, Creatinine, Diabetes mellitus, Ultrasonography

Mustafa Z. Mahmoud[1], Omer A. Mahmoud[2], Maram A. Fagiri[3]

Affiliations: [1]PhD, Associate Professor of Radiology and Ultrasonography, Radiology and Medical Imaging Department, College of Applied Medical Sciences, Prince Sattam bin Abdulaziz University, Al-Kharj, Al-Kharj, Saudi Arabia; [2]MSc, Specialist of Medical Ultrasound Imaging, Medical Ultrasound Imaging department, Dr. Mohamed Abdel Mageed Ali Medical Complex, Alnohood, West of Kordofan, Sudan; [3]MSc, Lecturer of Diagnostic Radiologic Technology Science, Radiology and Medical Imaging Department, College of Applied Medical Sciences, Prince Sattam bin Abdulaziz University, Al-Kharj, Al-Kharj, Saudi Arabia.

Corresponding Author: Mustafa Z. Mahmoud, Radiology and Medical Imaging Department, College of Applied Medical Sciences, Prince Sattam bin Abdulaziz University, Al-Kharj, Al-Kharj, Saudi Arabia 11942; Email: m.alhassen@psau.edu.sa

Received: 05 October 2016
Accepted: 10 November 2016
Published: 01 February 2017

How to cite this article

Mahmoud MZ, Mahmoud OA, Fagiri MA. Chronic renal failure secondary to diabetes mellitus. Int J Case Rep Images 2017;8(2):124–128.

Article ID: Z01201702CR10761MM

****

doi:10.5348/ijcri-201722-CR-10761

INTRODUCTION

Diabetes mellitus has become the primary cause of end stage renal disease (ESRD) in the United States, and the incidence of type 2 diabetes mellitus continues to grow in the United States and worldwide [1, 2]. Type 2 diabetes has a more variable course. Patients often present at diagnosis with microalbuminuria because of delays in diagnosis and other factors affecting protein excretion. Some patients with microalbuminuria progressing to advanced renal disease. Without intervention, approximately 30% progress to overt nephropathy and, after 20 years of nephropathy, approximately 20% develop ESRD. Differential diagnosis of diabetic nephropathy is usually based on the history, physical examination, laboratory evaluation, and imaging of kidneys [2, 3]. An important step in the screening and diagnosis of diabetic nephropathy is to measure albumin in a spot urine sample, collected either as the first urine in the morning or at random. Screening should not be performed in the presence of conditions that increase urinary albumin excretion, such as urinary tract infection with the presence of hematuria, febrile illness, hyperglycemia, hypertension, heart failure, and after exercise [4].

Ultrasound has become an ideal imaging test for chronic renal failure (CRF) as well as a valuable tool in nephrology because of its safety, simplicity and low cost, as well as the ease visualization of the kidneys. In ultrasonography, finding an atrophied kidney with a thin, echogenic parenchyma or cortex indicates irreversible damage, and thus helps to avoid any further unnecessary workup, biopsy, immunosuppressive therapy, and allows for the optimal planning for renal replacement therapy [5, 6]. Increased renal echogenicity can be the consequence of not only sclerosis, but also of infiltration as well. The size of the kidneys varies with body size, which should be taken into account when diagnosing irreversible renal damage on the basis of kidney size and echogenicity [6]. In CRF, Doppler ultrasonography of intrarenal vessels can provide additional information about microvascular and parenchymal lesions, which is helpful in deciding for or against therapeutic intervention and timely planning for optimal renal replacement therapy option [7].

CASE REPORT

A 48-year-old male, with 22 years history of type 2 diabetes mellitus that require insulin, and with complications of CRF primarily due to diabetic nephropathy, was admitted to the hospital for dialysis. Since last year, he had been undergoing hemodialysis three times via an arteriovenous fistula for 2.5–3 hours for each time. His last dialysis was four days before the day of the admission.

On physical examination the patient was in a fair condition, with no related distress, hydrate, and well oriented. The patient's blood pressure was 129/86 mmHg, heart rate was 79 beats per minute, body temperature was 36.9°C, and respiratory rate was 15 breaths/min.

Due to the patient history of CRF secondary to diabetic nephropathy, laboratory investigation blood testing revealed that a hemoglobin of 14.1 g/dl (normal value 13.5–17.5 g/dl), platelets of $290\times10^3/mm^3$ (normal value $150–350\times10^3/mm^3$), leukocytes of $9\times10^3/mm^3$ (normal value $4.5–11\times10^3/mm^3$), neutrophils of 65% (normal value 40–70%), and lymphocytes of 32% (normal value 22–44%). An elevated levels of creatinine 6.9 mg/dl (normal value <1.5 mg/dl) and blood urea nitrogen (BUN) 49 mg/dl (normal value 10–20 mg/dl) were noted. Normal levels for sodium 140 mg/dl (normal value 136–145 mg/dl) was detected, but there was an increased level of potassium 7 mg/dl (normal value 3.5–5 mg/dl), calcium 11.9 mg/dl (normal value 9–10.5 mg/dl), and phosphorus 5.8 mg/dl (normal value 3–4.5 mg/dl). Urine analysis demonstrates yellow urine, with a urine pH of 7 (normal value 4.5-8), and protein of 136 mg/d (normal value ≤ 150 mg/d). Also, there was no bacteria, yeasts, crystals, and RBCs were detected during the urine analysis.

Abdominal ultrasound scanning presented both kidneys with decreased length and width measurements (Figure 1); atrophied (right kidney of 8.41×3.91 cm and left kidney of 8.06×3.34 cm for both length and width respectively), poor corticomedullary differentiation (Figure 2), and increased renal echogenicity; hyperechoic (Figure 3).

The patient was discharged from the hospital after completing his regular check-up, and was also recommended to continue with hemodialysis, waiting for the availability of donor for kidney transplant.

DISCUSSION

This case report described a condition of CRF developed in a patient with 22 years history of type 2 diabetes mellitus that require insulin. Chronic renal failure has become a global epidemic, and one of the major causes of CRF is diabetes. Patients with diabetes and nephropathy commonly exhibit concurrent diabetic neuropathy [8]. Diabetic nephropathy is the most common cause of CRF. As stated more than 43.3% of new patients undergoing renal transplantation treatment program in the USA are mostly of type 2 diabetes mellitus [9]. As the progress of diabetic nephropathy, kidneys are shrinking in size and a progressive loss of renal function is occurring [7]. The prediction of renal function irreversibility in CRF has been often difficult on the basis of renal length or thickness of the renal parenchyma. Also, about 20% of diabetes mellitus patients can develop CRF as a result of non-diabetic renal pathologies, thus ultrasonography had the ability to show all the characteristics of CRF, as reduced in renal size and the atrophy of the renal parenchyma [10].

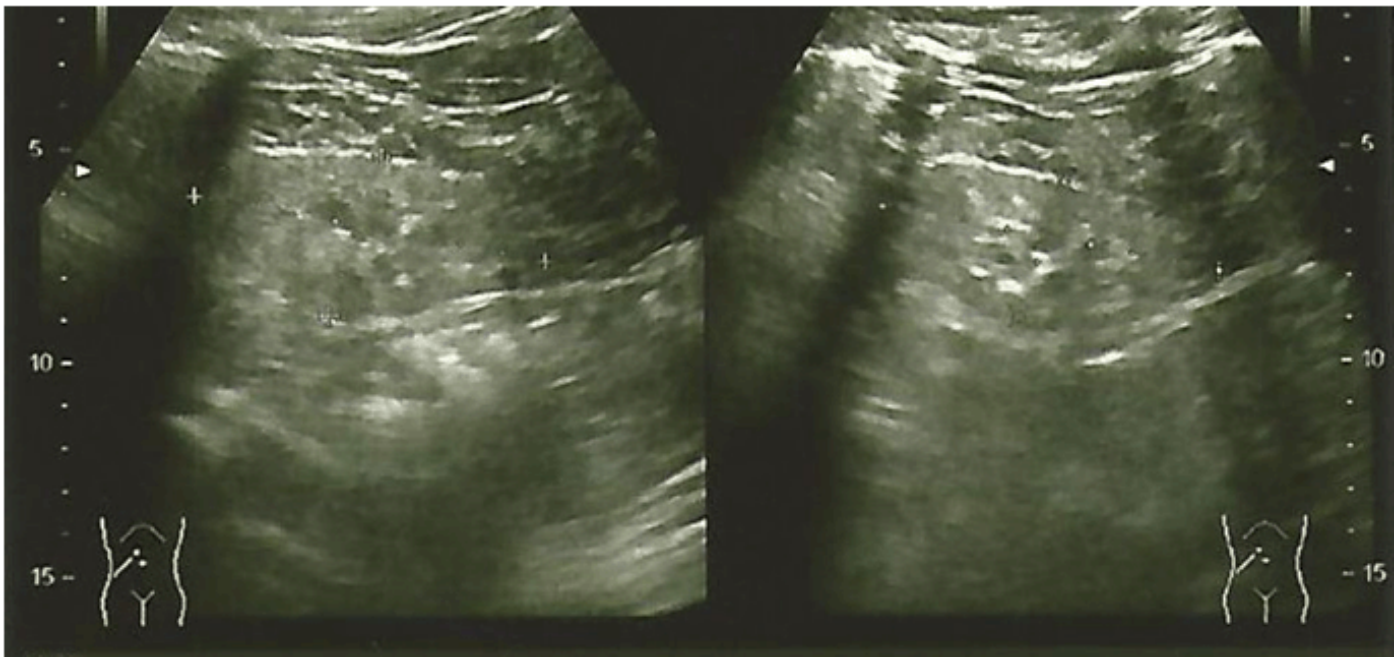


Figure 1: Gray scale ultrasound image on the right and left kidneys, where both kidneys were decreased in size; atrophied secondary to diabetic nephropathy.

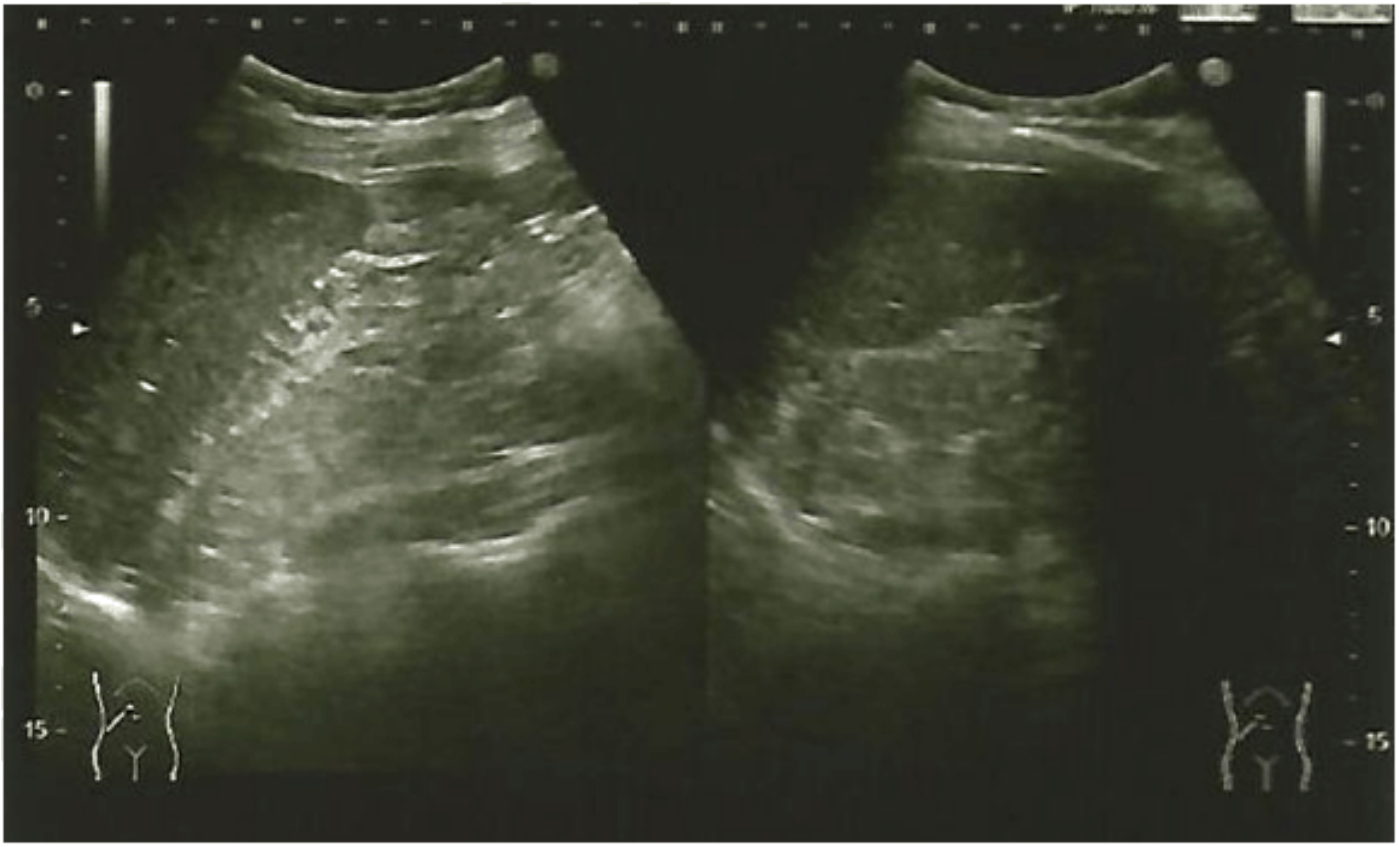


Figure 2: Chronic renal failure, were kidneys presented with poor corticomedullary differentiation. Note that the lower pole of the left kidney is obliterated by ribs shadow.

Blood tests as blood urea nitrogen (BUN) and serum creatinine are the simplest way to monitor renal function. A test can be done to measure the amount of urea nitrogen in the blood. In kidney disease, these substances are not excreted normally, and so they accumulate in the body, thus causing an increase in blood levels of urea [11]. The

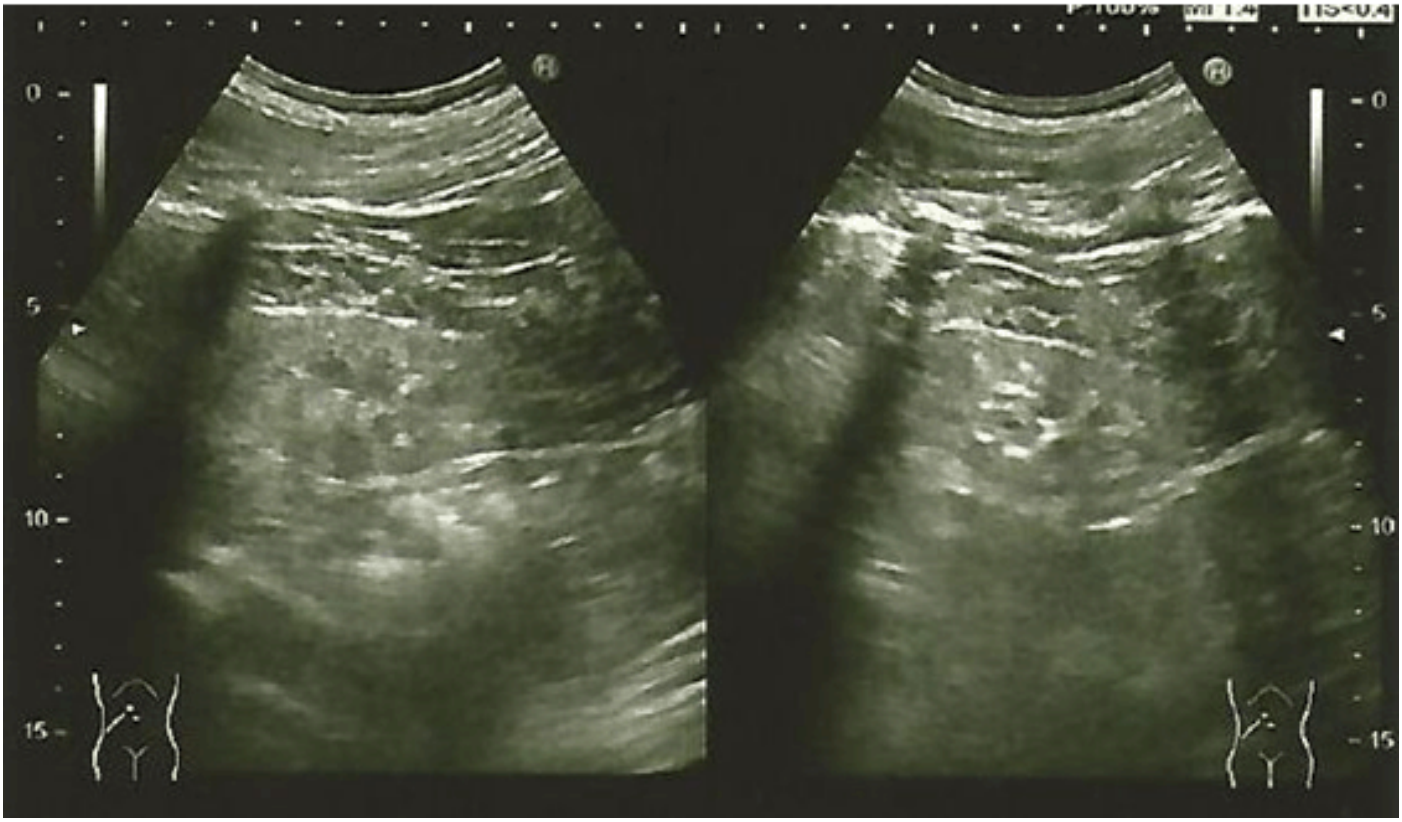


Figure 3: Kidneys affected with chronic renal failure and presented an increase of renal echogenicity; hyperechoic.

development of CRF as a consequence of diabetes mellitus was found to be related to disorders of vasodilatation and metabolic abnormalities that mediated by endothelial derived nitric oxide. Angiotensin II and aldosterone, interacting with pulse pressure and increased systolic blood pressure, activate NADPH oxidase, which acts as mediator of oxidative stress. Angiotensin II increases metabolism of nitric oxide to peroxynitrite, which further impairs endothelial-derived vasodilation [12]. In another mechanism, decrease in the ability to produce endothelial progenitor cells (EPCs), which derived from bone marrow, play a role in replacing damaged endothelium and are reduced in people with decreased endothelium-dependent vasodilation [13].

Ultrasonography examinations usually concern the ability to identify a pathological condition, to distinguish between different histopathological lesions, and to identify patients with CRF. The ultrasound brightness mode (B-mode) depends on morphological parameters to diagnose incidence of CRF. These morphological parameters are interpolar diameter, parenchymal thickness, and renal echogenicity. The right kidney mean length is 10.74±1.35 cm and the mean length of the left kidney is 11.10±1.15 cm, measured on a posterior oblique image as the longest diameter, with a lower limit of normality indicated as 9 cm. Where a renal length less than 8 cm is attributed to CRF. Also parenchymal thickness less than 15 mm and echogenicity identical to that of the renal sinus are strongly suggested a CRF condition [14].

CONCLUSION

B-mode ultrasonography is a valuable diagnostic tool and often required for the diagnostic workup of patients with chronic renal failure. Accurate description of bilateral renal size, parenchymal thickness, and renal echogenicity are needed because they can influence further diagnostic and therapeutic interventions in such malady.

****

Acknowledgements

We would like to thank the staff of the Radiology and Medical Imaging Department, College of Applied Medical Sciences, Prince Sattam bin Abdulaziz University for their continuous support during writing this case report.

Author Contributions

Mustafa Z. Mahmoud – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Omer A. Mahmoud – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Maram A. Fagiri – Substantial contributions to conception and design, Acquisition of data, Analysis

and interpretation of data, Drafting the article, revising it critically for important intellectual content, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

Copyright

© 2017 Mustafa Z. Mahmoud et al. This article is distributed under the terms of Creative Commons Attribution License which permits unrestricted use, distribution and reproduction in any medium provided the original author(s) and original publisher are properly credited. Please see the copyright policy on the journal website for more information.

REFERENCES

1. United States Renal Data System: Incidence of reported end-stage renal disease. [Available at: http://www.usrds.org/2008/ref/A_Incidence_08.pdf]
2. Hall PM. Prevention of progression in diabetic nephropathy. Diabetes Spectrum 2006;19(1):18–24.
3. Christensen PK, Larsen S, Horn T, Olsen S, Parving HH. Renal function and structure in albuminuric type 2 diabetic patients without retinopathy. Nephrol Dial Transplant 2001 Dec;16(12):2337–47.
4. Mogensen CE, Vestbo E, Poulsen PL, et al. Microalbuminuria and potential confounders. A review and some observations on variability of urinary albumin excretion. Diabetes Care 1995 Apr;18(4):572–81.
5. O'Neill WC. Sonographic evaluation of renal failure. Am J Kidney Dis 2000 Jun;35(6):1021–38.
6. O'Neill WC. Chronic renal failure. In: O'Neill WC ed. Atlas of Renal Ultrasonography, 1ed. Philadelphia: W.B. Saunders Company; 2001. p. 41–3.
7. Buturovic-Ponikvar J, Visnar-Perovic A. Ultrasonography in chronic renal failure. Eur J Radiol 2003 May;46(2):115–22.
8. Wen CP, Cheng TY, Tsai MK, et al. All-cause mortality attributable to chronic kidney disease: A prospective cohort study based on 462 293 adults in Taiwan. Lancet 2008 Jun 28;371(9631):2173–82.
9. Ritz E, Rychlík I, Locatelli F, Halimi S. End-stage renal failure in type 2 diabetes: A medical catastrophe of worldwide dimensions. Am J Kidney Dis 1999 Nov;34(5):795–808.
10. Remuzzi G, Schieppati A, Ruggenenti P. Clinical practice. Nephropathy in patients with type 2 diabetes. N Engl J Med 2002 Apr 11;346(15):1145–51.
11. Dabla PK. Renal function in diabetic nephropathy. World J Diabetes 2010 May 15;1(2):48–56.
12. Coresh J, Astor BC, Greene T, Eknoyan G, Levey AS. Prevalence of chronic kidney disease and decreased kidney function in the adult US population: Third National Health and Nutrition Examination Survey. Am J Kidney Dis 2003 Jan;41(1):1–12.
13. Hill JM, Zalos G, Halcox JP, et al. Circulating endothelial progenitor cells, vascular function, and cardiovascular risk. N Engl J Med 2003 Feb 13;348(7):593–600.
14. Fiorini F, Barozzi L. The role of ultrasonography in the study of medical nephropathy. J Ultrasound 2007 Dec;10(4):161–7.

Access full text article on other devices

Access PDF of article on other devices

CASE REPORT PEER REVIEWED | OPEN ACCESS

A case of inversely fused tooth of impacted maxillary third molar and supernumerary tooth

Eiji Mitate, Shintaro Kawano, Yurie Mikami, Tamotsu Kiyoshima, Tetsuro Ikebe, Seiji Nakamura

ABSTRACT

We report an extremely rare case of an impacted maxillary third molar inversely fused with a supernumerary tooth in a 51-year-old male. The panoramic and dental radiographs show a radiopaque and tooth-like mass of 20×15 mm located in the third maxillary region of the right maxilla. Computed tomography revealed that the tooth-like structure was a union of the impacted maxillary third molar with a inversed supernumerary tooth, creating a fused tooth with a common dental pulp. To our knowledge, only one case of inversely fusion of a maxillary third molar with a supernumerary tooth has been reported.

Keywords: Anomaly. Impacted third molar, Inversed fused tooth, Maxilla, Supernumerary tooth

Eiji Mitate[1,2], Shintaro Kawano[2], Yurie Mikami[2], Tamotsu Kiyoshima[3], Tetsuro Ikebe[1], Seiji Nakamura[2]

Affiliations: [1]Section of oral surgery, Department of Oral and Maxillofacial Surgery, Fukuoka Dental College, 2-15-1, Tamura, Sawara-ku, Fukuoka 814-0175, Japan; [2]Section of Oral and Maxillofacial Oncology, Division of Maxillofacial Diagnostic and Surgical, Sciences, Faculty of Dental Science, Kyushu University, 3-1-1, Maidashi, Higashi-ku, Fukuoka 812-8582, Japan; [3]Laboratory of Oral Pathology, Division of Maxillofacial Diagnostic and Surgical Sciences, Faculty of Dental Science, Kyushu University, 3-1-1, Maidashi, Higashi-ku, Fukuoka 812-8582, Japan.

Corresponding Author: Eiji Mitate, DDS, PhD, Section of oral surgery, Department of Oral and Maxillofacial Surgery, Fukuoka Dental College, 2-15-1, Tamura, Sawara-ku, Fukuoka 814-0175, Japan; Email: mitate@college.fdcnet.ac.jp; mitate@dent.kyushu-u.ac.jp

Received: 11 October 2016
Accepted: 16 November 2016
Published: 01 February 2017

How to cite this article

Mitate E, Kawano S, Mikami Y, Kiyoshima T, Ikebe T, Nakamura S. A case of inversely fused tooth of impacted maxillary third molar and supernumerary tooth. Int J Case Rep Images 2017;8(2):129–132.

Article ID: Z01201702CR10762EM

****

doi:10.5348/ijcri-201723-CR-10762

INTRODUCTION

Fused teeth result from the union of two or more tooth germs in the developing stage. This is common in the lower anterior, but rare in maxilla, and found in both sexes [1]. They are more commonly found in the deciduous dentition than in the permanent dentition. The reported incidence rate ranges from 0.5–2.5% [1]. But the etiology is still unknown. We report an extremely rare case of an impacted maxillary third molar inversely fused with a supernumerary tooth.

CASE REPORT

A 51-year-old male patient visited oral and maxillofacial surgery of Kyushu University Hospital complaining of slight pain in the right maxillary molar region. Clinical examination revealed slight percussion pain on maxillary tuberosity, but with no sign of infection such as swelling and redness of the gingival mucosa, fistula formation, and pus discharge in the right maxilla. The right maxillary third molar was not erupted. First, we take a panoramic X-ray for overview, and dental X-ray for detail. The panoramic and dental radiographs showed a radiopaque mass (20x15 mm) located in the third

maxillary molar region of the right maxilla (Figure 1A–B). To detect its shape and location, we took computed tomography (CT). Computed tomography scan revealed a tooth-like mass composed of an enamel-like and a dentin-like region, and dental pulp-like cavities near arteria palatina major (Figure 1C–D). Furthermore, the mass consisted of two tooth-like structures that shared a dental pulp cavity. The lesion was partially projecting into the right maxillary sinus (Figure 1D). The clinical diagnosis of the lesion was odontoma or fused tooth and slight pericoronitis may cause slight pain. As he had slight pain, removal of the mass through the canine fossa of maxilla was planed. From the point of view of operation time and procedure, this operation was performed under general anesthesia. The mass was divided into two pieces at removal, because the size of the mass was larger than that of the canine fossa aperture. The extirpated mass is shown in Figure 2.

DISCUSSION

Levitas [2] suggested that dental malformation might be classified as gemination, twinning, concrescence, and fusion. A fused tooth is defined as one in which there is union of dentin from two separate tooth germs during development. On the other hand, occurrence of two teeth with cementum union indicates concrescence. In this case, the mass consisted of two tooth-like structures that shared one dental pulp cavity, as revealed by CT scan. This mass was found to result from an inversely fusion of the third molar with a supernumerary tooth because the mass was located at the site of the third maxillary molar region and since the number of erupted teeth was normal.

With respect to fusion of the third molar and the supernumerary tooth, a number of reports in English scientific literature have described this occurrence primarily in the mandible [3–7], but there is only one report of a fused tooth in the maxilla [8]. On the other hand, we found many reports in Japanese scientific literature. For example, Uchida [8] reviewed 46 cases of supernumerary teeth fused with maxillary or mandibular third molars reported between 1934 and 2008 in the Japanese literature. Of the 46 cases (24 males and 22 females), 12 were in the right maxilla and 10 were in the left maxilla. However, it was unclear whether these included a case of inversely fused tooth. Nakanishi [9] reported that the prevalence of supernumerary tooth is almost 1% in all regions of the dentition, and only 0.063% in the upper molar region. This suggests that the frequency of fusion of the maxillary third molar and supernumerary tooth may be even lower. These facts, considered together, indicate the extreme rarity of this case. Sugibayashi [10] proposed that the causes of supernumerary teeth might be heredity, physical forces, and/or trauma in the tooth germ. Some cases of fusion between maxillary third molar and supernumerary tooth have been reported in English literature. The association of hereditary, racial, or environmental factors with the pathogenesis of this anomaly may not have been explored in detail.

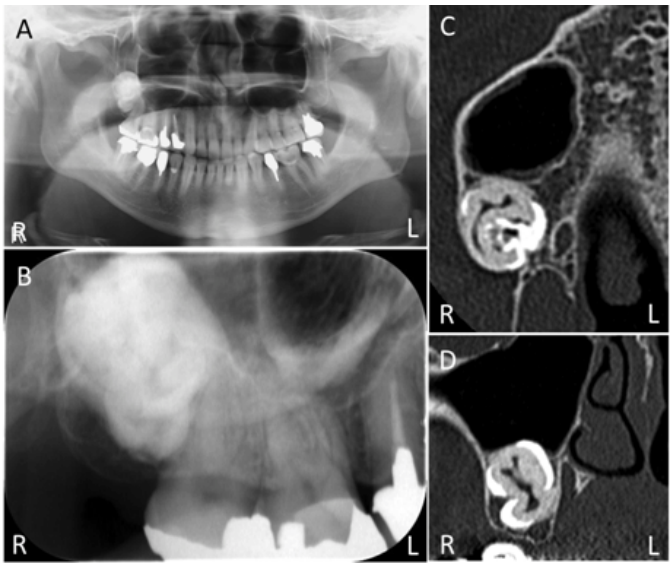


Figure 1: Radiography and computed tomography (A) Panoramic, (B) Dental radiography showing a radiopaque mass is located in the right maxilla, (C) Transverse plane of computed tomography images. The radiopaque mass is located in the right maxillary sinus. Some cavities can be seen in the mass, and (D) Coronal plane of CT images reveals that the radiopaque mass has pulp-like cavity. (R: right side, L: left side)

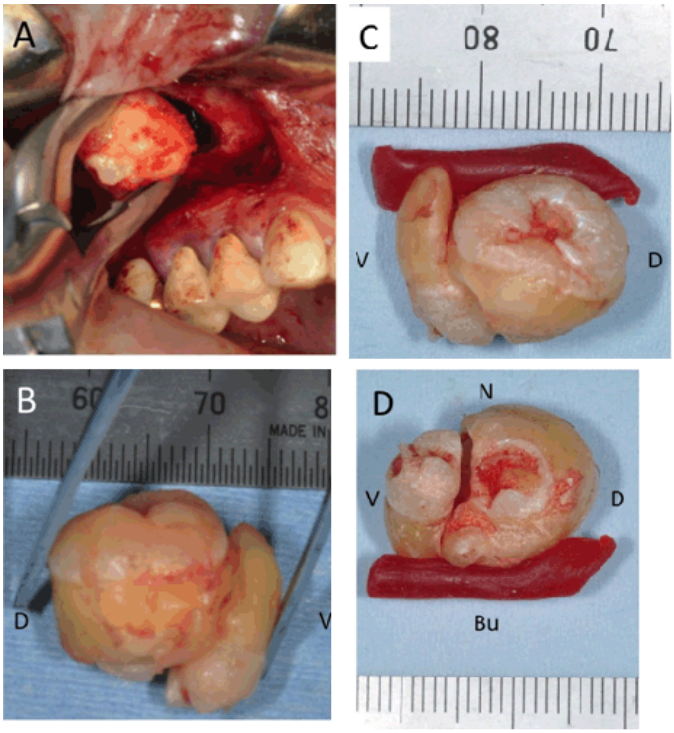


Figure 2: Operation procedure (A) and extracted material (fixed with dental utility wax) (B) Buccal side, (C) Nasal side, (D) Caudal side Bu: buccal side, N: nasal side, V: ventral side, D: dorsal side. The mass was 18x12x10 mm with enamel-like region and dentin-like region.

About the treatment, most of the cases were extracted with local anesthesia. These kinds of cases were found after eruption. In this case, fused teeth were fully impacted, and located near arteria palatina major and pterygoid plexus. As we have to remove more carefully, operation under local anesthesia is intolerable.

CONCLUSION

We reported a rare case of inversely fusion of impacted maxillary third molar and supernumerary tooth. In previous reports, the reason may be hereditary, racial, or environmental factors and so on. To remove the fused teeth, we have to take better procedure of removal and anesthesia.

****

Author Contributions

Eiji Mitate – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Shintaro Kawano – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Yurie Mikami – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Tamotsu Kiyoshima – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Tetsuro Ikebe – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Seiji Nakamura – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

Copyright

© 2017 Eiji Mitate et al. This article is distributed under the terms of Creative Commons Attribution License which permits unrestricted use, distribution and reproduction in any medium provided the original author(s) and original publisher are properly credited. Please see the copyright policy on the journal website for more information.

REFERENCES

1. Hou GL, Tsai CC. Fusion of maxillary third and supernumerary fourth molars. Aust Dent J 1989 Jun;34(3):219–22.
2. Levitas TC. Germination Fusion Twinning and Concrescence. ASDC J Dent Child 1965;32:93–100.
3. Morris DO. Fusion of mandibular third and supernumerary fourth molars. Dent Update 1992 May;19(4):177–8.
4. Conte M, Lombardi P, Linfant J. A supernumerary tooth fused to the distal surface of a mandibular third molar. J N J Dent Assoc 2002 Winter-Spring;73(1–2):8–9.
5. Goho CD Jr. Mandibular third molar and supernumerary molar fusion. Va Dent J 1988 Oct-Dec;65(4):26–7.
6. Ozcan G, Sekerci AE, Soylu E, Nazlim S, Amuk M, Avci F. Role of cone-beam computed tomography in the evaluation of a paradental cyst related to the fusion of a wisdom tooth with a paramolar: A rare case report. Imaging Sci Dent 2016 Mar;46(1):57–62.
7. Abrams RA, Nelson DL. Fusion of a third molar to a supernumerary tooth with an associated dentigerous cyst. Aust Dent J 1979 Jun;24(3):141–2.
8. Uchida K, Kuroiwa H, Uchiyama M, et al. A case of fusion of the supernumerary teeth in the right maxilla and the third molar. Matsumoto Shigaku 2010;36:134–8.
9. Nakanishi H, Nomura J, Yanase S, et al. A case of fused tooth consisted of the maxillary third molar and supernumerary tooth with review of the Japanese literature. Jpn J Oral Diag/Oral Med 2004;17(2):217–20.
10. Sugibayashi Y, Inoue K, Yamazaki Y, Arai T, Sato T. Fusion of maxillary third and supernumerary fourth molars: A histological analysis. Tsurumi Univ Dent J 2003;29(2):201–8.

Access full text article on other devices

Access PDF of article on other devices

CASE REPORT PEER REVIEWED | OPEN ACCESS

Intralobar pulmonary sequestration: A rare presentation in adults

Ahmed Ehab, Marwa Ghanem, Ahmed Ibrahim Twafik, Magda Abdel-salam, Nasef Abdel-Salam A. Rezk, Asem A. Hewidy

ABSTRACT

Introduction: Pulmonary sequestration represents a rare congenital anomaly of the lower respiratory tract. Interlobar sequestration is the most common form and usually presents in the left hemithorax during the second decade of life or earlier. Its blood supply usually arises from systemic circulation. Surgical intervention is the treatment of choice in patients with pulmonary sequestration. Case Report: We describe a late presentation of an intralobar sequestration in a 59-year-old male. It presented with recurrent attack of hemoptysis. Radiological investigations revealed pulmonary sequestration. Two unusual features were found in this case: being in the right side and having double blood supply from both systemic and pulmonary circulation. Conclusion: In this case report, we described a case of unusual presentation of ILS in old patients, right sided with double blood supply from both pulmonary artery and abdominal aorta.

Keywords: Intralobar pulmonary sequestration, Lung anomalies, Pulmonary angiography

Ahmed Ehab[1], Marwa Ghanem[1], Ahmed Ibrahim Twafik[2], Magda Abdel-salam[1], Nasef Abdel-Salam A. Rezk[1], Asem A. Hewidy[1]

Affiliations: [1]Department of Chest Medicine, Faculty of Medicine, Mansoura University, Egypt; [2]Department of Diagnostic and Interventional Radiology Faculty of Medicine, Mansoura University, Egypt.

Corresponding Author: Ahmed Ehab, Chest Medicine Department, Mansoura University Hospital, Elgomhoria Street, Mansoura Egypt, 35516; E-mail: dr.a.ehba@gmail.com

Received: 18 August 2016
Accepted: 26 October 2016
Published: 01 February 2017

How to cite this article

Ehab A, Ghanem M, Twafik AI, Abdel-Salam M, Abdel-Salam N, A. Rezk NA, Hewidy AA. Intralobar pulmonary sequestration: A rare presentation in adults. Int J Case Rep Images 2017;8(2):133–137.

Article ID: Z01201702CR10763AE

****

doi:10.5348/ijcri-201724-CR-10763

INTRODUCTION

Pulmonary sequestration is a rare congenital malformation of the lower respiratory tract. It defined as a nonfunctioning primitive solid or cystic aberrant lung mass which has no connection with tracheobronchial tree and has systemic arterial supply [1]. Intralobar sequestration (ILS) is overall the most common form. In 50–60% of cases, the diagnosis of ILS is made during the second decade or younger. Later presentation is rare [2].

Surgical excision is the treatment of choice of pulmonary sequestration. The preoperative assessment includes besides confirmation of the diagnosis the proper identification of the vascular supply of the sequestartion [3].

We describe intralobar pulmonary seqestration in a male patient 59-year-old, who presented with recurrent attacks of hemoptysis. The discovered ILS was abnormally supplied with a double blood supply.

CASE REPORT

A 59-year-old male current smoker presented with recurrent attacks of hemoptysis. The first attack started 12 years ago, after that he experienced recurrent attacks

of blood-tinged sputum (average 3 attacks per year). The patient was first investigated in the primary health care facility where chest X-ray (Figure 1) was done and was interpreted as a case of bronchitis despite the obvious right para cardiac opacity. The patient was treated with supportive measures. Then the patient was admitted to a general hospital due to another attack of hemoptysis with syncope. Computed tomography scan of thorax with contrast revealed right lower lobe opacity with calcification (Figure 2). Due to the suspicious of malignancy, ultrasonographic-guided aspiration was done twice and the pathological examination revealed fibrous tissue with dilated vascular spaces. Finally, the patient was referred to our chest medicine department for further evaluation and bronchoscopy. After revision of his history and chest radiology, the pulmonary sequestration was suspected besides other benign lung lesions and CT angiography was ordered before any further interventions. It revealed evidence of right intralobular pulmonary sequestration. It has dual arterial supply from both right pulmonary artery and multiple branches from aorta arise, the venous drainage into right pulmonary vein (Figure 2 and Figure 3). Finally, the patient was transferred to the surgical department for surgical intervention, but the patient refused to perform any interventions.

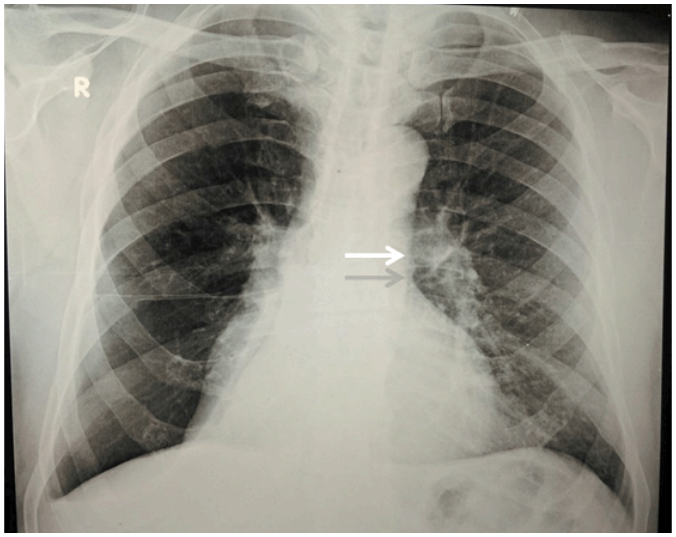


Figure 1: Chest X ray: Right paracardiac shadow.

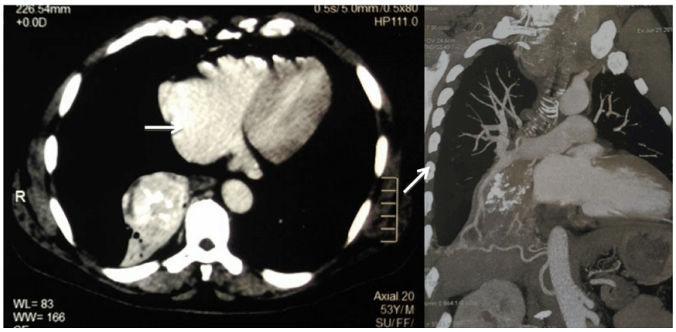


Figure 2: (A) Computed tomography scan of chest with contrast reveled right lower opacity, and (B) Chest angiography with appearance of the aberrant blood supply of pulmonary sequestration.

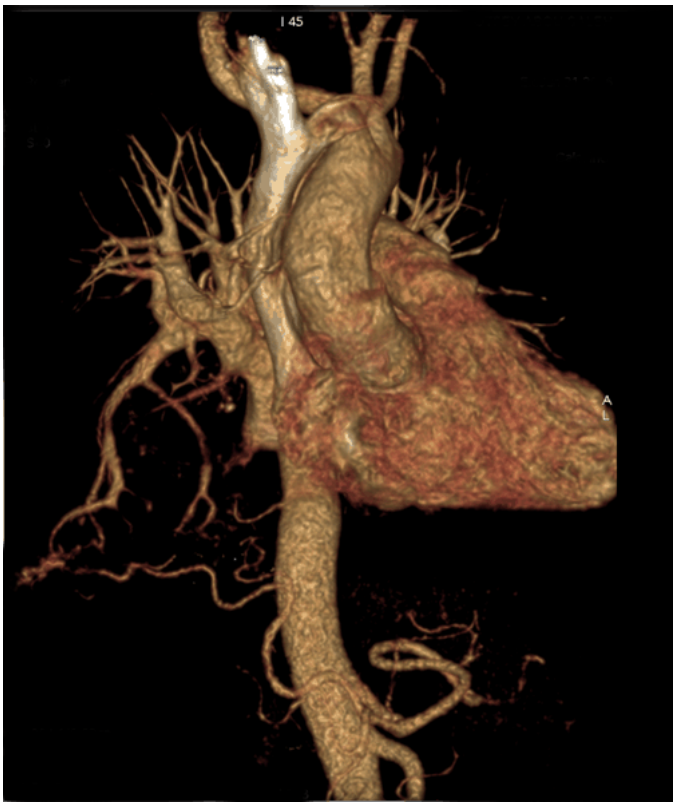


Figure 3: Computed tomography angiography (volume rendering) show the arterial blood supply of pulmonary sequestration from both right pulmonary artery and multiple branches from aorta.

DISCUSSION

Pulmonary sequestration was first described by Pryce in 1946. Its name was derived from the Latin verb 'sequestrate' which means to set apart. It can be defined as a developmental lung disease with non-functioning pulmonary tissue; which has no communication with the bronchial tree and receive a systemic blood flow [4].

Pulmonary sequestration is classified into three subtypes:

- Intralobar pulmonary sequestration (located within normal lung lobe and has its own visceral pleura),
- Extra lobar pulmonary (located outside the lung lobe and has its visceral pleura), and
- Bronchopulmonary- foregut malformation (BPFM) which is a rare variant of sequestration and is connected to the gastrointestinal tract [5].

The incidence of the pulmonary sequestration is rare and represented about 0.15–6.4% of all congenital pulmonary malformations [6]. Generally, ILS is the most common form of pulmonary sequestration. It represents about 75–90% with no sex differentiation [5].

The embryologic basis of the pulmonary sequestration remained unclear. Many possibilities had been suggested. The first referred to very early abnormality in the development during lung bud formation. Another

theory suggested mechanical separation of a portion of the developing lung due to compression or traction by aberrant vascular structure or inadequate pulmonary blood flow. However, the mechanical theory cannot fully explain all types of the pulmonary sequestration especially BPFM [7]. The third theory suggested that ILS may be an acquired rather than developmental lesion [8]. Recently, researchers found that the abnormal vascular development in the pulmonary arterial blood supply can lead to retention and proliferation of the nascent systemic capillary network [1].

Intralobar sequestration (ILS) usually presents in patients less than 20 years of age in 50% of the patients and rarely found in patients older than 50 years. Lower lobe above the diaphragm is typically the mostly affected area and left side is common in 55–60% of the patients [2, 4].

Herein, we describe presentation of ILS in a male patient 59-year-old which is not only atypical due to the old age but also due to the atypical site in the right lower lobe. The time of ILS presentation is variable. It usually present in late childhood or adolescence. It presented with recurrent lower respiratory tract infection. Hemoptysis and chest pain were also reported. It may be also asymptomatic and discovered in routine chest radiology in 15.5% of the patients with ILS [6]. In rare cases, heart failure occurs due to high flow through the anomalous artery [1].

The radiological assessment aimed not only to confirm the sequestration diagnosis, but also to evaluate its vascular supply for further management. Besides the chest radiography, ultrasonography, computed tomography scan and magnetic resonance imaging scan, the relatively recent introduced angiography represent the diagnostic tool of choice to define the vascular supply prior to any surgical intervention [3]. Single feeding blood supply was detected in 71% of the patients. It arises from the thoracic aorta in 36% [6].

The intralobar sequestration can be classified according to Pryce et al. into:

- type 1 presence of aberrant artery without sequestration,
- type 2 the aberrant artery supply the sequestration as well as the adjacent normal lung and type
- type 3 the aberrant supply only the sequestration [2].

The main management of the pulmonary sequestration in symptomatic patient is surgical resection for curative purposes. Proper identification and ligation of the feeding vessels is crucial. While in asymptomatic patients with ILS, the surgery is also recommended to prevent recurrent infections and the unfavorable cardiac influence caused by the existing aortopulmonary shunt. Recently, published data has introduced the emerging successful role of video-assisted thoracoscopic surgery (VATS) for pulmonary sequestration resection despite the difficulties in surgical dissection due to recurrent inflammation and fibrosis owing to the recurrent infections [9]. Arterial embolization of the feeding vessels has been also reported [10].

CONCLUSION

The interesting points in our case include the late presentation of the patient (59-year-old), unusual right-sided Intralobar sequestration (ILS) and the double blood supply from both pulmonary artery and abdominal aorta. Finally, we should emphasize the importance of proper interpretation of the chest radiology by the primary medical care providers as a standard tool of chest examination for early and adequate diagnosis of catastrophic hidden chest diseases.

****

Author Contributions

Ahmed Ehab – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published
Marwa Ghanem – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published
Ahmed Ibrahim Twafik – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published
Magda Abdel-salam – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published
Nasef Abdel-Salam A. Rezk – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published
Asem A. Hewidy – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

Copyright

© 2017 Ahmed Ehab et al. This article is distributed under the terms of Creative Commons Attribution License which permits unrestricted use, distribution and reproduction in any medium provided the original author(s) and original publisher are properly credited. Please see the copyright policy on the journal website for more information.

REFERENCES

1. Hertzenberg C, Daon E, Kramer J. Intralobar pulmonary sequestration in adults: Three case reports. J Thorac Dis 2012 Oct;4(5):516–9.
2. Hirai S, Hamanaka Y, Mitsui N, Uegami S, Matsuura Y. Surgical treatment of infected intralobar pulmonary sequestration: A collective review of patients older than 50 years reported in the literature. Ann Thorac Cardiovasc Surg 2007 Oct;13(5):331–4.
3. Kang M, Khandelwal N, Ojili V, Rao KL, Rana SS. Multidetector CT angiography in pulmonary sequestration. J Comput Assist Tomogr 2006 Nov-Dec;30(6):926–32.
4. Alizadeh E, Suliman H. Intralobular pulmonary sequestration. JBR-BTR 2013 Jul-Aug;96(4):208–9.
5. Stocker JT, Drake RM, Madewell JE. Cystic and congenital lung disease in the newborn. Perspect Pediatr Pathol 1978;4:93–154.
6. Van Raemdonck D, De Boeck K, Devlieger H, et al. Pulmonary sequestration: A comparison between pediatric and adult patients. Eur J Cardiothorac Surg 2001 Apr;19(4):388–95.
7. Kravitz RM. Congenital malformations of the lung. Pediatr Clin North Am 1994 Jun;41(3):453–72.
8. Holder PD, Langston C. Intralobar pulmonary sequestration (a nonentity?). Pediatr Pulmonol 1986 May-Jun;2(3):147–53.
9. Kestenholz PB, Schneiter D, Hillinger S, Lardinois D, Weder W. Thoracoscopic treatment of pulmonary sequestration. Eur J Cardiothorac Surg 2006 May;29(5):815–8.
10. Cho MJ, Kim DY, Kim SC, Kim KS, Kim EA, Lee BS. Embolization versus surgical resection of pulmonary sequestration: Clinical experiences with a thoracoscopic approach. J Pediatr Surg 2012 Dec;47(12):2228–33.

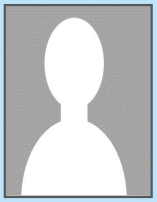
ABOUT THE AUTHORS

Article citation: Ehab A, Ghanem M, Twafik AI, Abdel-Salam M, Abdel-Salam N, A. Rezk NA, Hewidy AA. Intralobar pulmonary sequestration: A rare presentation in adults. Int J Case Rep Images 2017;8(2):133–137.

Ahmed Ehab is Assistant lecturer of chest medicine at chest medicine department, Faculty of medicine, Mansoura University, Mansoura, Egypt. He earned undergraduate degree (M.B.B.Ch) and postgraduate degree (MD in Chest Medicine) from from Mansoura University, Mansoura, Egypt. He has published two papers and one book. His research interests include bronchoscopic intervention, medical thoracoscopy, and respiratory critical care.
E-mail: dr.a.ehab@gmail.com

Marwa Ghanem is Chest medicine resident at Chest medicine department, Faculty of medicine, Mansoura University, Mansoura Egypt. She earned the undergraduate degree (M.B.B.Ch) from Mansoura University, Mansoura, Egypt). Her research interests include interventional bronchoscopy and respiratory critical care.
E-mail: marwasalah366@gmail.com

Ahmed Ibrahim Twafik is lecturer of diagnostic and interventional Radiology at Department of diagnostic and interventional radiology, faculty of medicine, Mansoura University, Mansoura, Egypt. He earned undergraduate degree (M.B.B.Ch) and postgraduate degree (MD in Radiology) from Mansoura University, Mansoura, Egypt. He has published one research paper and one book. His interest includes interventional radiology.
E-mail: ahmedtawfik82@yahoo.com



Magda Abdel-Salam is Chest medicine Assistant Professor at Chest medicine department, Faculty of medicine, Mansoura University, Mansoura, Egypt. She earned undergraduate degree (M.B.B.Ch) and postgraduate degree (MD in Chest Medicine) from Mansoura University, Mansoura, Egypt.
E-mail: drmagy2020@gmail.com

Nasef Abdel-Salam A. Rezk is Chest medicine Assistant Professor at Chest Medicine Department, Faculty of medicine, Mansoura University, Mansoura, Egypt. He earned undergraduate degree (M.B.B.Ch) and postgraduate degree (MD in Chest Medicine) from Department of chest medicine, faculty of medicine, Mansoura University, Mansoura, Egypt. He has published ten research papers in national and international academic journals.
E-mail: rawanmn@yahoo.com

Asem A. Hewidy is Chest medicine Assistant Professor at Chest Medicine Department, Faculty of Medicine, Mansoura University, Mansoura, Egypt. He earned undergraduate degree (M.B.B.Ch) and postgraduate degree (MD in Chest Medicine) from Chest Medicine Department, Faculty of Medicine, Mansoura University, Mansoura, Egypt. He has published eleven research papers in national and international academic journals. His research interests include interventional bronchoscopy, and thoracoscopy.
E-mail: ahewidy@yahoo.com

Access full text article on other devices

Access PDF of article on other devices

CASE REPORT PEER REVIEWED | OPEN ACCESS

Use of a fracture table for irreducible bipolar hemiarthroplasty dislocation: A case report

Chelsea S. Mathews, Robert L. Garrison, Regis L. Renard

ABSTRACT

Introduction: This article describes a case in which a fracture table was used to successfully reduce a hip dislocation which was seemingly irreducible using other common methods. Case Report: A 53-year-old female presented to the authors' facility with pain in the right hip and an infected posterolateral hip incision. Radiographs showed a posterior dislocation of the right hip status post bipolar hemiarthroplasty. The patient was taken to the operating room and successfully closed reduced using the Allis method. The infection was eradicated with IV antibiotics and I & D. After six weeks of post reduction, the patient presented to the emergency room with another right hip dislocation. A closed reduction was attempted using the same technique as before, but it was unsuccessful. Based on the patient's chronic diagnosis (stage IV lung cancer) we chose to treat this again in a closed manner. The patient was transferred to a fracture table, and using longitudinal traction, a successful reduction was achieved. At final follow-up the patient had maintained reduction without signs or symptoms of infection. Conclusion: Use of the fracture table for hip reduction may be an option in certain selected patients.

Keywords: Bipolar, Dislocation, Hemiarthroplasty, Irreducible

Chelsea S. Mathews[1], Robert L. Garrison[2], Regis L. Renard[3]

Affiliations: [1]MD, PGY-3 Resident, Department of Orthopedic Surgery, University of Arkansas for Medical Sciences, Little Rock, Arkansas, USA; [2]MD, Assistant Professor, Department of Orthopedic Surgery, University of Arkansas for Medical Sciences, Little Rock, Arkansas, USA; [3]MS, MD, Assistant Professor, Department of Orthopedic Surgery, University of Arkansas for Medical Sciences, Little Rock, Arkansas, USA.

Corresponding Author: Robert L. Garrison II, MD, UAMS Department of Orthopedic Surgery, 4301 West Markham Street, #531, Little Rock, AR, USA; E-mail: rlgarrison@uams.edu

Received: 19 August 2016
Accepted: 10 November 2016
Published: 01 February 2017

How to cite this article

Mathews CS, Garrison RL, Renard RL. Use of a fracture table for irreducible bipolar hemiarthroplasty dislocation: A case report. Int J Case Rep Images 2017;8(2):138–142.

Article ID: Z01201702CR10764CM

****

doi:10.5348/ijcri-201725-CR-10764

INTRODUCTION

Over a quarter-million femoral neck fractures occur annually and these numbers are expected to rise to 500,000 by 2040 [1]. The optimal treatment of displaced femoral neck fractures is still debated and includes unipolar and bipolar hemiarthroplasty, as well as total hip arthroplasty. Although there are reported advantages to each method, the general tendency amongst arthroplasty surgeons is to perform hemiarthroplasty. In a survey of AAHKS members, 85% reported this to be their primary method of treatment for patients age 65 or older with a displaced femoral neck fracture [1].

The incidence of dislocation after hip hemiarthroplasty after a femoral neck fracture can range from 1.5–3.8% [2]. Risk factors that appear to increase the risk of dislocation include a smaller center-edge angle and lack of tendon to bone reconstruction of the short external rotators [3]. Patients who are receiving hemiarthroplasty for hip fracture are also at a greater

risk for dislocation than from other diagnoses [2]. Dislocation after a hemiarthroplasty can be a devastating issue that is often difficult to treat. Closed reduction is the primary method of treatment after the first dislocation. Use of a fracture table in this instance can provide a gentle method of reduction with more control than with manual manipulation.

CASE REPORT

A 53-year-old female presented to the emergency room with pain in the right hip. She was unable to ambulate and had drainage from a right posterolateral hip incision. Ten days prior to presentation she underwent hemi-arthroplasty of the right hip by a physician at another hospital. She was unhappy with her postoperative care at the outside hospital and thus presented to our emergency department for evaluation and treatment. She was unable to recall any recent trauma or incident that caused her worsening pain. The patient was not a community ambulator prior to her surgery but would ambulate at home with assistive devices. She had a history of stage IV lung cancer, with chemotherapy and radiation treatment, as well as brain metastasis. She was receiving palliative care.

Upon examination, the patient was sitting up in bed and appeared to be comfortable. It was noted that the right leg was shortened and internally rotated. The patient was afebrile and vital signs were stable. Staples were in place over the posterolateral hip incision. The area was erythematous and had some mild drainage. There was no obvious wound dehiscence or purulent material. Her right leg was well perfused with a normal sensorimotor examination.

Laboratory examinations revealed a C-reactive protein 13.8, erythrocyte sedimentation rate (ESR) 15 and white blood cell count 8.09 cm^3. Imaging of the right hip showed a posterior dislocation of the right hip status post bipolar hemiarthroplasty (Figure 1).

After obtaining consent, under propofol sedation, the patient's hip was reduced in the emergency room using the Allis maneuver. Reduction was confirmed by radiograph (Figure 2). The patient was admitted with an abduction pillow and knee immobilizer. She was started on antibiotic therapy for the surgical site infection. She was placed on posterior hip precautions with touch-down weight-bearing status to the right lower extremity.

For several days, the wound was monitored and it was felt that the infection would resolve with IV antibiotic therapy. However, no improvement was noted. So on hospital day-5, the patient returned to the operating room for irrigation and debridement of the right hip. Cultures from the wound were positive for multiple organisms and the patient was started on a course of vancomycin and levaquin.

On postoperative day-1, hospital day-6, the patient's right lower extremity was noted to be shorter than the left. X-rays of the right hip revealed a dislocated right hemiarthroplasty (Figure 3).

The patient was taken back to the operating room that day for reduction. The hip was reduced using a sheet over the pelvis for counter traction. The right leg was gently adducted and flexed with longitudinal traction. Reduction was confirmed on two views and the patient was transferred back to the floor with an abduction pillow.

On hospital day-9, the patient was discharged to rehab with an abduction brace and was instructed on touch-down weight-bearing to the right leg.

Three weeks from discharge, the patient presented to clinic for follow-up. The hip remained reduced (Figure 4). Sutures were removed and there were no additional concerns.

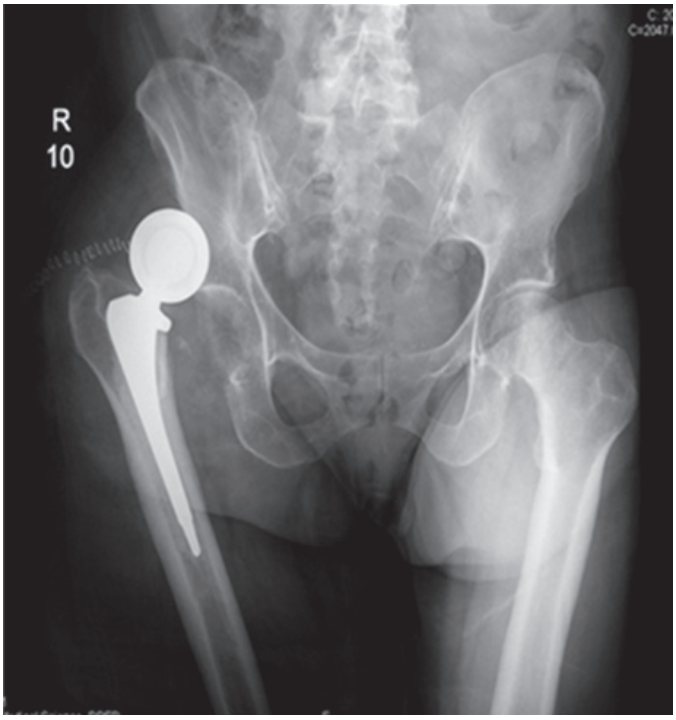


Figure 1: Radiograph of initial right prosthetic hip dislocation.

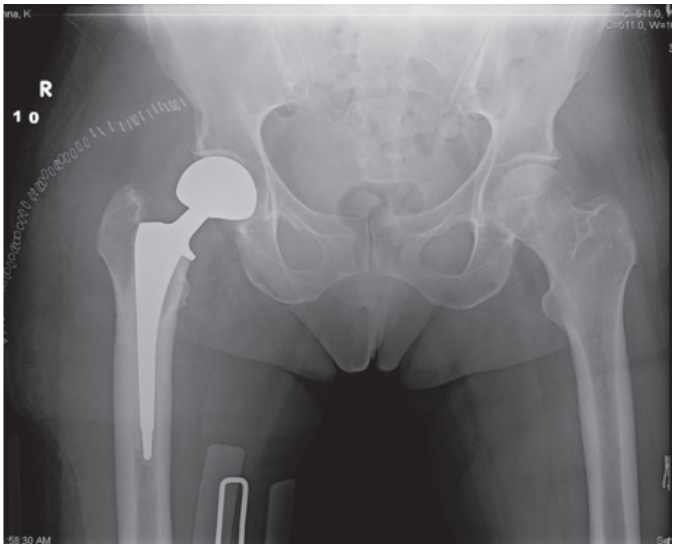


Figure 2: Radiograph of right hip post-reduction (Staples present from original outside surgeon's surgery).

Two weeks after the first clinic visit, the patient presented to the emergency room with another right hip dislocation (Figure 5). The surgical site remained clean with no signs or symptoms of infection. She, admittedly, had been non-compliant with her abduction brace. She was admitted overnight for reduction the next morning.

We discussed with the patient the possibility of a girdle stone procedure versus closed reduction based on her chronic diagnosis of stage IV lung cancer. Patient elected to proceed with a closed reduction with an understanding that compliance with bracing would be needed.

The next morning, the patient was brought to the operating room and placed on a radiolucent flat top table. A sheet was placed over the pelvis for countertraction and the same reduction maneuver was performed as before. After an unsuccessful attempt at reduction, the patient was transferred to a fracture table with a well-padded perineal post. Bilateral lower extremities were placed in longitudinal traction. With longitudinal traction, the right leg was externally rotated, then internally rotated and abducted. The hip was reduced and remained stable through range of motion (Figure 6). The patient was then transferred back to the floor with an abduction pillow.

The next day, the patient was discharged back to the rehab facility. She was instructed on touchdown weight-bearing to the right leg. She was strongly encouraged to be compliant with her abduction brace at all times out of bed and with an abduction pillow in bed.

At the time of this writing, the patient is three months status post right hip reduction on fracture table. She has returned to clinic on three occasions. She has remained compliant with the brace and pillow. Her hip remains reduced and her surgical site is well healed (Figure 7). She has graduated to full weight-bearing status.

DISCUSSION

Dislocation of bipolar hemiarthroplasty is not a common complication, but it is a problem that needs acute management and can be difficult to correct. Many hip hemiarthroplasties that dislocate require open reduction and revision. The reduction of a hip hemiarthroplasty is a tenuous process that requires gentle reduction and great care to protect both the implant and the native bone.

In a study performed by Sierra et al. [4], dislocations at 1, 5, 10, and 20 years were 1.1%, 1.5%, 2.1% and 5% respectively. More than half of the dislocations occurred in the first six months postoperatively. Only 30% of patients were treated successfully with closed reduction and no need for additional surgery.

Another study published by Salem et al. [5], shows their dislocation rate of hip hemiarthroplasties to be 0.76%. The majority of dislocations in their study occurred in the first six weeks after surgery and closed reduction was the definitive treatment in only 23% of the cases.

There are many techniques described for reducing a hip dislocation, but the literature has not thoroughly explored the use of a fracture table as a reduction technique. Commonly described reduction maneuvers include Allis', Bigelow's, Rochester and the Gravity method of Stimson. Position of the patient varies per maneuver, but the hip is generally gently flexed, internally rotated and then externally rotated into reduction. An article published by Flint et al. [6], describes a similar technique to the fracture table technique. They describe

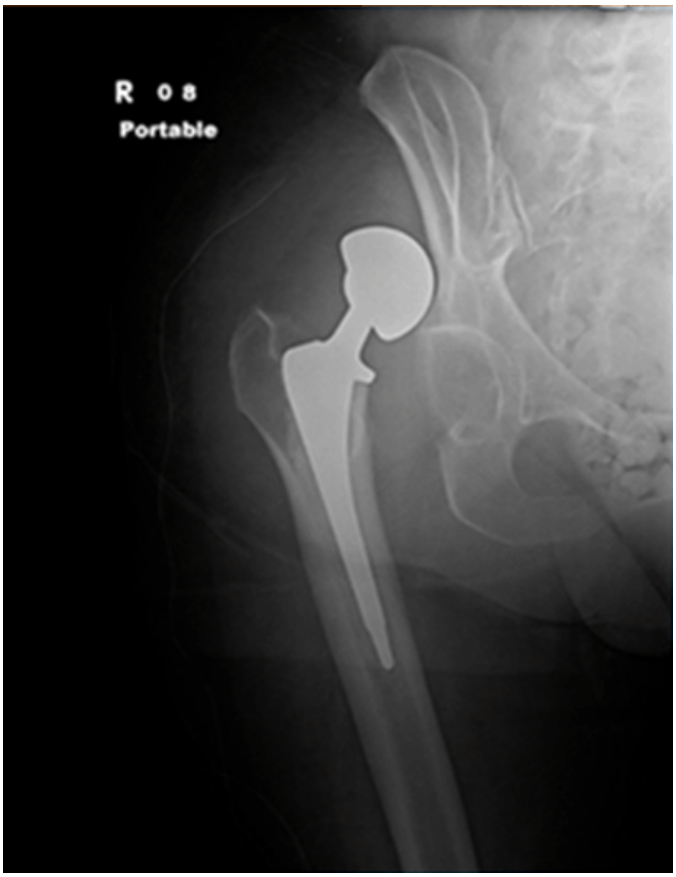


Figure 3: Radiograph of right hip dislocation on hospital day-6.

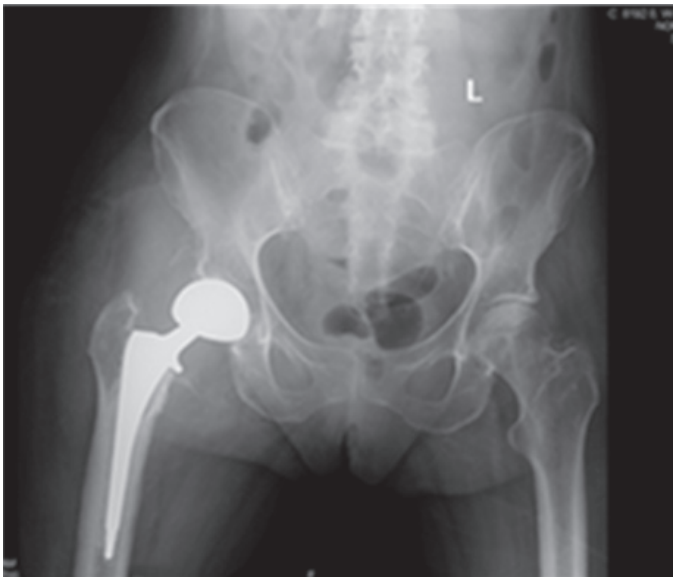


Figure 4: Radiograph of right hip three weeks post-discharge. hip remains reduced.

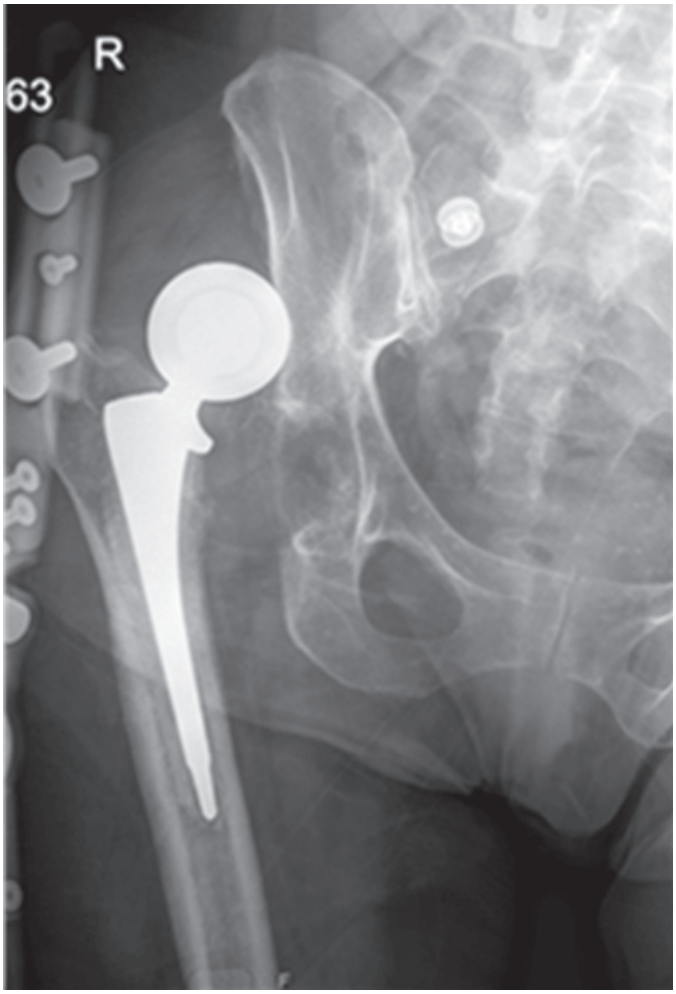


Figure 5: Radiograph of right dislocated hip two weeks after first clinic visit.

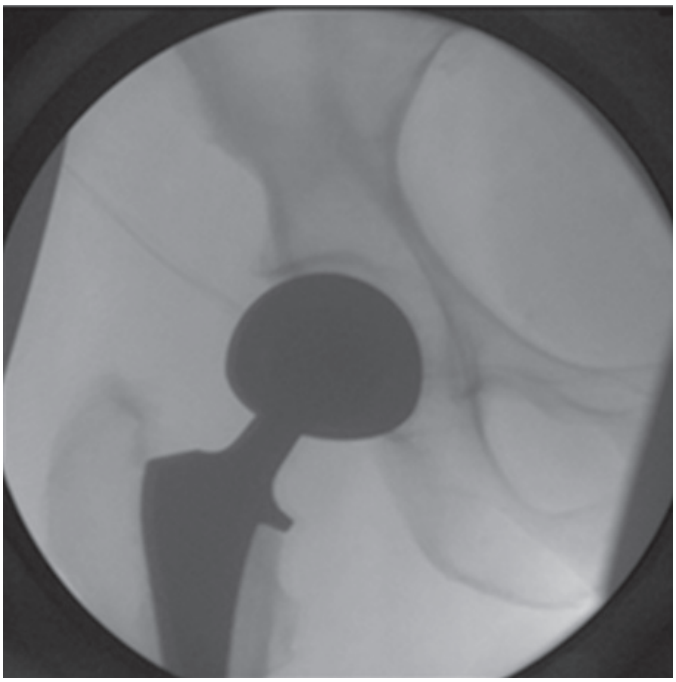


Figure 6: Radiograph of right hip post-reduction on fracture table. Hip is reduced.

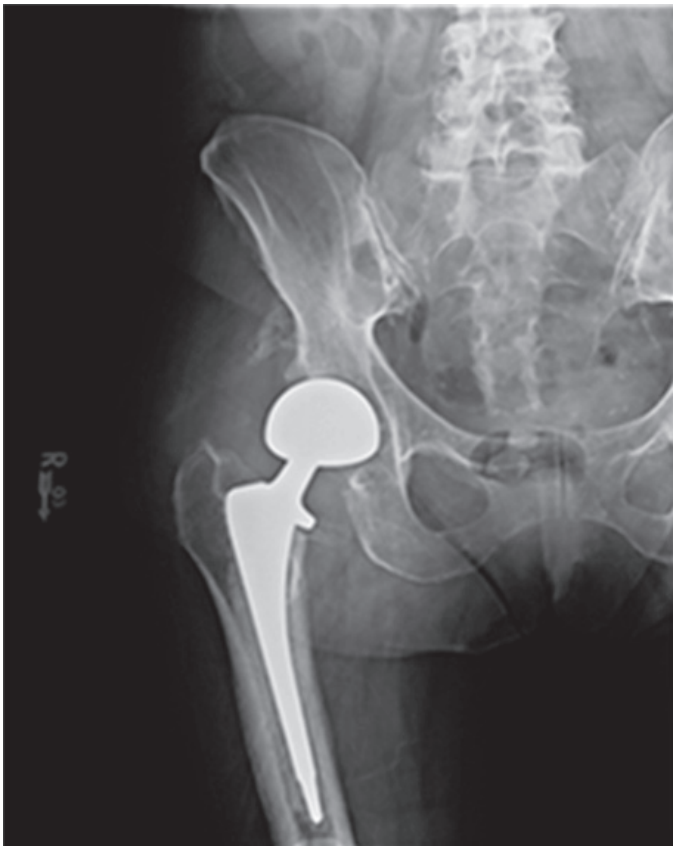


Figure 7: Radiograph of right hip, still reduced at third month post-fracture table reduction.

using an operating table with a peg board attached. They feel that this technique is more timely than placing the patient on a fracture table and may be of use in non-reducible hips requiring open reduction. The article describes a reduction technique combining longitudinal traction, internal rotation and pressure over the greater trochanter. While the peg board technique may allow for easier transition to open reduction, it does not offer the same gentle controlled traction that a fracture table provides.

CONCLUSION

In conclusion, an irreducible hemiarthroplasty dislocation may be treated with a fracture table using controlled traction. This technique may allow the hip to be closed reduced without the need for an open reduction.

****

Acknowledgements
We are thankful to Dana L. Garrison for her help and support in preparing this report.

Author Contributions

Chelsea S. Mathews – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Robert L. Garrison II – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Regis L. Renard – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

Copyright

© 2017 Chelsea S. Mathews et al. This article is distributed under the terms of Creative Commons Attribution License which permits unrestricted use, distribution and reproduction in any medium provided the original author(s) and original publisher are properly credited. Please see the copyright policy on the journal website for more information.

REFERENCES

1. Iorio R, Schwartz B, Macaulay W, Teeney SM, Healy WL, York S. Surgical treatment of displaced femoral neck fractures in the elderly: A survey of the American Association of Hip and Knee Surgeons. J Arthroplasty 2006 Dec;21(8):1124–33.
2. Barnes CL, Berry DJ, Sledge CB. Dislocation after bipolar hemiarthroplasty of the hip. J Arthroplasty 1995 Oct;10(5):667–9.
3. Kim Y, Kim JK, Joo IH, Hwang KT, Kim YH. Risk factors associated with dislocation after bipolar hemiarthroplasty in elderly patients with femoral neck fracture. Hip Pelvis 2016 Jun;28(2):104–11.
4. Sierra RJ, Schleck CD, Cabanela ME. Dislocation of bipolar hemiarthroplasty: Rate, contributing factors, and outcome. Clin Orthop Relat Res 2006 Jan;442:230–8.
5. Salem KM, Shannak OA, Scammell BE, Moran CG. Predictors and outcomes of treatment in hip hemiarthroplasty dislocation. Ann R Coll Surg Engl 2014 Sep;96(6):446–51.
6. Flint JH, Phisitkul P, Callaghan JJ. Closed reduction of a dislocated constrained total hip arthroplasty using a novel technique with a PEG board. Orthopedics 2010 Mar;33(3).

Access full text article on other devices

Access PDF of article on other devices

CASE REPORT PEER REVIEWED | OPEN ACCESS

A child with fulminant acute myocarditis rescued with extracorporeal membrane oxygenation

Colaco Sylvia M., Raghavan Subramanyam,
Anto Sahayaraj R., Cherian K.M.

ABSTRACT

Introduction: Fulminant acute myocarditis (FAM) occurs rapidly, causes pump failure or lethal arrhythmias, sometimes leading to death by cardiogenic shock. Case Report: We hereby present a three-year-old girl, previously asymptomatic, who developed rapid onset tachycardia, hypotension and cardiorespiratory arrest following an episode of respiratory tract infection. The patient was treated with anti-arrhythmic drugs, inotropes, and cardioversion after being diagnosed as atrial tachycardia, but the rhythm did not revert to sinus rhythm. Due to deteriorating condition patient was put on extra corporeal membrane oxygenation (ECMO) and supported for 131 hours along with supportive and IVIG treatment. After improvement in ejection fraction, patient was weaned off ECMO. The rhythm reverted to sinus after three days of admission and antiarrhythmics were gradually tapered. Patient was discharged on 17th day of admission in a stable condition with an ejction fraction of 58%. Conclusion: Timely extracorporeal membrane oxygenation (ECMO) support in fulminant acute myocarditis (FAM) with refractory atrial tachycardia and shock due to myocarditis, along with medical treatment could prevent lethal outcomes.

Keywords: Child, Extracorporeal membrane oxygenation (ECMO), Fulminant acute myocarditis (FAM)

Colaco Sylvia M.[1], Raghavan Subramanyam[2], Anto Sahayaraj R.[3], Cherian K.M.[4]

Affiliations: [1]Fellow in Pediatric Cardiology, Frontier lifeline hospital, Chennai; [2]Head of Department of Pediatric Cardiology, Frontier Lifeline Hospital, Chennai; [3]Consultant Surgeon, Frontier Lifeline Hospital, Chennai; [4]Director and Head Surgeon, Frontier Lifeline Hospital, Chennai.

Corresponding Author: Sylvia Michael Colaco, Philrose, Behind Manickpur Cricket Ground, Vasai road (west), Dist-Palghar, Maharashtra, India, 401202; Email: drsylviac@gmail.com

Received: 18 September 2016
Accepted: 21 October 2016
Published: 01 February 2017

How to cite this article

Colaco SM, Subramanyam R, Sahayaraj AR, Cherian KM. A child with fulminant acute myocarditis rescued with extracorporeal membrane oxygenation. Int J Case Rep Images 2017;8(2):143–146.

Article ID: Z01201702CR10765CM

****

doi:10.5348/ijcri-201726-CR-10765

INTRODUCTION

Myocarditis is an inflammatory process of the myocardium that can have multiple etiologies such as infection, systemic disease and/or exposure to medications and toxins [1]. Fulminant acute myocarditis (FAM) presents with rapid onset of cardiac manifestations, which typically manifest after nonspecific flu-like symptoms and rapidly progresses to severe hemodynamic deterioration and severe heart failure, cardiogenic shock and potentially fatal arrhythmias [2]. In pediatric patients, FAM accounts for 30–40% of cases of myocarditis and has a mortality rate of up to 48% [3]. Cases have been reported in literature of severe myocarditis being treated successfully with extra corporeal membrane oxygenation (ECMO) both in adults and children, but none in India. We present a case of FAM with heart failure and refractory atrial tachycardia in a child where life support with ECMO was effective.

CASE REPORT

A three-year-old girl, previously asymptomatic, with completed vaccination and no history of receiving any viral vaccines developed cold and cough without any recorded fever. Due to worsening symptoms, the child was taken to a pediatric hospital and diagnosed to have supraventricular tachycardia. Two doses of injection adenosine failed to control the tachycardia. The child was then referred to our centre. On examination the child had cold peripheries, feeble pulses, and a heart rate of 235/minute. The child was started on inotropic supports and again an injection adenosine (5 mg) was given in vain. Injection amiodarone (5 mg/kg) was infused rapidly and continued at 15 mics/kg/min. Two shocks of cardioversion with energy of 0.5 J/Kg and 2 J/kg were tried but were unsuccessful in reverting the rhythm to sinus rhythm (Figure 1). On the same day the child had an episode of cardiorespiratory arrest but was successfully resuscitated. Bedsides echocardiogram showed an ejection fraction of 15% with generalized left ventricular hypokinesia. The basic investigations are summarized in Table 1. In view of the refractory arrhythmia, falling pressures and ventricular dysfunction a decision was made to support the child on ECMO. Extracorporeal membrane oxygenation insertion was taken through the left iliac arterial and venous cannulations followed by distal femoral cannulation to perfuse the left lower limb. The ECMO circuit used was an indigenously prepared circuit (Figure 2). All the tubings were heparin coated, and activated clotting time (ACT), which is a test to monitor heparin therapy in clinical situations where intensive anticoagulation needed was maintained between 180–200 s. Extracorporeal membrane oxygenation was started on high flow at 2 L/min and a total of 131 hours ECMO support was provided. Patient was also treated with intravenous immunoglobulin, (2 g/kg over 48 hours), pulse therapy of methylprednisolone, anti-failure medications (frusemide, spironolactone and digoxin) and IV antibiotics. The tachycardia was identified as ectopic atrial tachycardia. Since the tachycardia did not respond to higher doses of amiodarone, flecainide was started with mild reduction in the rate. Carvedilol was introduced once the blood pressure was stable. The rhythm was reverted to sinus rhythm on the third day of admission following which patient was gradually weaned off ECMO and then ventilator. After 48 hour observation in the PICU, the patient remained hemodynamically and neurologically stable, was shifted to the ward on the eighth day. The child had an unremarkable course in the ward and was discharged on day-17.

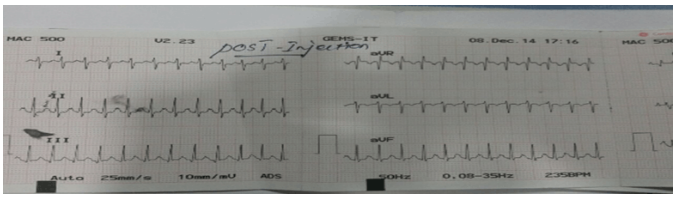


Figure 1: Electrocardiogram on admission which did not revert on injection adenosine.

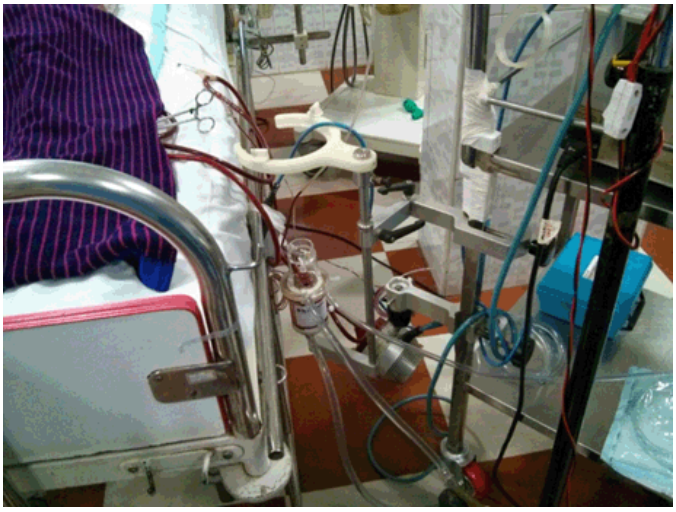


Figure 2: Extracorporeal membrane oxygenation (ECMO) circuit.

Table 1: Hemodynamics

Parameters	DAY 1	DAY 2	DAY 4	DAY 17
Hemoglobin	11.7	13.8	11.8	10.6
TLC	19500	26100	12100	9400
Platelets	1.35	86000	1L	1.20
Urea	78		129	60
Creatinine	1.2		1.3	0.7
CPKMB	158		560	
TROP	0.14		0.09	
BNP	>25,000			
CRP	2.1			
D-dimer	positive			

DISCUSSION

Fulminant acute myocarditis (FAM) carries a high mortality, ranging from 50–75%, without immediate mechanical circulatory support [4]. Prompt diagnosis, as well as proper mechanical circulatory support, improves survival [5]. Our patient presented in an emergency situation with a past history of flu-like illness for seven days and no improvement with antibiotics and sudden onset refractory arrhythmia. This was followed by rapid development of severe heart failure, cardiogenic shock, refractory atrial tachycardia, and cardiac arrest. The diagnosis of FAM was initially made on clinical grounds supported by echocardiographic evidence of severe left ventricular impairment. This was later corroborated by elevated cardiac enzymes and BNP levels.

Echocardiography is an important tool in the diagnosis of myocardial dysfunction and is able to exclude other anatomical causes of heart failure and helps to identify the

fulminant course of the disease. Classical findings include global hypokinesia, with or without pericardial effusion, variable degree of myocardial dilation, and atrioventricular regurgitation [6]. Our patient had severe left ventricular systolic dysfunction, an ejection fraction of 15% and severe mitral regurgitation. Ejection fraction of 15%, severely raised BNP levels (> 25,000), cardiogenic shock and refractory arrhythmias indicated severe and rapidly worsening condition; hence the decision of mechanical circulatory support was taken. There are many case reports and some studies which have show the usefulness of ECMO in FAM in children [2, 5, 7, 8]. These treatment methods have been applied to patients under conditions as refractory cardiac arrest or lethal arrhythmias, and circulatory failure by low output syndrome.

The assisted circulation used a 10 F tube for blood inflow and a 12-F tube for blood outflow which were sufficient for securing the blood volume and proper placement without causing any damage to the vessels. A problem with arterial cannulation was the inadequate perfusion of the lower limb below the site of cannulation. For this a 4-F catheter was introduced into the iliac vessel distally. The left lower limb pulses, calf girth and color of the toes and limbs were monitored during the entire ECMO duration. No vascular complications were observed in our patient. The mean duration for ECMO support varies from 20–126 hours, [8] our patient was weaned off support after 131 hours. Invasive mechanical ventilation is often required in cases of fulminant myocarditis with cardiogenic shock. Mild to moderate hyperventilation may help to correct acidemia in initial phases [9].

Since myocarditis is an immune-mediated inflammatory myocardial damage, targeting that process may improve outcome. Immune modulation with intravenous immunoglobulin (IVIG) in high dose (2 g/kg) has been reported to be beneficial in children with improved survival [10]. It has multiple effects including neutralization of pathogens, reduction in inflammatory cytokines and antiviral action [11]. However, a major randomized control studies have been conducted which concluded that these therapies may alter the course of the disease but the results are statistically not significant [12]. The Cochrane database review concluded that IVIG therapy did not conclusively improve the outcome.

Immunesuppression with methylprednisolone has been used with a view to suppress cytokine production inflammation and myocardial damage especially in severely ill patients [13]. However, RCT have not proved a definite benefit with steroid therapy and may potentially cause side-effects [8]. Both immune modulation and immunesuppression are presently considered adjuvants to hemodynamic support.

Extracorporeal membrane oxygenation is a labor-intensive and expensive modality with an estimated total hospital cost of $20,000 to $90,000 per patient in developed countries [14]. Similarly, in a developing country like ours where the hospital cost burden is borne mainly by the patient as compared to insurance coverage in the west, ECMO support is a financial exhausting treatment. With our indigenous circuit, where the basic set up cost of the machine and tubings was around 1,00,000 Indian rupees ($2000) and the ECMO ongoing cost was 35,000 Indian rupees/day (700 $). Early recognition of the clinical picture and prompt ECMO support along with immunomodulation treatment may provide better chances of recovery for patients with FAM but the major problem in our country is the cost associated with ECMO.

CONCLUSION

Timely extracorporeal membrane oxygenation (ECMO) support in fulminant acute myocarditis (FAM) with refractory atrial tachycardia and shock due to myocarditis, along with medical treatment could prevent lethal outcomes.

****

Author Contributions

Colaco Sylvia M. – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published
Raghavan Subramanyam – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published
Anto Sahayaraj R. – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published
Cherian K.M. – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

Copyright

© 2017 Colaco Sylvia M. et al. This article is distributed under the terms of Creative Commons Attribution License which permits unrestricted use, distribution and reproduction in any medium provided the original author(s) and original publisher are properly credited. Please see the copyright policy on the journal website for more information.

REFERENCES

1. Bowles NE, Vallejo J. Viral causes of cardiac inflammation. Curr Opin Cardiol 2003 May;18(3):182–8.

2. Lobo ML, Taguchi Â, Gaspar HA, Ferranti JF, de Carvalho WB, Delgado AF. Fulminant myocarditis associated with the H1N1 influenza virus: Case report and literature review. [Article in English, Portuguese]. Rev Bras Ter Intensiva 2014 Jul-Sep;26(3):321–6.
3. Saji T, Matsuura H, Hasegawa K, et al. Comparison of the clinical presentation, treatment, and outcome of fulminant and acute myocarditis in children. Circ J 2012;76(5):1222–8.
4. Fenoglio JJ Jr, Ursell PC, Kellogg CF, Drusin RE, Weiss MB. Diagnosis and classification of myocarditis by endomyocardial biopsy. N Engl J Med 1983 Jan 6;308(1):12–8.
5. Wu ET, Huang SC, Chen YS, Wang JK, Wu MH, Ko WJ. Children with fulminant myocarditis rescued with extracorporeal membrane oxygenation. Heart 2006 Sep;92(9):1325–6.
6. Felker GM, Boehmer JP, Hruban RH, et al. Echocardiographic findings in fulminant and acute myocarditis. J Am Coll Cardiol 2000 Jul;36(1):227–32.
7. Nahum E, Dagan O, Lev A, et al. Favorable outcome of pediatric fulminant myocarditis supported by extracorporeal membranous oxygenation. Pediatr Cardiol 2010 Oct;31(7):1059–63.
8. Taoka M, Shiono M, Hata M, et al. Child with fulminant myocarditis survived by ECMO support: Report of a child case. Ann Thorac Cardiovasc Surg 2007 Feb;13(1):60–4.
9. Cooper HA, Panza JA. Cardiogenic shock. Cardiol Clin 2013 Nov;31(4):567–80, viii.
10. Robinson J, Hartling L, Vandermeer B, Crumley E, Klassen TP. Intravenous immunoglobulin for presumed viral myocarditis in children and adults. Cochrane Database Syst Rev 2005 Jan 25;(1):CD004370.
11. Levine MC, Klugman D, Teach SJ. Update on myocarditis in children. Curr Opin Pediatr 2010 Jun;22(3):278–83.
12. Hia CP, Yip WC, Tai BC, Quek SC. Immunosuppressive therapy in acute myocarditis: An 18 year systematic review. Arch Dis Child 2004 Jun;89(6):580–4.
13. Feltes TF, Adatia I. Immunotherapies for acute viral myocarditis in the pediatric patient. Pediatr Crit Care Med 2006;7:S17–S20.
14. Ayad O, Dietrich A, Mihalov L. Extracorporeal membrane oxygenation. Emerg Med Clin North Am 2008 Nov;26(4):953–9, ix.

Access full text article on other devices

Access PDF of article on other devices

CASE REPORT PEER REVIEWED | OPEN ACCESS

Very delayed coronary stent fracture presenting as unstable angina: A case report

Saurabh Mehrotra, Praful Sharma P., Yashpaul Sharma Y.P.

ABSTRACT

Introduction: Coronary stent fracture represents an under diagnosed clinical event of drug-eluting stents which is often associated with adverse clinical outcomes of in-stent restenosis. Numerous risk factors are associated with stent fracture that include stent overexpansion, creation of hinge points due to stent overlapping, use of longer stents for complex lesions as well as mechanical fatigue causing stent distortion in the right coronary artery and vein grafts. Case Report: A 64-year old male, a cigarette smoker, presented with rest angina. Coronary angiogram showed discrete 99% stenosis in proximal left anterior descending artery and a mid-eccentric 90% lesion in the right coronary artery (RCA). The patient was taken up for angioplasty of both the vessels. A type V fracture was detected after four years of zotarolimus-eluting stent placement in the right coronary artery. Conclusion: Despite the recent advances in drug-eluting stents design, there remains a potential of stent fracture especially when a long drug-eluting stents is implanted in a tortuous vessel and is exposed to torsion forces at the hinge points.

Keywords: Drug-eluting stent, Stent fracture, Zotarolimus-eluting stent

How to cite this article

Mehrotra S, Sharma PP, Sharma YYP. Very delayed coronary stent fracture presenting as unstable angina: A case report. Int J Case Rep Images 2017;8(2):147–150.

Article ID: Z01201702CR10766SM

****

doi:10.5348/ijcri-201727-CR-10766

Saurabh Mehrotra[1], Praful Sharma P.[2], Yashpaul Sharma Y.P.[3]

Affiliations: [1]MD, DM, Assistant Professor, Department of Cardiology, Post Graduate Institute of Medical Education and Research (PGIMER), Chandigarh, Punjab, India; [2]MD, DM, Senior Resident, Department of Cardiology, Post Graduate Institute of Medical Education and Research (PGIMER), Chandigarh, Punjab, India; [3]MD, DM, Head, Cardiology, Department of Cardiology, Post Graduate Institute of Medical Education and Research (PGIMER), Chandigarh, Punjab, India.

Corresponding Author: S. Mehrotra, Assistant Professor, Cardiology, Post Graduate Institute of Medical Education and Research (PGIMER), Sector-12, Chandigarh- 160012, Punjab, India; Email: smehrotrapgi@gmail.com

Received: 24 July 2016
Accepted: 21 November 2016
Published: 01 February 2017

INTRODUCTION

The introduction of drug-eluting stents has marked a new era in the field of interventional cardiology with significant reduction in the incidence of restenosis as well as repeat revascularization [1, 2]. Although drug-eluting stents has become the standard of care for percutaneous coronary intervention, the occurrence of late stent thrombosis has raised concern over their long-term safety. Stent fracture is being increasingly recognized as a potential cause of in-stent restenosis and stent thrombosis with the clinical manifestation of recurrent angina, myocardial infarction and even sudden death [3–6]. We report a rare case of delayed stent fracture after percutaneous coronary intervention with zotarolimus-eluting stent (ZES).

CASE REPORT

A 64-year old male, a cigarette smoker, presented to us with rest angina. The patient was not on any medication at the time of presentation. Clinical evaluation revealed ST segment elevation in anterior precordial leads on electrocardiogram along with raised troponins (29.834 ng/ml, normal level <1.5 ng/ml). His white blood cell count was 11.26×10^9/l (normal range $4–10\times10^9$/l), granulocyte proportion was 79.7% (normal range 46–75%), red blood cell count was 4.07×10^{12}/l (normal range $4.0–5.5\times10^{12}$/l), blood platelet count was 167×10^9/l (normal range $100–300\times10^9$/l), potassium was 3.54 mmol/l (normal range 3.5–5.3 mmol/l), sodium was 137 mmol/l (normal range 135–145 mmol/l) and urea was 6.6 mmol/l (normal range 2.9–8.6 mmol/l. The patient was thrombolysed with streptokinase (1.5 MU over 45 min). Coronary angiogram showed discrete 99% stenosis in proximal left anterior descending artery along with diffuse disease distally. The right coronary artery showed a mid-eccentric 90% lesion (Figure 1).

The patient was taken up for angioplasty of both the vessels. Endeavour Resolute stent 3.0×38 mm (Medtronic Inc. Santa Rosa, CA, USA) was deployed in mid to distal left anterior descending, followed by another overlapping Endeavour Re solute stent 3.5×38 mm in mid to proximal left anterior descending. Both the stents were post-dilated sequentially with 3.0×10 mm and 4.0×9 mm Dura Star noncompliant balloons (Cordis Corp, Johnson & Johnson, Miami Lakes, FL 33014) respectively. A third Endeavour Resolute stent 3.0×38 mm was deployed in the right coronary artery at 10 atmospheric pressure then post dilated with 4.0×12 mm noncompliant balloon at 10, 12 and 14 atm distal to proximally, with good angiographic results (Figure 2A–B).

The patient was subsequently discharged in stable condition and followed-up as outpatient uneventfully. His medications included ecosprin 150 mg daily, metoprolol 50 mg daily, clopidogrel 75 mg daily, and atorvastatin calcium 40 mg daily. After four years, patient once again presented with unstable angina. Laboratory tests revealed an HbA1c of 6.1%, lipid profile within goal range with total cholesterol 3.4 mmol/l, triglyceride 1.10 mmol/l, HDL 1.64 mmol/l and LDL 1.29 mmol/l. Serum creatinine was elevated at 120 mmol/l (normal range 58–110 mmol/l) and 24-h urinary protein 1.13 g. Further investigations revealed a normal troponin I of 0.3 ng/ml on the day of admission. Repeat coronary angiogram showed stents in left anterior descending. The right coronary artery stent showed a Type V fracture (multiple strut fractures with acquired transaction with gap in the stent body) in the middle with a clear gap between the two fractured segments (Figure 3A–C). The patient was advised bypass graft to right coronary artery and is currently asymptomatic at 6 month follow-up.

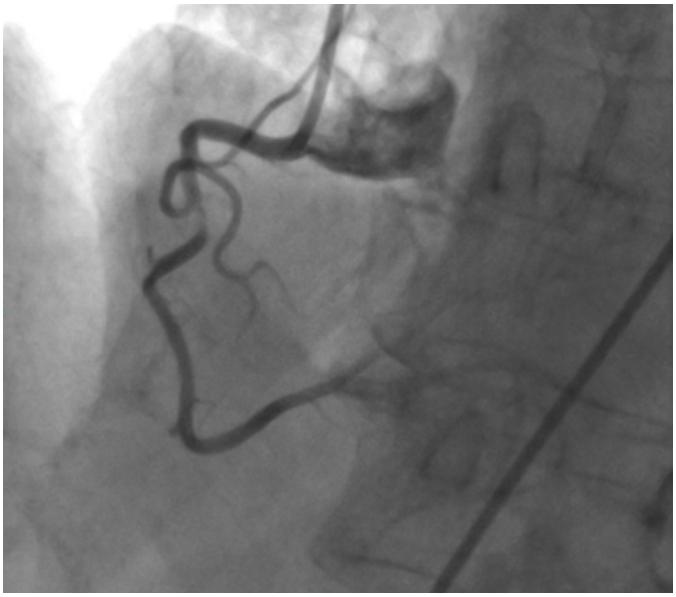


Figure 1: Right coronary artery mid discrete stenosis.

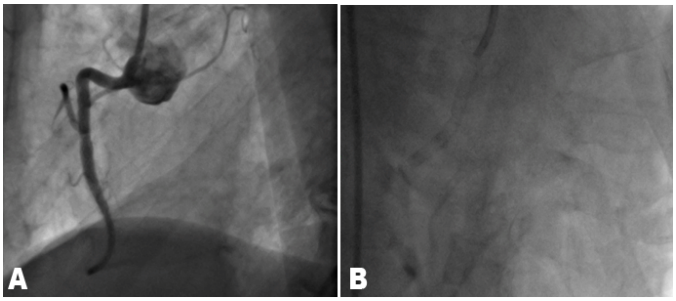


Figure 2: (A) Right coronary artery post stenting-with contrast, (B) Right coronary artery post stenting-without contrast.

DISCUSSION

Stent fracture is an important yet underestimated clinical entity associated with adverse clinical sequelae. Various clinical studies have reported the incidence of stent fracture in the range of 0.84–7.7% [7] whereas an autopsy study by Nakazawa et al. reported a 29% incidence of stent fracture in drug-eluting stents at autopsy [8]. Stent fracture is classified as isolated strut fractures (type 1, single-strut fracture; type 2, incomplete trans-verse fracture) and complete fracture (type 3, complete transverse fracture without displacement; type 4, transverse fracture with displacement).

Risk factors associated with stent fracture that has been reported in clinical studies include longer stent length, extremely angular and calcified lesion, post-dilatation with high pressure, right coronary artery or saphenous vein graft lesion location, lesion with high motion, overlapping stent and use of sirolimus eluting stent [9]. The rigid, and closed cell design of the sirolimus eluting stent results in greater straightening of the vessel thereby subjecting the stent to greater forces during the cardiac cycle [1, 10].

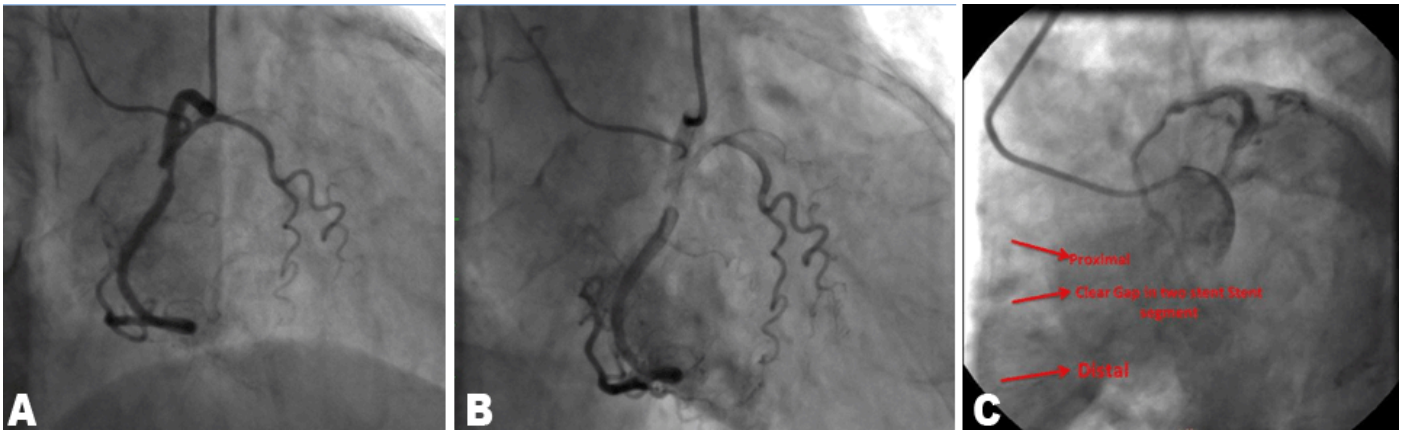


Figure 3: (A) Type V stent fracture in right coronary artery with contrast, (B) Type V stent fracture in right coronary artery without contrast, and (C) Proximal and distal portions of right coronary artery stent following the Type V stent fracture with the gap in between.

Zotarolimus-eluting stent with its open cell flexible design is less affected by torsion forces and there are only isolated case reports of stent fracture with ZES. In our case, a single ZES was implanted in non-calcified right coronary artery and the post dilatation pressure was well within the prescribed burst pressure. Symptomatic stent fracture four years after implantation in our case is possibly due to the chronic stretch at the bend point in the tortuous vessel resulting from the forceful exaggerated motion of the RCA. Stent fracture can present as recurrent angina, myocardial in farction or even sudden death with the adversity of clinical presentation having a direct relation to the severity of stent fracture [8]. Our case of Type V stent fracture has presented as unstable angina while the more significant clinical presentation of stent thrombosis was probably prevented due to the extended dual antiplatelet therapy of the patient.

As on date there is no clear cut consensus statement for the treatment of stent fracture and the decision is generally guided by the type of stent fracture , presence of ischemia as well as likelihood of recurrence. Since the patient in our case presented with rest angina along with a Type V stent fracture despite being on dual antiplatelet therapy, revascularization was indicated. Considering the high likelihood of recurrence in our case, the decision to graft the right coronary artery was taken.

CONCLUSION

This case highlights the importance of considering stent fracture, a not so rare entity as previously believed, as an etiological factor for causing late drug-eluting stents related complications with adverse clinical sequelae. Despite the recent advances in drug-eluting stents design, there remains a potential of this dreaded complication more so when a long drug-eluting stents is implanted in a tortuous vessel (right coronary artery in our case) and is exposed to torsion forces at the hinge points.

****

Author Contributions

Saurabh Mehrotra – Substantial contribution to Conception and design, Analysis and interpretation of data, Drafting the article, Final approval of the version to be published

Praful Sharma P. – Substantial contribution to Conception and design, Analysis and interpretation of data, Final approval of the version to be published

Yashpaul Sharma Y.P. – Substantial contribution to Conception and design, Analysis and interpretation of data, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

Copyright

© 2017 Saurabh Mehrotra et al. This article is distributed under the terms of Creative Commons Attribution License which permits unrestricted use, distribution and reproduction in any medium provided the original author(s) and original publisher are properly credited. Please see the copyright policy on the journal website for more information.

REFERENCES

1. Shaikh F, Maddikunta R, Djelmami-Hani M, Solis J, Allaqaband S, Bajwa T. Stent fracture, an incidental finding or a significant marker of clinical in-stent restenosis? Catheter Cardiovasc Interv 2008 Apr 1;71(5):614–8.
2. Betriu A, Masotti M, Serra A, Randomized comparison of coronary stent implantation and balloon angioplasty in the treatment of de novo coronary artery lesions

(START): A four-year follow-up. J Am Coll Cardiol 1999 Nov 1;34(5):1498–506.
3. Aoki J, Nakazawa G, Tanabe K, Incidence and clinical impact of coronary stent fracture after sirolimus-eluting stent implantation. Catheter Cardiovasc Interv 2007 Feb 15;69(3):380–6.
4. Lee MS, Jurewitz D, Aragon J, Forrester J, Makkar RR, Kar S. Stent fracture associated with drug-eluting stents: Clinical characteristics and implications. Catheter Cardiovasc Interv 2007 Feb 15;69(3):387–94.
5. Park JS, Shin DG, Kim YJ. Fractured DES with a patent coronary artery: Clinical implications. J Invasive Cardiol 2007 Feb;19(2):E43–5.
6. Shite J, Matsumoto D, Yokoyama M. Sirolimus-eluting stent fracture with thrombus, visualization by optical coherence tomography. Eur Heart J 2006 Jun;27(12):1389.
7. Alexopoulos D, Xanthopoulou I. Coronary stent fracture: How frequent it is? Does it matter? Hellenic J Cardiol 2011 Jan-Feb;52(1):1–5.
8. Nakazawa G, Finn AV, Vorpahl M, et al. Incidence and predictors of drug-eluting stent fracture in human coronary artery a pathologic analysis. J Am Coll Cardiol 2009 Nov 17;54(21):1924–31.
9. Kim HS, Kim YH, Lee SW, et al. Incidence and predictors of drug-eluting stent fractures in long coronary disease. Int J Cardiol 2009 Apr 17;133(3):354–8.
10. Canan T, Lee MS. Drug-eluting stent fracture: Incidence, contributing factors, and clinical implications. Catheter Cardiovasc Interv. 2010 Feb 1;75(2):237–45.

Access full text article on other devices

Access PDF of article on other devices

CLINICAL IMAGES PEER REVIEWED | OPEN ACCESS

Traumatic asphyxia: A rare syndrome in trauma children

Mohamed Adnane Berdai, Smael Labib, Mustapha Harandou

CASE REPORT

A 12-year-old boy, weight 31 kg, with no medical history, had falling from a horse-drawn carriage and was crashed by its wheels at the thorax and upper limbs for approximately 30 seconds. He was admitted to emergency department 45 minutes later. On arrival, he was lethargic, with a Glasgow Coma Score of 12 (E3V4M5); both pupils were equal and reactive to light. His blood pressure was 110/60 mmHg and his heart rate was 110/min. He had tachypnea with respiratory rate of 41/min, the pulsed oxygen saturation in ambient air was 94%.

The patient had facial purple congestion, diffuse head and neck edema and petechiae in the entire face, neck and upper chest (Figure 1). Ophthalmologic examination revealed the presence of sub-conjunctival hemorrhages without impact on visual acuity with a normal fundus (Figure 2). Abdominal examination showed epigastric abrasion without tenderness. Examination revealed a deformation of the right arm and ecchymotic bruises and abrasions on the right hip. The rest of physical examination was unremarkable.

Thoracic computed tomography (CT) scan showed bilateral pulmonary contusions, low abundance bilateral pneumothorax, and fractures of the 3rd and 4th left ribs. Cerebral CT scan was normal. A shoulder X-ray revealed

Mohamed Adnane Berdai[1], Smael Labib[2], Mustapha Harandou[2]

Affiliations: [1]MD, Assistant Professor of anesthesiology and intensive care / Child and mother intensive care unit, University hospital Hassan II, Fes, Morocco; [2]MD, Professor of anesthesiology and intensive care / Child and mother intensive care unit, University hospital Hassan II, Fes, Morocco.

Corresponding Author: Mohamed Adnane Berdai, Child and mother intensive care unit, University hospital Hassan II, Sidi Hrazem Avenue, 30000, Fes, Morocco; E-mail: adnane.berdai@yahoo.fr

Received: 24 September 2016
Accepted: 04 November 2016
Published: 01 February 2017

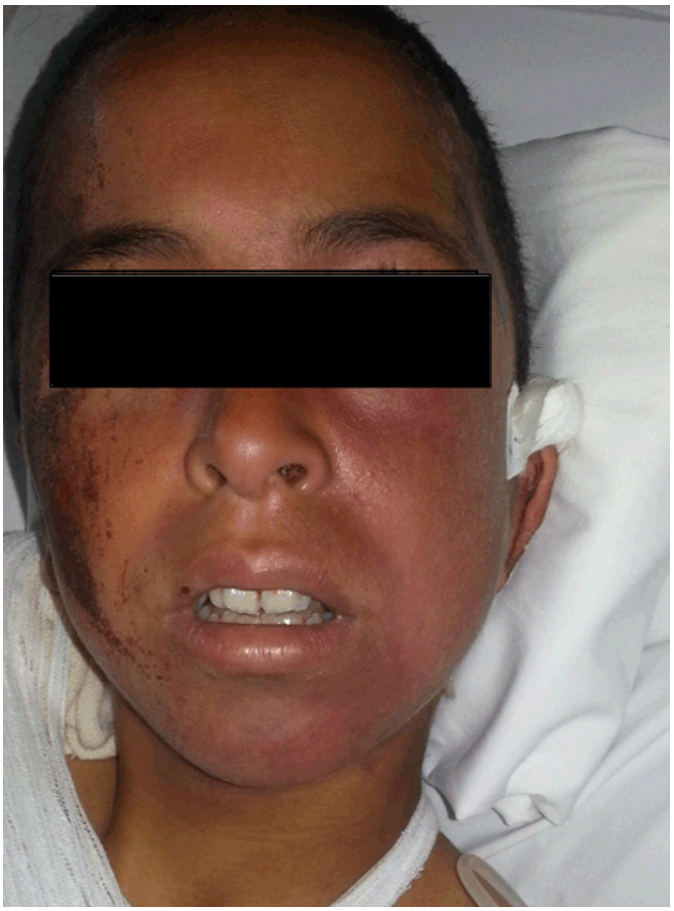


Figure 1: Purple congestion and petechiae in the superior vena cava territory.

Figure 2: Bilateral sub-conjunctival hemorrhage.

right humeral fracture. The electrocardiogram showed a sinus tachycardia. Arterial blood gases on 3 L/min facial mask oxygen showed: pH 7.41, PaCO2 36 mmHg, PaO2 180 mmHg. The blood cell count and the coagulation test were normal. Renal and liver function tests and troponin Ic were unremarkable.

The clinical and radiological presentation of our case was in favor of traumatic asphyxia syndrome, because of the mechanism of the trauma which was the compression of the chest between the ground and a heavy object and because of the presence of classical triad of traumatic asphyxia in the head and neck region. The differential diagnosis in our case was obstruction of the superior vena cava and the skull base fracture, these diagnosis were ruled out by cerebral and thoracic CT scan.

The patient was hospitalized in the intensive care unit and was monitored continuously. Support was symptomatic including facial mask oxygen therapy at 6 L/min, fluid replacement, and multimodal analgesia including paracetamol 15 mg/kg/6H associated to morphine 20 µg/kg/H. The head of the bed was elevated to 30 degrees to help venous drainage of the head and the neck. Pneumothorax was minimal, it spontaneously regressed and there were no indication to chest drainage. Consciousness of the patient gradually improved and he became alert after six hours, although agitation and confusion that lasted for one day. Tachypnea regressed 48 hours later. Thoracic X-ray showed disappearance of pneumothorax and contusions. The humeral fracture was not displaced and was treated by plaster. The outcome was favorable, marked by the decline of the facial edema after three days and the progressive disappearance of petechiae and conjunctival hemorrhages three weeks later.

DISCUSSION

Traumatic asphyxia is a type of mechanical asphyxia, where external pressure on the body inhibits respiratory movements and compromise venous return from the head. The thoracic compression must be preceded by a Valsalva maneuver. It is a rare syndrome, first described in 1937 by Oliver d'Angers as the ecchymotic mask. Others names are also used to describe this syndrome: Traumatic cyanosis, compressive cyanosis, traumatic apnea, Oliver's syndrome, and Perthes syndrome [1, 2].

Perthes syndrome is characterized by the association of edema and cyanosis of the head and neck, sub-conjunctival hemorrhage, and petechial hemorrhages of the face, neck and chest, secondary to a sudden compression of the thoracoabdominal region [3]. All of these findings were present in our patients. The weight and the duration and of compression affect the outcome. Important weight can be tolerated for a short period, whereas a lower weight associated to a longer period can result in severe consequences [4].

Our patient presented traumatic asphyxia due to a compression between the ground and a heavy object, this mechanism is common in reported cases. Other etiologies of Perthes syndrome are: motor vehicle crashes, crushing in a panicked crowd, entrapment beneath vehicles or falling down in a narrow space [5].

The combination of sudden increase in chest pressure and a deep breath with closed glottis leads to elevated pressure in the valveless head and neck venous system, which is responsible of venous stasis and breaking capillaries and veins [6]. The lower venous territory is protected by the presence of valves and by the obliteration or the compression of inferior vena cava after thoracic hyper pressure [4].

Perthes syndrome is frequently associated with other injuries: hemothorax, pneumothorax, pulmonary contusion, prolonged loss of consciousness, confusion and seizures, ophthalmic injuries such as retinal hemorrhages and visual loss [7].

Our patient presented pulmonary contusions, low abundance pneumothorax, and fractures of two ribs, with a minimal and transitory impact on respiratory function. This disparity between chest injuries and trauma mechanism is probably due to elastic chest cage in children.

Neurological involvement, which makes the severity of this syndrome, is common (90%). Its variable from confusion to coma, the frequency of neurological disorders contrast with the rarity of radiological findings [8]. The mechanism of neurological injury includes cerebral hypoxia, ischemia and venous hypertension, which lead to cortical dysfunction [2]. Usually, neurological events are reversible within 24–48 hours under early and adequate treatment [8]. Our patient had alteration of consciousness that lasted six hours, and was confused during 24 hours, but recovered a normal neurological status under symptomatic treatment.

Visual disturbances occur in some cases [9], secondary to the same mechanism as neurological involvement with multiple presentations: retinal hemorrhage, retrobulbar hemorrhage and vitreous exudates [2]. Therefore, ophthalmological follow-up is important. Our case had no ophthalmological abnormalities in fundoscopy.

The differential diagnosis of this syndrome includes obstruction of the superior vena cava, skull base fracture which clinical presentation contains: sub-conjunctival hemorrhage, periorbital ecchymosis, epistaxis and otorrhagia. Tamponade can also induce cyanosis, respiratory distress, but more likely hemodynamic instability [10].

Traumatic asphyxia cases should be monitored after securing the airway. Oxygen therapy and fluid replacement need to be initiated and the patient shall be intubated and followed on mechanical ventilation as needed [1]. The management should include the elevation of the head at 30 degrees; and specific treatments may be needed for associated injuries.

The elasticity of children chest makes the difference of this syndrome in comparison with adults. Thus, in some pediatric cases, even with severe chest and abdominal compression, thoracic lesions were not associated with rib fracture [10]. In children, the prognosis is generally favorable in the absence of severe associated lesions, with the exception of possible visual sequelae, and the mortality rate is usually low [4].

CONCLUSION

Traumatic asphyxia should always be considered as a possible complication of injuries of the chest and abdomen. The prognosis of this syndrome depends on the nature and duration of the compressive force and the presence of others injuries. However, despite the dramatic appearance of Perthes syndrome, mortality remains low, especially in children, due to chest elasticity.

Keywords: Pediatrics, Perthes syndrome, Trauma, Traumatic asphyxia

How to cite this article

Berdai MA, Labib S, Harandou M. Traumatic asphyxia: A rare syndrome in trauma children. Int J Case Rep Images 2017;8(2):151–154.

Article ID: Z01201702CL10113MB

****

doi:10.5348/ijcri-201703-CL-10113

****

Author Contributions

Mohamed Adnane Berdai – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, revising critically for important intellectual content, Final approval of the version to be published
Smael Labib – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published
Mustapha Harandou – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

Copyright

© 2017 Mohamed Adnane Berdai et al. This article is distributed under the terms of Creative Commons Attribution License which permits unrestricted use, distribution and reproduction in any medium provided the original author(s) and original publisher are properly credited. Please see the copyright policy on the journal website for more information.

REFERENCES

1. Gulbahar G, Kaplan T, Gundogdu AG, et al. A rare and serious syndrome that requires attention in emergency service: Traumatic asphyxia. Case Rep Emerg Med 2015;2015:359814.
2. Sertaridou E, Papaioannou V, Kouliatsis G, Theodorou V, Pneumatikos I. Traumatic asphyxia due to blunt chest trauma: A case report and literature review. J Med Case Rep 2012 Aug 30;6:257.
3. Richards EC, Wallis DN. Asphyxiation: A review. Trauma 2005;7(1):37–45.
4. Nishiyama T, Hanaoka K. A traumatic asphyxia in a child. Can J Anaesth 2000;47:1196–201.
5. Byard RW, Wick R, Simpson E, Gilbert JD. The pathological features and circumstances of death of lethal crush/traumatic asphyxia in adults–a 25-year study. Forensic Sci Int 2006 Jun 2;159(2-3):200–5.
6. Karamustafaoglu YA, Yavasman I, Tiryaki S, Yoruk Y. Traumatic asphyxia. Int J Emerg Med 2010 Aug 25;3(4):379–80.
7. Eken C, Yigit O. Traumatic asphyxia: A rare syndrome in trauma patients. Int J Emerg Med 2009 Aug 1;2(4):255–6.
8. Barakat M, Belkhadir ZH, Belkrezia R, et al. Traumatic asphyxia or Perthe's syndrome: Six case reports. [Article in French]. Ann Fr Anesth Reanim 2004 Feb;23(1):59–62.
9. Baldwin GA, Macnab AJ, McCormick AQ. Visual loss following traumatic asphyxia in children. J Trauma 1988 Apr;28(4):557–8.
10. El koraichi A, Benafitou R, Tadili J, et al. Traumatic asphyxia or Perthe's syndrome: About two paediatric cases. [Article in French]. Ann Fr Anesth Reanim 2012 Mar;31(3):259–61.

Access full text article on other devices

Access PDF of article on other devices

CLINICAL IMAGES PEER REVIEWED | OPEN ACCESS

Anomalous origin of left circumflex coronary artery: An easy 'pick' on transthoracic echocardiography

Keyur Vora, Alok Ranjan

CASE REPORT

A 52-year-old male with a personal history of smoking and systemic arterial hypertension since two years presented with what he described as a squeezing pain in the left side of his chest. He also had associated dizziness and diaphoresis. An initial electrocardiogram (ECG) revealed acute inferior wall infarction with sinus bradycardia. As per the institutional protocol, a transthoracic echocardiography was performed prior to coronary angiography. Transthoracic 2D-echocardiography was consistent with inferior wall myocardial infarction and no significant mitral regurgitation.

Remarkably, in apical five-chamber view, a prominent vessel was seen; arising from right side of aortic root; entering into left atrioventricular (AV) groove (Figure 1 A–B; blue arrows). Part of this vessel was also apparent in apical two chamber view and in parasternal long axis views (Figure C; red dotted lines, red arrow and D; green arrow). An anomalous origin of coronary artery was diagnosed. The course of the vessel towards left AV groove was suggestive of left circumflex coronary artery (LCX). In parasternal short axis (PSAX) view, left main coronary artery (Figure E; orange arrow) and right coronary artery (Figure E; yellow arrow) were arising from their respective sinuses. The origin of the anomalous LCX was not seen in PSAX view. Finally, the anomalous LCX was confirmed on coronary angiography (CAG), arising from ostioproximal part of right coronary artery (Figure F; white arrow).

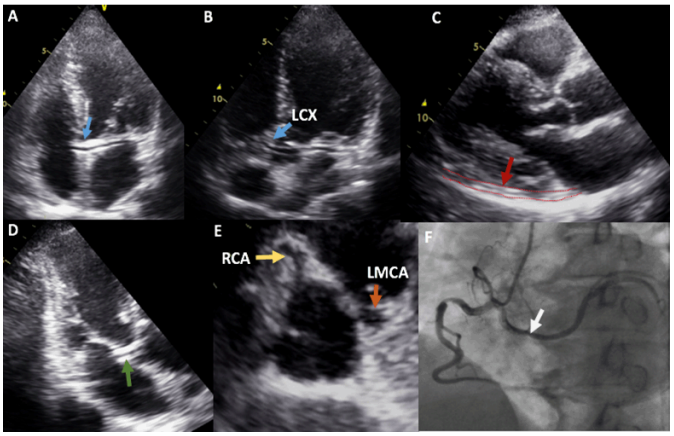


Figure 1: (A) Anomalous coronary artery on echocardiography. In apical four-chamber view, a coronary artery is found arising from right side of aortic root, (blue arrow), (B) Anomalous coronary artery on echocardiography. In apical five-chamber view, a coronary artery is found arising from right side of aortic root, entering into left atrioventricular groove (blue arrow), (C) Course of anomalous coronary artery. The part of the anomalous course of coronary artery was also depicted on parasternal long axis (PLAX) view (red dotted lines), (D) Course of anomalous coronary artery. The part of the anomalous course of coronary artery was also depicted on apical two-chamber view (green arrow), (E) Origin of coronary arteries. Left main coronary artery (LMCA) is arising from left coronary sinus as shown on parasternal short axis view (orange arrow), RCA is arising from right coronary sinus as shown on parasternal short axis view (yellow arrow), and (F) Anomalous artery on coronary angiogram. Anomalous origin of left circumflex coronary artery is depicted on conventional coronary angiogram (white arrow).

Keyur Vora[1], Alok Ranjan[1]

Affiliations: [1]Department of Cardiology, CARE Hospitals, the Institute of Medical Sciences, Surat, Gujarat, India.

Corresponding Author: Keyur Vora, Department of Cardiology, CARE Hospitals, the Institute of Medical Sciences, Surat, Gujarat 395009, India; Email: drkeyurvora@gmail.com

Received: 28 September 2016
Accepted: 21 October 2016
Published: 01 February 2017

DISCUSSION

Anomalous origin of LCX from right coronary sinus is the most common congenital variant with prevalence of 0.18–0.67% [1]. An aberrant but normal LCX arising from the right coronary sinus (common or separate ostium with the RCA) has no clinical significance per se, and it does not predispose the LCX to a higher incidence of obstructive disease [2]. Although the LCX anomaly

is classified as benign and asymptomatic, it can cause myocardial ischemia, and in some cases sudden death, myocardial infarction, and angina pectoris in the absence of atherosclerotic lesions. These manifestations might be due to repeated compression of the anomalous artery by a dilated aortic root or to unusual angling as a result of the retroaortic course of the LCX, which can compress the coronary ostium and restrict blood flow.

On the other hand, the presence of obstructive disease, however, especially in a vessel of large distribution, makes it mandatory that the anomaly be recognized and angiographically demonstrated, especially in acute myocardial infraction. Sometimes, in acute myocardial infraction, no evidence of an occluded coronary artery can be seen during angiography. This might lead to a large spectrum of differential diagnoses to explain the acute chest pain or the electrocardiogram modification [3]. The absence of an epicardial vessel or its branch, which is anatomically supposed to supply a myocardial ischemic area identified at the left ventriculography, suggests that an anatomical variation of the normal coronary tree (i.e. the aberrant artery) has to be actively searched.

Computed tomography angiography (CTA) is more useful than conventional angiography. Three dimensional information of the course of the coronary arteries in relationship to the great vessels and the origin are clearly detected by CTA. The transthoracic echocardiography is most limited in such cases as it can only detect part of the anomalous course of the artery. Usually, the origins of coronary arteries from its respective coronary sinuses are relatively easy to detect on transesophageal echocardiography. The subsequent course of artery is even more difficult and most of the times only a proximal part of the course is detected on TTE. The distal course and intramural course of the artery are not seen on TTE. The role of TTE is even more limited in adult as compared to children in detecting coronary abnormalities.

Transesophageal echocardiography may be more useful in detecting the origin and the proximal part of coronary arteries but the subsequent course is even more difficult to detect. Transesophageal echocardiography offers several advantages that may potentially overcome the technical problems associated with the transthoracic approach, including closer proximity of the transducer to the proximal coronary arteries and avoidance of anterior chest wall structures that cause degradation of the ultrasonic signal. This allows the routine use of higher frequency transducers and thus better spatial resolution and more detailed image quality. With these advantages, TEE is only helpful in detecting proximal coronary abnormalities [4].

CONCLUSION

In conclusion, our imaging experience highlights the significance of evaluation of coronary arteries on emergency echocardiography study and proactive preparation for uneventful interventional procedures. Careful evaluation of coronary arteries on echocardiography is inexpensive, quick and time saving modality as well as contrast and radiation exposure is saved. Such a high level of anticipation can be extremely useful information for emergency percutaneous coronary interventions (PCI). The reliable anticipation and identification is of paramount importance to the interventional cardiologists. Appropriate anatomical and technical understanding is vital for a successful interventional treatment of anomalous coronary arteries.

Keywords: Anomalous Coronary Artery, Acute Coronary Syndrome, Transthoracic Echocardiography

How to cite this article

Vora K, Ranjan A. Anomalous origin of left circumflex coronary artery: An easy 'pick' on transthoracic echocardiography. Int J Case Rep Images 2017;8(2):155–157.

Article ID: Z01201702CL10114KV

****

doi:10.5348/ijcri-201704-CL-10114

****

Author Contributions

Keyur Vora – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting of article, Revising it critically for important intellectual content, Final approval of the version to be published

Alok Ranjan – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting of article, Revising it critically for important intellectual content, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

Copyright

© 2017 Keyur Vora et al. This article is distributed under the terms of Creative Commons Attribution License which permits unrestricted use, distribution and reproduction in any medium provided the original author(s) and original

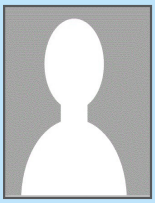
publisher are properly credited. Please see the copyright policy on the journal website for more information.

REFERENCES

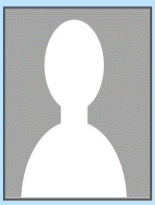
1. Dursunoglu D, Ozalp G, Tasköylü O, Semiz E. Anomalous origin of the left circumflex coronary artery: A case report. Exp Clin Cardiol 2007 Winter;12(4):207–8.
2. Page HL Jr, Engel HJ, Campbell WB, Thomas CS Jr. Anomalous origin of the left circumflex coronary artery: Recognition, antiographic demonstration and clinical significance. Circulation 1974 Oct;50(4):768–73.
3. Hendiri T, Alibegovic J, Bonvini RF, Camenzind E. Successful angioplasty of an occluded aberrant coronary artery: A rare cause of acute myocardial infarction. Acute Card Care 2006;8(2):125–7.
4. Samdarshi TE, Nanda NC, Gatewood RP Jr, et al. Usefulness and limitations of transesophageal echocardiography in the assessment of proximal coronary artery stenosis. J Am Coll Cardiol 1992 Mar 1;19(3):572–80.

ABOUT THE AUTHORS

Article citation: Vora K, Ranjan A. Anomalous origin of left circumflex coronary artery: An easy 'pick' on transthoracic echocardiography. Int J Case Rep Images 2017;8(2):155–157.



Keyur P. Vora is an Internist and Imaging Cardiologist at CARE Institute of Medical Sciences, India. He holds MS Bioinformatics degree from Georgia State University, Atlanta as well as Postdoctoral Research Fellowship from Emory University/CDC, Atlanta USA. His research interests include echocardiography, Cardiac CT, Cardiac MR studies and computational medicine projects.
E-mail: drkeyurvora@gmail.com



Alok Ranjan, Department of Cardiology, CARE Hospitals, the Institute of Medical Sciences, Surat, Gujarat, India.

Access full text article on other devices

Access PDF of article on other devices

CLINICAL IMAGES PEER REVIEWED | OPEN ACCESS

Ochronotic arthropathy

Kayahan Karaytug, Eren Yildiz

CASE REPORT

A 56-year-old female was referred to our clinic with severe pain on her right knee. She was admitted to physiotherapy and took analgesics previously. On physical examination, there was moderate pain on flexion and extension of the right knee with crepitations. Osteoarthritis was detected in X-ray of both knees (Figure 1A). After conservative treatment there was no significant progress. We offered surgical treatment and after the patient confirm the surgery, cemented total knee replacement surgery was performed. During the operation black discoloration of the synovial tissue, capsules and surfaces of the tibiofemoral joint was observed (Figure 1B–C). Microbiological analysis detected no organism. Some analyses were made for metabolic diseases because of discoloration of the soft tissue and the bones. Blood and urine samples were taken for screening. Metabolic disease was suspected due to organic acids in urine screening. Urine levels of homogentisic acid was 20 times higher than upper level of normal range. A diagnosis of alkaptonuria was made. Histopathological examination was consistent with ochronotic arthropathy (Figure 1D). A childhood metabolic disease called alkaptonuria was diagnosed in a 56-year-old patient, due to findings during surgery. Postoperative two years later physical examination showed 0–120° range of motion and patient reported no pain with normal findings on X-ray (Figure 1E).

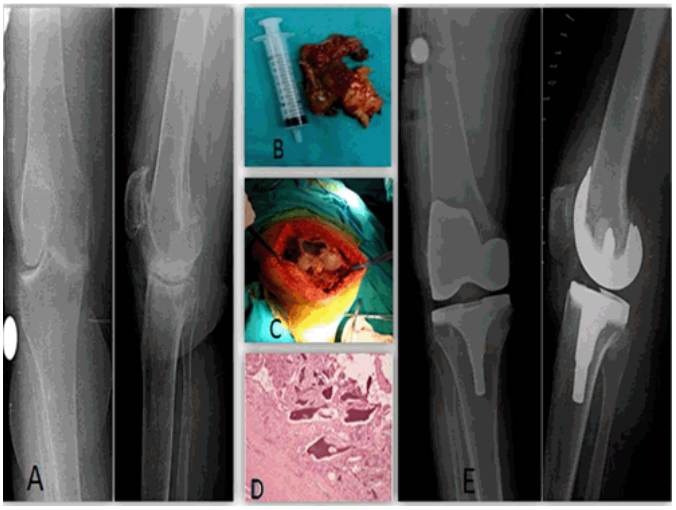


Figure 1: (A) Preoperative anteroposterior and lateral radiographs, (B) Black discoloration of the synovial tissue, capsules, (C) Black discoloration of surface of the tibiofemoral joint and the femur, (D) Mononuclear cell infiltration in focal focus is observed around the synovial cartilage fragments, and (E) Postoperative anteroposterior and lateral radiographs of the patient.

DISCUSSION

Alkaptonuria is rare autosomal recessive metabolic disease [1]. Homogentisic acid deficiency occurs due to mutations in the gene 1-2 dioxygenase [2]. It is seen in one out of one million population [3, 4]. Alkaptonuria is a disease caused due to deficiency of the homogentisic acid oxidase enzyme. Homogentisic acid polymers condense in the urine and accumulate in the urine (called alkaptonuria), soft tissue and connective tissues as brownish black pigmentation (called ochronosis). Changes in cartilage, internal organs and osteoporosis are lead to pathognomonic change [5]. Alkaptonuria is often the first sign of Madiran darkening of the urine [6].

Ochronotic arthropathy is a manifestation of the alkaptonuria developed after a long period of time. Ochronotic homogentisic acid occurs in the articular

Kayahan Karaytug[1], Eren Yildiz[2]
Affiliations: [1]State Hospital, Department of Orhopedic Surgery Sarıkamış-Kars, Turkey; [2]State Hospital, Department of Orhopedic Surgery Nusaybin-Mardin, Turkey.
Corresponding Author: Kayahan Karaytuğ, Sarıkamış, State Hospital, Yeni Mahalle Erzurum Yolu, Kars, Turkey; Email: Kayhan_karaytug@hotmail.com

Received: 04 October 2016
Accepted: 02 November 2016
Published: 01 February 2017

cartilage with increasing age as a result of osteoarthritis. In this case, there were observed black soft tissue and bone tissue during knee joint replacement surgeries. Tissue sampling was done. Histopathological examination diagnosed with ochronosis and alkaptonuria diagnosed in the urine sample taken from the patient was placed as a result. This metabolic disease is diagnosed in early age of life. In these cases, based on the findings of secondary diagnosis at an advanced age was put alkaptonuria

CONCLUSION

Ochronosis is a rare metabolic disease involving the periphery joints. Although there is no definitive treatment, joint replacement is one of the most effective treatments recommended. It provides painless quality of life and patients have close to a full joint motion.

Keywords: Alkaptonuria, Black discoloration, Ochronotic arthropathy, Total knee replacement

How to cite this article

Karaytug K, Yildiz E. Ochronotic arthropathy. Int J Case Rep Images 2017;8(2):158–160.

Article ID: Z01201702CL10115SK

****

doi:10.5348/ijcri-201705-CL-10115

****

Author Contributions

Kayahan Karaytug – Substantial contributions to conception and design, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published
Eren Yildiz – Substantial contributions to conception and design, Drafting the article, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

Copyright

© 2017 Kayahan Karaytug et al. This article is distributed under the terms of Creative Commons Attribution License which permits unrestricted use, distribution and reproduction in any medium provided the original author(s) and original publisher are properly credited. Please see the copyright policy on the journal website for more information.

REFERENCES

1. Keller JM, Macaulay W, Nercessian OA, Jaffe IA. New developments in ochronosis: Review of the literature. Rheumatol Int 2005 Mar;25(2):81–5.
2. Phornphutkul C, Introne WJ, Perry MB, et al. Natural history of alkaptonuria. N Engl J Med 2002 Dec 26;347(26):2111–21.
3. Sahin G, Milcan A, Bagis S, Köktürk A, Pata C, Erdogan C. A case of ochronosis: upper extremity involvement. Rheumatol Int 2001 Oct;21(2):78–80.
4. Manoj Kumar RV, Rajasekaran S. Spontaneous tendon ruptures in alkaptonuria. J Bone Joint Surg Br 2003 Aug;85(6):883–6.
5. Acar MA, Erkocak OF, Aydin BK, Altan E, Senaran H, Elmadag NM. Patients with black hip and black knee due to ochronotic arthropathy: Case report and review of literature. Oman Med J 2013 Nov;28(6):448–9.
6. Resnick D. Alkaptonuria. In: Resnick D, Niwayama G eds. Diagnosis of Bone and Joint Disorders. Philadelphia, PA: WB Saunders; 1988. p. 1787–803.

ABOUT THE AUTHORS

Article citation: Karaytug K, Yildiz E. Ochronotic Arthropathy. Int J Case Rep Images 2017;8(2):158–160.

Kayahan Karaytug is Specialist of Orthopaedics and Traumatology, Sarikamıs State Hospital Kars, Turkey.
E-mail: kayhan_karaytug@hotmail.com

Eren Yildiz is Specialist of Orthopaedics and Traumatology, Nusaybin State Hospital Mardin, Turkey.

Access full text article on other devices

Access PDF of article on other devices

CLINICAL IMAGES PEER REVIEWED | OPEN ACCESS

Perforated inferior vena cava filter removal by concurrent femoral and internal jugular vein approaches

Raymond Yap, Ankur Sidhu, Mark Brooks, Mehrdad Nikfarjam

CASE REPORT

It is commonly accepted that insertion of inferior vena cava (IVC) filters is safe, effective and their use is constantly increasing [1]. Inferior vena cava filters are commonly used adjuncts in the prevention of pulmonary embolism from deep vein thrombosis, particularly in the setting of gastrointestinal malignancy. Although their use is widespread, there is some misunderstanding in regards to the commonality of potential serious consequences from their deployment. We highlight the pitfalls in their use, as well as a technique for percutaneous removal of filter that may have migrated beyond the lumen of the IVC, using a combined internal jugular and femoral vein approach.

A 70-year-old female with colorectal liver metastases had an inferior vena caval (IVC) filter inserted prior to a major liver resection. She had developed a deep vein thrombosis (DVT) and pulmonary embolus (PE) four months previously following a right hemicolectomy, despite perioperative use of graduated compression stockings, intermittent pneumatic compression and use of low molecular weight heparin (LMWH). Following the diagnosis of DVT and pulmonary embolus, she was treated by with therapeutic LMWH, while receiving neoadjuvant chemotherapy, prior to her liver surgery. It was anticipated that she was at high risk of developing further thrombus in the setting of prolonged abdominal surgery, and it was decided that she should have an IVC filter inserted (Cook Celect® Vena Cava filter, Cook medical, Brisbane, QLD, Australia). She had an uncomplicated liver operation and a decision was made for her to have adjuvant chemotherapy prior to considering removal of her IVC filter. She received therapeutic does of LMWH during her postoperative follow-up.

At six months following completion of adjuvant therapy the patient had restaging computed tomography imaging of the chest, abdomen and pelvis that showed no evidence of disease recurrence. The imaging did, however, demonstrate perforation of the tines of the filter through the walls of the inferior vein cava (Figure 1), with one in close proximity to the aorta and resulting in some aortic wall thickening. The filter had also become tilted and its removal hook was projecting through the anterior wall of the cava. These findings were not present on the initial postoperative computed tomography scan. She was asymptomatic, but there were concerns of possible future complications, in particular the development of aorto-caval fistula.

An attempt to remove the filter was made percutaneously through the right internal jugular vein approach. Venogram confirmed that the tines and hook of the filter lying outside the vein. Initial attempts to snare the filter with a retrieval kit were unsuccessful through both internal jugular and femoral approaches. A second percutaneous attempt was made with a combined right internal jugular and femoral vein approach under general anesthesia (Figure 2). Two wires were looped under the filter legs from a 16Fr internal jugular sheath. A 3rd wire was passed from the jugular sheath to an 8Fr femoral vein sheath. With the filter trapped between the tips of the two sheaths it was mobilized with repeated cranial and caudal movements. The tines and the hook were thus maneuvered back into the IVC lumen. Once this was accomplished, the filter was enclosed by a snare. The IVC filter was then inverted and removed through the right femoral vein. A completion venogram showed no injury to the IVC or contrast extravasation. The patient's stay

Raymond Yap[1], Ankur Sidhu[1], Mark Brooks[2], Mehrdad Nikfarjam[1]

Affiliations: [1]Department of Surgery, University of Melbourne Austin Health, Heidelberg, Melbourne, Victoria, Australia; [2]University of Melbourne Department of Radiology, Austin Health, Heidelberg, Melbourne, Victoria, Australia.

Corresponding Author: Mehrdad Nikfarjam MD, PhD, FRACS, University Department of Surgery, Austin Health, LTB 8, Studley Rd, Heidelberg Melbourne, Victoria 3084, Australia; Ph: +613 9496 5466; Fax: +613 9458 1650; Email: mehrdad.nikfarjam@gmail.com

Received: 29 September 2016
Accepted: 16 November 2016
Published: 01 February 2017

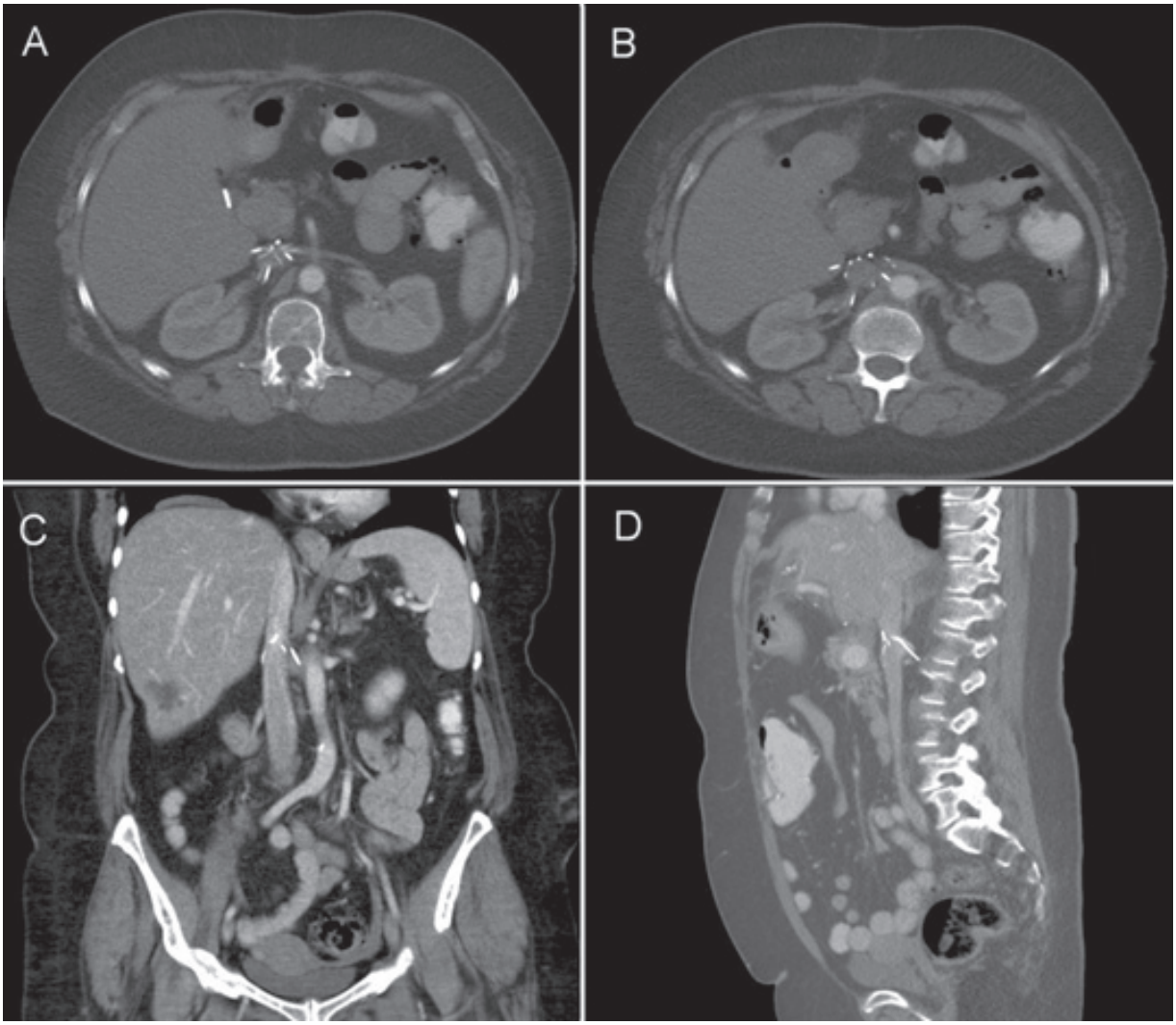


Figure 1: (A–D). Computed tomography scan showing perforation of vena cava by filter tines the close proximity of the tines to the aorta can be appreciated (B, C). Note the hook of the filter outside the lumen (D).

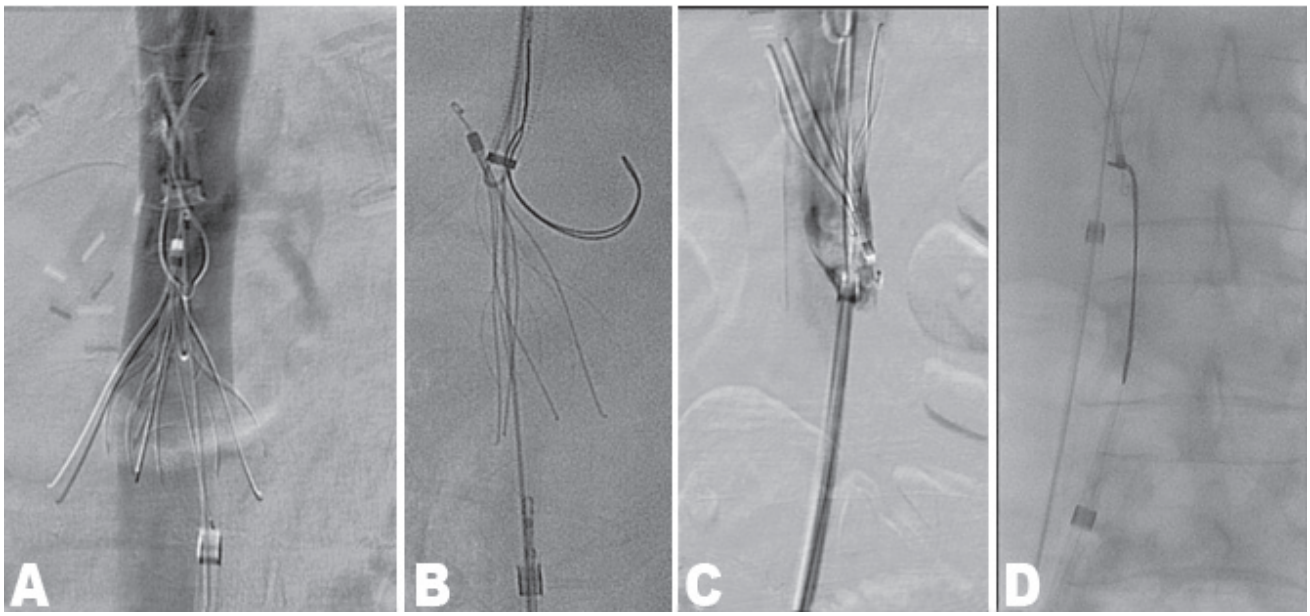


Figure 2: (A–D) Filter grasped though simultaneous internal jugular and femoral vein approaches and dislodged. The filter was then rotated to enable removal through the femoral vein.

had no complications and was discharged home the following day.

DISCUSSION

Inferior vena cava (IVC) filters have been well established as useful alternatives or adjuncts to anticoagulation for deep vein thrombosis to prevent pulmonary embolism [1]. They are generally considered safe. There are, however, numerous case reports in literature of serious complications including IVC filter perforation into the aorta or duodenum, stent migration and occlusion [2]. These complications can have serious consequences, including death and significant morbidity.

Durack et al. [3] published a case series of 50 IVC filters which had CT scans for reasons other than concerns of stent complications. Eighty-six percent of filters showed perforation through the IVC on review CT scan, and the earliest of this was at six days. All filters imaged after 71 days showed a degree of perforation which appeared to be progressive. Although this is a small case series using one type of filter, it does demonstrate that caution should be used in IVC filter use. The clinical significance of CT-detected IVC filter perforation is not known, however, due to the possible catastrophic complications, these findings should always be taken seriously.

Our patient had no symptoms related to her filter, but we had concerns for future complications if we were to leave it in situ. While we have shown it is possible to remove an IVC filter with tines and hook projecting beyond the cava the procedure was prolonged and difficult and carried some potential risk. We strongly suggest that IVC filters should be removed as soon as the clinical need for them has passed.

If their use is to continue, past three months interval CT imaging should be considered even if the patient is asymptomatic and if perforation is detected, strong consideration should be given to removal of the IVC filter. The technique described using concurrent internal jugular and femoral vein approach appears to be useful method to consider for the difficult to remove IVC filter, even in the setting tine perforation.

Further research is needed to delineate the natural history of IVC filters and perforation. While the current generation of removable IVC filters appear safe and effective for short-term protection from pulmonary embolus more work is required to develop removable filters which also perform safely as long-term devices.

CONCLUSION

Vena caval filer perforation of the vena cava by filter tines can occur. Filter removal can be difficult in some cases and a combined internal jugular and femoral vein approach may be required for removal.

Keywords: Deep venous thrombosis, Filter, Pulmonary emboli, Inferior Vena Cava

How to cite this article

Yap R, Sidhu A, Brooks M, Nikfarjam M. Perforated inferior vena cava filter removal by concurrent femoral and internal jugular vein approaches. Int J Case Rep Images 2017;8(2):161–164.

Article ID: Z01201702CL10116RY

****

doi:10.5348/ijcri-201706-CL-10116

****

Author Contributions

Raymond Yap – Acquisition of data, Drafting the article, Final approval of the version to be published
Ankur Sidhu – Acquisition of data, Revising it critically for important intellectual content, Final approval of the version to be published
Mark Brooks – Substantial contributions to conception and design, Revising it critically for important intellectual content, Final approval of the version to be published
Mehrdad Nikfarjam – Substantial contributions to conception and design, Revising it critically for important intellectual content, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

Copyright

© 2017 Raymond Yap et al. This article is distributed under the terms of Creative Commons Attribution License which permits unrestricted use, distribution and reproduction in any medium provided the original author(s) and original publisher are properly credited. Please see the copyright policy on the journal website for more information.

REFERENCES

1. Athanasoulis CA, Kaufman JA, Halpern EF, Waltman AC, Geller SC, Fan CM. Inferior vena caval filters: Review of a 26-year single-center clinical experience. Radiology 2000 Jul;216(1):54–66.

2. Greenfield LJ, Proctor MC. Filter complications and their management. Semin Vasc Surg 2000 Sep;13(3):213–6.
3. Durack JC, Westphalen AC, Kekulawela S, et al. Perforation of the IVC: Rule rather than exception after longer indwelling times for the Günther Tulip and Celect retrievable filters. Cardiovasc Intervent Radiol 2012 Apr;35(2):299–308.

Access full text article on other devices

Access PDF of article on other devices

CLINICAL IMAGES PEER REVIEWED | OPEN ACCESS

Renal cell carcinoma of the native kidney in a renal transplant recipient

Vikrampal Bhatti, Ananthalaxmi Vangapalli, Deepti Bhattacharya, Abhilash Koratala

CASE REPORT

A 73-year-old white male was seen for fever, back pain and dysuria of three-day duration. His past medical history was significant for end stage renal disease secondary to hypertensive nephropathy status post deceased donor kidney transplant ten years ago and multiple skin cancers status post resection. His maintenance immunosuppression included tacrolimus 3 mg twice a day, azathioprine 75 mg daily and prednisone 10 mg daily. He was treated with antibiotics for Urinary tract infection. Ultrasound of the transplanted kidney was normal and the serum creatinine was at baseline. However, native kidney ultrasound done because of the back pain showed ~6 cm mass in the right kidney (Figure 1A). He never had gross or microscopic hematuria. His symptoms improved but MRI scan of abdomen showed heterogeneous mass (5.2x5.1x4.8 cm) in the superior pole of the right kidney suggestive of renal cell carcinoma along with bony lesions suspicious for metastases. Also, there were multiple cysts in both native kidneys and he did not have cystic renal disease prior to transplant (Figure 1B–C). He developed acquired cystic renal disease of native kidneys which unfortunately, was never screened for.

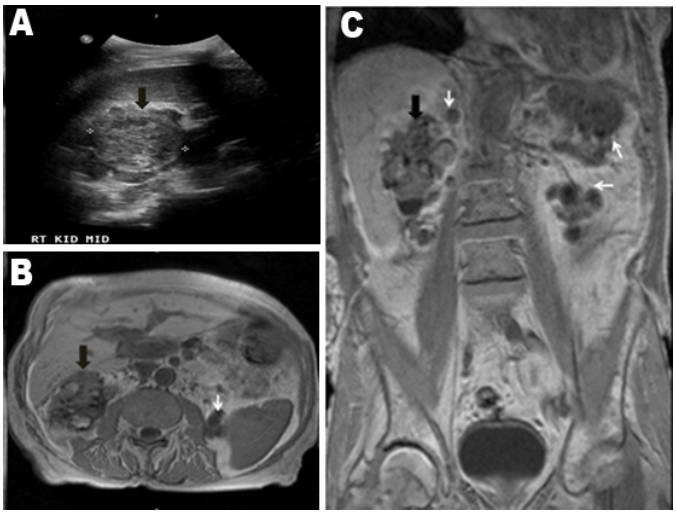


Figure 1: (A) Renal ultrasound showing the mass in the right kidney (arrow), (B, C) MRI abdomen showing heterogeneous mass suggestive of Renal cell carcinoma (black arrow) and bilateral native kidney cysts (arrows).

DISCUSSION

The incidence of renal cell carcinoma is increased in kidney transplant recipients and, particularly those with acquired cystic disease of the kidneys [1]. Chronic renal failure (particularly in patients on maintenance dialysis) is frequently associated with the development of multiple and bilateral renal cysts [2, 3]. There is conflicting data on whether to screen patients with advanced kidney disease for acquired cystic disease and thereby early detection of premalignant or malignant lesions [4–6]. Current major guidelines do not recommend routine screening of average risk patients for renal cell carcinoma. The learning point from our case is that till we have detailed guidelines designed by the major international transplantation societies, it is prudent to screen transplant recipients with periodic ultrasound of the native kidneys (6 months to 1 year) [7] irrespective of the presence or absence of microscopic hematuria. In fact, a Japanese study has shown that in dialysis patients with renal cell carcinoma, the survival rate in the group detected by screening was better than that in the group detected by symptoms [8]. If the ultrasound demonstrates cysts, a computed tomography (CT) scan with contrast or magnetic resonance imaging (MRI) scan

Vikrampal Bhatti[1], Ananthalaxmi Vangapalli[2], Deepti Bhattacharya[1], Abhilash Koratala[1]

Affiliations: [1]University of Florida, Gainesville, USA; [2]Hebei Medical University, China.

Corresponding Author: Abhilash Koratala, M.D., Division of Nephrology, Hypertension and Renal Transplantation, P.O. Box 100224, Gainesville, FL 32610, USA; E-mail: akoratsla@ufl.edu

Received: 21 October 2016
Accepted: 24 November 2016
Published: 01 February 2017

without gadolinium may be performed at yearly intervals especially in patients with good life expectancy and those with larger lesions to screen for the possible development of malignancy [6, 9]. Urology consultation should be sought for patients with Bosniak class III and IV cysts, to discuss the surgical options including total nephrectomy or nephron-sparing (partial) nephrectomy.

CONCLUSION

Our case of renal cell carcinoma that developed in a renal transplant patient and acquired cystic disease of the native kidneys emphasizes the need for establishment of routine screening protocol in such patients.

Keywords: Acquired cystic renal disease, Renal cell carcinoma, Screening

How to cite this article

Bhatti V, Vangapalli A, Bhattacharya D, Koratala A. Renal cell carcinoma of the native kidney in a renal transplant recipient. Int J Case Rep Images 2017;8(2):165–167.

Article ID: Z01201702CL10117VB

****

doi:10.5348/ijcri-201707-CL-10117

****

Author Contributions

Vikrampal Bhatti – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published
Ananthalaxmi Vangapalli – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published
Deepti Bhattacharya – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published
Abhilash Koratala – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

Copyright

© 2017 Vikrampal Bhatti et al. This article is distributed under the terms of Creative Commons Attribution License which permits unrestricted use, distribution and reproduction in any medium provided the original author(s) and original publisher are properly credited. Please see the copyright policy on the journal website for more information.

REFERENCES

1. Wong G, Chapman JR, Craig JC. Cancer screening in renal transplant recipients: What is the evidence? Clin J Am Soc Nephrol 2008 Mar;3 Suppl 2:S87–S100.
2. Ishikawa I. Acquired cystic disease: Mechanisms and manifestations. Semin Nephrol 1991 Nov;11(6):671–84.
3. Matson MA, Cohen EP. Acquired cystic kidney disease: Occurrence, prevalence, and renal cancers. Medicine (Baltimore) 1990 Jul;69(4):217–26.
4. Sarasin FP, Wong JB, Levey AS, Meyer KB. Screening for acquired cystic kidney disease: A decision analytic perspective. Kidney Int 1995 Jul;48(1):207–19.
5. Truong LD, Krishnan B, Cao JT, Barrios R, Suki WN. Renal neoplasm in acquired cystic kidney disease. Am J Kidney Dis 1995 Jul;26(1):1–12.
6. Marple JT, MacDougall M, Chonko AM. Renal cancer complicating acquired cystic kidney disease. J Am Soc Nephrol 1994 Jun;4(12):1951–6.
7. Filocamo MT, Zanazzi M, Li Marzi V, et al. Renal cell carcinoma of native kidney after renal transplantation: Clinical relevance of early detection. Transplant Proc 2009 Dec;41(10):4197–201.
8. Ishikawa I, Honda R, Yamada Y, Kakuma T. Renal cell carcinoma detected by screening shows better patient survival than that detected following symptoms in dialysis patients. Ther Apher Dial 2004 Dec;8(6):468–73.
9. Levine E, Slusher SL, Grantham JJ, Wetzel LH. Natural history of acquired renal cystic disease in dialysis patients: A prospective longitudinal CT study. AJR Am J Roentgenol 1991 Mar;156(3):501–6.

Access full text article on other devices

Access PDF of article on other devices

LETTERS TO THE EDITOR PEER REVIEWED | OPEN ACCESS

The gastrocnemius muscle flap for coverage of soft tissue defect of the proximal third of lower leg

Ingo Schmidt

To the Editor,

Postoperative soft tissue defect with exposure of relevant structures such as bone with or without osteosynthesis plates of the proximal third of lower leg represents a challenging problem. A 57-year-old male presented with a highly comminuted open fracture of the proximal right tibia (Figure 1A). First, the fracture was stabilized by knee joint-bridging external fixation. After four debridements and negative-pressure vacuum assisted closure (VAC) therapies including incorporation of polymethyl methacrylate (PMMA) beads containing gentamycin (Figure 1B), the pre-tibial soft tissue defect could be covered with a medial gastrocnemius muscle flap and additional splitted skin grafts (Figure 1C). Then, the fracture was definitively treated with open reduction and internal fixation (ORIF). After eight weeks of injury, there was uncomplicated fracture and wound healing with complete restoration of knee joint function (Figure 1D–E), and 12 weeks after injury the patient could be mobilized with full weight-bearing on the affected leg.

The use of local flaps for coverage of soft tissue defects of the proximal third of lower leg and knee is an option for treatment in patients who are not willing or healthy enough to undergo free microvascular tissue transplantation, and do not require microsurgical expertise. The use of the gastrocnemius muscle flap is one method of choice for reconstruction [1]. There is only one vasculonervous pedicle for each of both muscle heads composed of a sural artery and one or two veins, and is classified as type I according to the classification of Mathes and Nahai [2]. It is possible to divide the muscle in two sections longitudinally according to the needs. However, the lateral head has to be rotated around the proximal fibula, therefore, it has a lower rotation angle than the medial head. There is an option to safely harvest a skin paddle overlying the muscle [3]. The gastrocnemius muscle flap is probably one of the safest flap, however, muscle flaps for reconstruction of legs are generally not free of any complications. Neale et al. [4] reported on major and minor complications in 32% of a total of 95 muscle flaps and they agreed that the causes were mainly technical errors, inadequate debridement, use of diseased and traumatized muscle, and unrealistic objectives. When

Ingo Schmidt

Affiliation: SRH Poliklinik Gera GmbH, Straße des Friedens 122, 07548 Gera, Germany.

Corresponding Author: Dr. Ingo Schmidt, SRH Poliklinik Gera GmbH, Straße des Friedens 122, 07548 Gera, Germany; Email: schmidtingo62@googlemail.com

Received: 01 July 2016
Accepted: 11 November 2016
Published: 01 February 2017

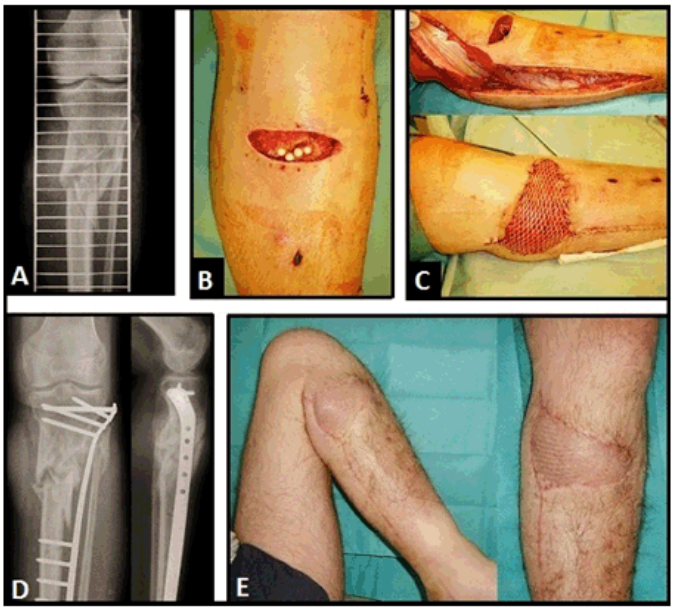


Figure 1: (A) Posteroanterior radiograph demonstrating highly comminuted fracture of proximal tibia, (B) Clinical photograph showing soft tissue defect at the ventral aspect of proximal tibia with incorporated polymethyl methacrylate beads, (C) Clinical photographs showing the harvest and transposition after skin grafting of the medial gastrocnemius head, (D) Posteroanterior and lateral radiographs demonstrating fracture healing after open reduction and internal fixation, (E) Clinical photographs showing uncomplicated wound healing and complete restoration of knee joint function.

a gastrocnemius muscle flap is not indicated, the use of random pattern skin transposition flaps is one salvage option [5].

Keywords: Gastrocnemius muscle flap, Proximal third lower leg, Soft tissue defect

How to cite this article

Schmidt I. The gastrocnemius muscle flap for coverage of soft tissue defect of the proximal third of lower leg. Int J Case Rep Images 2017;8(2):168–170.

Article ID: Z01201702LE10024IS

****

doi:10.5348/ijcri-201708-LE-10024

****

Author Contribution

Ingo Schmidt – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Author declare no conflict of interest.

Copyright

© 2017 Ingo Schmidt. This article is distributed under the terms of Creative Commons Attribution License which permits unrestricted use, distribution and reproduction in any medium provided the original author(s) and original publisher are properly credited. Please see the copyright policy on the journal website for more information.

REFERENCES

1. Le Nen D, Hu W, Liot M, Moineau G, Gerard R. Gastrocnemius medial flaps. Interact Surg 2007;2(2):118–27.
2. Mathes SJ, Nahai F. Classification of the vascular anatomy of muscles: Experimental and clinical correlation. Plast Reconstr Surg 1981 Feb;67(2):177–87.
3. McCraw JB, Fishman JH, Sharzer LA. The versatile gastrocnemius myocutaneous flap. Plast Reconstr Surg 1978 Jul;62(1):15–23.
4. Neale HW, Stern PJ, Kreilein JG, Gregory RO, Webster KL. Complications of muscle-flap transposition for traumatic defects of the leg. Plast Reconstr Surg 1983 Oct;72(4):512–7.
5. Haroon-Ur-Rashid, Hafeez K, Abbas K. Use of distally based random flap in the management of soft tissue defects in upper two thirds of leg. J Pak Med Assoc 2014 Dec;64(12 Suppl 2):S15–8.

ABOUT THE AUTHOR

Article citation: Schmidt I. The gastrocnemius muscle flap for coverage of soft tissue defect of the proximal third of lower leg. Int J Case Rep Images 2017;8(2):168–170.

Ingo Schmidt is a surgeon in the Department of Traumatology SRH Poliklinik, Waldklinikum Gera GmbH, Germany. From 1983 to 1989, he studied human medicine at the Friedrich-Schiller-University in Jena (Germany). From 1990 to 1999, Dr. Schmidt graduated his training for general surgery, traumatology, orthopaedics, and hand surgery at the University hospital in Jena. In 1994, he successfully defended his scientific work to gain the title as a medical doctor. He has published more than 20 scientific articles. His areas of interest include hip replacement, coverage of soft tissue defects, and hand surgery with special focus on total wrist replacement and arthroplasties of all other joints of the hand.

Access full text article on other devices

Access PDF of article on other devices