EDITORIAL BOARD
International Journal of Case Reports and Images (IJCRI)

Achuta Kumar Guddati  USA
Aditya Gupta  USA
Adria Arboix  Spain
Adriana Handra-Luca  France
Afra Hadjizadeh  Iran
Ahmed El Said Lasheen  Egypt
Akbar Dorgalaleh  Iran
Ali Soltani  USA
Alok Kumar  India
Altacilio Aparecido Nunes  Brazil
Amin F Majdalawieh  United Arab Emirates
Amit Sengupta  India
Antonio La Cava  USA
Asher Bashiri  Israel
Athanasios Tsakris  Greece
Awad Alawad  Sudan
Banavar Ravi Spoorthi  India
Behzad Foroutan  Iran
Benson Chellaikan  USA
Selvanesan
Bhagya Lakshmi Atla  India
Bhavinder Arora  India
Bhoopendra Singh  India
Chandrakeshwar  India
Rajasekharan
Christopher CK Ho  malasiya
Claudio Feliciani  Italy
Daniela Cabibi  Italy
Deepa Rastogi  USA
Deepak Sharma  USA
Ekamol Tantisattamo  USA
Emre Karasahin  Turkey
Federico Bizzarri  Italy
Gavin A Falk  USA
George Konstantinos Paraskevas  Greece
Gerardo Gómez-Moreno  Spain
Germana Casaccia  Italy
Gil Atzmon  USA
Giovanni Leuzzi  Italy
Giovanni Tuccari  Italy
Gokulakrkrishna Subhas  USA
Hajimi Orita  Japan
Ho-Sheng Lin  USA
Hong Seng Gan  Malaysia
Hua Zhong  USA
Huseyin Avni Balcioglu  Turkey
Imtiaz Wani  India
James Cheng-Yi Lin  Taiwan
Jonathan D. Solomon  USA
Kyuzi Kamoi  Japan
Luca Bertolaccini  Italy
Makoto Adachi  Japan
Mehmet Inal  Turkey
Mehmet Uludag  Turkey
Mohamed Radhi  USA
Mohannad Al-Qudah  Jordan
Mohit Kumar Patralekh  India

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)
EDITORIAL BOARD
International Journal of Case Reports and Images (IJCRI)

Montish Singla USA
Naila Khalil USA
Natunya Semiletova USA
Oner Dikensoy Turkey
Ozlem Guneysel Turkey
P.S. Srikumar Malaysia
Parijat Saurav Joy USA
Paolo Cardelli Italy
Paul Rea United Kingdom
Pengcheng Luo China
Piaray Lal Kariholu India
Piraye Kervancioglu Turkey
Prabin Sharma USA
Radhika Muzumdar USA
Rajan Arora USA
Rajinder PS Bajwa USA
Rajesh Pareta USA
Ranjan Agrawal India
Ranji Cui China
Ranjit Nair USA
Ricardo Correa USA
Ricardo S. Macarenco Brazil
Rohini Karunakaran Malaysia
Roy Rillera Marzo Malaysia
Sangeeta Singh India
Sanju George United Kingdom
Santhosh USA
Gheevarghese John USA
Saurabh Khakharia USA
Sergio Gabriel Israel
Susmallian USA
Shashideep Singhal USA
Shekhar Gogna India
Shervin Assari USA
Shilpa Jain USA
Shivilal Vishnoi India
Siddharth Mathur USA
Sinirath Sirivisoot USA
Slobodan Marinkovic Slovenia
Sofya Asfaw USA
Stefan Hagmann USA
Stefano Romagnoli Italy
Tapas Saha USA
Teguh Haryo Sasonko Malaysia
Tun Hing Lui China
Tushar Chandra USA
Varun Menon. P India
Yulin Li China
Yupeng Chen USA

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)
Contents

Editorial

358 Alloimmunization against Rh and Kell blood groups antigens is the main obstacle for blood transfusion in transfusion dependent thalassemia patients in Iran
Akbar Dorgalaleh, Mohammad Saeed Gholami, Mohammad Shokuhayan, Mohsen Valikhani, Esmaei Sameei Moghaddam, Majid Naderi

Case Series

364 Impact of collagen hydrolysate in middle-aged athletes with knee and ankle osteochondral lesions: A case series
Fábio Krebs Gonçalves

370 Abdominal menstruation: A dilemma for the gynecologist
Seema Singhal, Sunesh Kumar, Yamini Kansal, Deepika Gupta, Mohit Joshi

Case Reports

376 Pulmonary embolism and atrial fibrillation: A complicated relationship
Saroj Lohani, Niranjan Tachamo, Bidhya Timilsina, Salik Nazir

380 An indirect sinus floor elevation by using piezoelectric surgery with platelet-rich fibrin for sinus augmentation: A short surgical practice
Ali H. Neamat, Shakhawan M. Ali, Saman W. Boskani, Payman Kh. Mahmud

385 A rare case of primary squamous cell carcinoma of the stomach
Xu Chen, Chengxin Luo, Hu Zhang

390 Live Ascaris in anterior chamber causing hypertensive uveitis
Anadi Khatri, Bal Kumar Khatri

394 Acute colonic pseudoobstruction (Ogilvie’s syndrome) as a postoperative complication: A case report and literature review
Alaa Sedik, Mufid Maaly, Salwa ElHoushy

397 A misdiagnosis of benign abdominal mass as a non-Hodgkin lymphoma
Luma Haj Kassem, Ahmad Ghazal, Khawa Mayoh, Najat Mehio Sailam

402 Spontaneous splenic rupture without trauma: A case report
Mürşit Dincer, Ahmet Kocakuşak, Gamze Çitlak, Ekrem Ferlengez, Muzaffer Akinci

405 A diagnostic dilemma: Sclerosing encapsulated peritonitis
Mürşit Dincer, Gamze Çitlak, Zehra Zeynep Keklikkiran, Ahmet Kocakuşak, Muzaffer Akinci

408 Congenital epulis: A rare case report
Jaya Naidu, Shreya Banerjee, Sapna Jyoti, Pavanalakshmi GP, Kirthana Satish

412 Complex esophageal reconstruction after esophagogastronomy with non-supercharged right colon interposition for the treatment of lye ingestion
Paige Finkelstein, Omar Picado, Elizabeth Paulus, Janeth Ng, Gabriel Ruiz, Danny Yákoub

417 Reversible stomatocytosis
Akanksha Agrawal, Deepanshu Jain, Mitchell Goldstein

420 Splenic artery aneurysm: Interesting images
Sayf Altabaqchali, Mohanad Hasan, Ahmed Altabaqchali

423 Kienböck’s disease mimicking gouty monoarthritis of the wrist
Ingo Schmidt

Clinical Images

Letters to the Editor
Alloimmunization against Rh and Kell blood group antigens is the main obstacle for blood transfusion in transfusion dependent thalassemia patients in Iran

Akbar Dorgalaleh, Mohammad Saeed Gholami, Mohammad Shokuhian, Mohsen Valikhani, Esmaei Saneei Moghaddam, Majid Naderi

ALLOIMMUNIZATION IN THALASSEMIA MAJOR

Thalassemia is the most common inherited single-gene disorder, causing by decrease or absence of α-globin or β-globin chain production. The disorder commonly inherited in autosomal recessive manner and is more common in areas with high rate of consanguinity [1–3]. Thalassemia belt is an extensive area which extend from Mediterranean east through Middle-East and India to Southeast Asia and south through Africa. Estimated incidence of thalassemia in this area is varies from 1–20% depend on area. Iran as a Middle-East country with high rate of consanguineous marriage has a considerable number of patients with β-thalassemia major [4–6]. The precise incidence of disorder is not clear in Iran but it was estimated that there are between two and three million beta thalassemia carriers and about 20,000 patients with beta thalassemia major. The main therapeutic choice in these patients is packed red blood cell (pRBC) transfusion. Continuous blood transfusion imposed a number of transfusion related complications, most importantly iron overload and related complications as well as alloimmunization against transfused red blood cell antigens [6–10]. The reported rate of alloimmunization among transfused dependent patients with thalassemia varies between 4–50% and has a lower incidence in homologues populations. Some of these alloantibodies are important and even cause severe life-threatening transfusion related hemolytic reactions while others are clinically insignificant. Both of alloantibodies and autoantibodies may decrease survival of transfused pRBC and increase transfusion rate. Such patients may require immunosuppressive drugs, splenectomy as well as other alternative treatments. Therefore, alloimmunization and autoimmunization can significantly affect patients’ quality of life and overall survival [11–13].

ALLOIMMUNIZATION IN THALASSEMIA MAJOR IN IRAN

A considerable number of studies were performed in patients with β-thalassemia major in different areas of Iran. The most common used method for detection of alloimmunization, was conventional tube technique (~80%), while gel method was used in minority (~20%) [5–8]. In the majority of studies, in addition to alloantibodies, autoantibody (8 out of 13 studies) (61.5%) also were detected [1, 3, 4, 12]. Among these studies, the rate of autoimmunization ranges from 1–~19% [4, 9]. The rate of alloimmunization varied between ~3–76%. The lower incidence of alloimmunization was reported in Tehran province, while the highest incidence was observed in Isfahan province [6, 9]. Among alloantibodies the majorities are against Rh and Kell blood group systems. The prevalence of alloantibody against, Rh system ranged from 7.5–100% and this prevalence for Kell system varied from 14–60% [3]. Among these studies on Iranian patients, the rate of splenectomy was reported from ~8–100%. The rate of alloimmunization against these totally splenectomies patients was ~4% (Table 1).

In Rh blood group system, most of antibodies directed against, E, C and c antigens respectively, while in Kell blood group system the majority were directed against K antigen (Table 2).
### Table 1: Prevalence of splenectomy and hemolytic reaction among Iranian patients with thalassemia major

<table>
<thead>
<tr>
<th>Study</th>
<th>Splenectomy</th>
<th>Without spleectomy</th>
<th>Hemolytic reaction</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>28 (7.3%)</td>
<td>375 (92.7%)</td>
<td>Without hemolytic reaction</td>
</tr>
<tr>
<td>2</td>
<td>18 (25.7%)</td>
<td>52 (74.3%)</td>
<td>-</td>
</tr>
<tr>
<td>3</td>
<td>346 (41.4%)</td>
<td>489 (58.6%)</td>
<td>21 (2.5%)</td>
</tr>
<tr>
<td>4</td>
<td>222 (50.3%)</td>
<td>219 (49.6%)</td>
<td>7 (1.6%)</td>
</tr>
<tr>
<td>5</td>
<td>-</td>
<td>-</td>
<td>-</td>
</tr>
<tr>
<td>6</td>
<td>9 (17.3%)</td>
<td>43 (82.7%)</td>
<td>-</td>
</tr>
<tr>
<td>7</td>
<td>-</td>
<td>-</td>
<td>-</td>
</tr>
<tr>
<td>8</td>
<td>28 (21%)</td>
<td>105 (78.9%)</td>
<td>-</td>
</tr>
<tr>
<td>9</td>
<td>80 (100%)</td>
<td>-</td>
<td>11 (13.75%)</td>
</tr>
<tr>
<td>10</td>
<td>7</td>
<td>-</td>
<td>-</td>
</tr>
<tr>
<td>11</td>
<td>10 (8.3%)</td>
<td>111 (91.7%)</td>
<td>-</td>
</tr>
<tr>
<td>12</td>
<td>17 (34.7%)</td>
<td>32 (65.3%)</td>
<td>-</td>
</tr>
<tr>
<td>13</td>
<td>203 (28.5%)</td>
<td>508 (71.5%)</td>
<td>-</td>
</tr>
<tr>
<td>Total</td>
<td>961</td>
<td>1934</td>
<td>39 (2.8%)</td>
</tr>
</tbody>
</table>

*All of the patients had their spleen removed prior to the time of antibody formation.*

### Table 2: Prevalence of alloimmunization and autoimmunization among Iranian patients with β-thalassemia major

<table>
<thead>
<tr>
<th>Study</th>
<th>Number of patients</th>
<th>Gender</th>
<th>Mean age</th>
<th>Province</th>
<th>Method</th>
<th>Allo-antibody</th>
<th>Majority of allo-antibodies</th>
<th>Number of antibody</th>
<th>Auto-antibody</th>
<th>Reference</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>385</td>
<td>Male: 221 Female: 164</td>
<td>13.8 y</td>
<td>Sistan and Baluchestan</td>
<td>Conventional</td>
<td>69 (17.9%)</td>
<td>Rh: 49.1% Kell: 14% Leu: 10.5%</td>
<td>Single</td>
<td>21 (5.5%)</td>
<td>1</td>
</tr>
<tr>
<td>2</td>
<td>70</td>
<td>Male: 31 Female: 39</td>
<td>16 y</td>
<td>Khuzestan</td>
<td>Conventional</td>
<td>6 (8.6%)</td>
<td>Rh: 50% Kell: 50%</td>
<td>Single</td>
<td>-</td>
<td>2</td>
</tr>
<tr>
<td>3</td>
<td>835 Adult (548) Pediatric (287)</td>
<td>Male: 416 Female: 419</td>
<td>Adult (24.5 y) Pediatric (10 y)</td>
<td>Tehran</td>
<td>Gel</td>
<td>100 (11.9%)</td>
<td>Rh: 45% Kell: 34% Colton: 2%</td>
<td>Single</td>
<td>72%</td>
<td>1 (1%) 3</td>
</tr>
<tr>
<td>4</td>
<td>441</td>
<td>Male: 234 Female: 207</td>
<td>22 y</td>
<td>Tehran + Qazvin</td>
<td>Gel</td>
<td>50 (11.3%)</td>
<td>Rh: 42% Kell: 28%</td>
<td>Single</td>
<td>74% Double: 16% UD: 8%</td>
<td>1 (2%)</td>
</tr>
<tr>
<td>5</td>
<td>458</td>
<td>Male: 221 Female: 237</td>
<td>16.96 y</td>
<td>Tehran</td>
<td>Gel</td>
<td>49 (11.8%)</td>
<td>Kell: 35% Rh: 47%</td>
<td>Single</td>
<td>71.5% Double: 14.3% UD: 14.2%</td>
<td>1 (2%)</td>
</tr>
<tr>
<td>6</td>
<td>52</td>
<td>Male: 36 Female: 16</td>
<td>18.2 y</td>
<td>Isfahan</td>
<td>Conventional</td>
<td>40 (76%)</td>
<td>Kell: 27.5% MNSs: 20% Rh: 7.5%</td>
<td>Single</td>
<td>67.5% Multiple: 27.5% UD: 5%</td>
<td>-</td>
</tr>
<tr>
<td>7</td>
<td>218</td>
<td>Male: 100 Female: 118</td>
<td>22.5 y</td>
<td>Mazandaran</td>
<td>Conventional</td>
<td>88 (40.4%)</td>
<td>Rh: 75% Le: 64%</td>
<td>Single</td>
<td>47%</td>
<td>-</td>
</tr>
<tr>
<td>8</td>
<td>133</td>
<td>Male: 66 Female: 67</td>
<td>17.5 y</td>
<td>Khuzestan</td>
<td>Conventional</td>
<td>25 (18.7%)</td>
<td>Rh: 55% Kell: 33%</td>
<td>Single</td>
<td>72% Double: 20% Triple: 8%</td>
<td>17 (12.7%)</td>
</tr>
<tr>
<td>9</td>
<td>80</td>
<td>Male: 37 Female: 43</td>
<td>8.35 y</td>
<td>Tehran</td>
<td>Conventional</td>
<td>3 (3.7%)</td>
<td>Rh: 67% Kell: 33%</td>
<td>Single</td>
<td>15 (18.8%)</td>
<td>9</td>
</tr>
<tr>
<td>Ref.</td>
<td>Country</td>
<td>Year(s)</td>
<td>Sample Size</td>
<td>Age (Mean)</td>
<td>Gender</td>
<td>Gel</td>
<td>Conventional</td>
<td>Rh</td>
<td>Single</td>
<td>Double</td>
</tr>
<tr>
<td>------</td>
<td>------------------------</td>
<td>---------</td>
<td>-------------</td>
<td>------------</td>
<td>--------</td>
<td>-----</td>
<td>--------------</td>
<td>----</td>
<td>--------</td>
<td>--------</td>
</tr>
<tr>
<td>10</td>
<td>313</td>
<td>Male: 187</td>
<td>14.46 y</td>
<td>Male: 187</td>
<td>14.46 y</td>
<td></td>
<td>Northeast of Iran (Mashhad)</td>
<td>Conventional</td>
<td>9 (2.87%)</td>
<td>Rh: 100%</td>
</tr>
<tr>
<td>11</td>
<td>121</td>
<td>Male: 55</td>
<td>13.0 y</td>
<td>Male: 55</td>
<td>13.0 y</td>
<td></td>
<td>Tehran</td>
<td>Conventional</td>
<td>9 (7.4%)</td>
<td>Rh: 22.2%</td>
</tr>
<tr>
<td>12</td>
<td>49</td>
<td>Male: 25</td>
<td>18.59 y</td>
<td>Female: 66</td>
<td></td>
<td></td>
<td>Zanjan</td>
<td>Conventional</td>
<td>8 (16.32%)</td>
<td>Rh: 40%</td>
</tr>
<tr>
<td>13</td>
<td>711</td>
<td>Male: 366</td>
<td>14.4 y</td>
<td>Female: 345</td>
<td></td>
<td></td>
<td>Shiraz</td>
<td>Conventional</td>
<td>38 (5.3%)</td>
<td>Rh: 36.3%</td>
</tr>
</tbody>
</table>

Table 3: Prevalence of alloimmunization and autoimmunization among different countries

<table>
<thead>
<tr>
<th>Study</th>
<th>Country</th>
<th>Year(s)</th>
<th>Sample Size</th>
<th>Age (Mean)</th>
<th>Gender</th>
<th>Gel</th>
<th>Conventional</th>
<th>Rh</th>
<th>Single</th>
<th>Double</th>
<th>Triple</th>
<th>Percentage</th>
</tr>
</thead>
<tbody>
<tr>
<td>Ahmed et al.</td>
<td>Egypt</td>
<td>2010</td>
<td>501</td>
<td>10.84 y</td>
<td></td>
<td></td>
<td>Gel and Conventional</td>
<td>57</td>
<td>Rh: 15%</td>
<td>K: 35%</td>
<td>E: 29.9%</td>
<td>Single: 145 (28.9%)</td>
</tr>
<tr>
<td>Haslina et al.</td>
<td>Malaysia</td>
<td>2004</td>
<td>63</td>
<td>Adult (24.5 y)</td>
<td></td>
<td></td>
<td>Conventional</td>
<td>-</td>
<td>-</td>
<td>Single</td>
<td>1 (1.6%)</td>
<td>15</td>
</tr>
<tr>
<td>Pahuja et al.</td>
<td>India</td>
<td>2008-2009</td>
<td>211</td>
<td>Adult &gt;5 y (80.6 y)</td>
<td></td>
<td></td>
<td>Conventional</td>
<td>8 (3.79%)</td>
<td>Rh: 25%</td>
<td>K: 25%</td>
<td>Single:75%</td>
<td>Double: 25%</td>
</tr>
<tr>
<td>Al-Mousawi et al.</td>
<td>Iraq</td>
<td>2014</td>
<td>401</td>
<td>10.0 y</td>
<td></td>
<td></td>
<td>Conventional</td>
<td>18 (4.5%)</td>
<td>Rh: 6%</td>
<td>K: 3%</td>
<td>C: 3%</td>
<td>Single: 88.9%</td>
</tr>
<tr>
<td>S. Jansuwan et al.</td>
<td>Thailand</td>
<td>2013</td>
<td>143</td>
<td>16.0 y</td>
<td></td>
<td></td>
<td>Gel and Conventional</td>
<td>24</td>
<td>Rh: 16.7%</td>
<td>E: 16.7%</td>
<td>C: 16.7%</td>
<td>Single: 68%</td>
</tr>
<tr>
<td>Köçyigit C, et al.</td>
<td>Turkey</td>
<td>2011-2012</td>
<td>139</td>
<td>Adult &gt; 2 (37 y) Pediatric &lt; 2 (102)</td>
<td></td>
<td></td>
<td>Gel and Conventional</td>
<td>9 (6.4%)</td>
<td>Rh: 27%</td>
<td>K: 27%</td>
<td>D: 18%</td>
<td>JKa:18%</td>
</tr>
<tr>
<td>Dogra, et al.</td>
<td>Jammu region</td>
<td>2009-2010</td>
<td>70</td>
<td>9.27 y</td>
<td></td>
<td></td>
<td>Conventional</td>
<td>6 (7.5%)</td>
<td>Rh: 50%</td>
<td>K: 33.3</td>
<td>D: 16.7</td>
<td>Single: 1 (1.42%)</td>
</tr>
<tr>
<td>AMEEN et al.</td>
<td>Kuwait</td>
<td>2002</td>
<td>190</td>
<td>12.7 y</td>
<td></td>
<td></td>
<td>Conventional</td>
<td>142</td>
<td>Rh: 74.7%</td>
<td>K: 41 (72%)</td>
<td>E: 26 (45.6%)</td>
<td>D: 12 (21.1%)</td>
</tr>
<tr>
<td>Hassan et al.</td>
<td>Pakistan</td>
<td>2003</td>
<td>75</td>
<td>6.5 y</td>
<td></td>
<td></td>
<td>Conventional</td>
<td>17</td>
<td>Rh: 22.7%</td>
<td>Kpa: 4</td>
<td>e: 3 (17.6%)</td>
<td>E: 2 (11.8%)</td>
</tr>
<tr>
<td>L.-Y. Wang et al.</td>
<td>Taiwan</td>
<td>2005</td>
<td>30</td>
<td>20 y</td>
<td></td>
<td></td>
<td>Conventional</td>
<td>11</td>
<td>Rh: 63.6%</td>
<td>C: 2 (18.2%)</td>
<td>Single: 72.8%</td>
<td>Double: 27.2%</td>
</tr>
<tr>
<td>N. Guirat-Dhouib et al.</td>
<td>Tunisia</td>
<td>2011</td>
<td>130</td>
<td>9.9 y</td>
<td></td>
<td></td>
<td>Conventional</td>
<td>10</td>
<td>Rh: 30%</td>
<td>C: 30%</td>
<td>Single: 80%</td>
<td>Double: 20%</td>
</tr>
<tr>
<td>Muhammad USMAN et al.</td>
<td>Pakistan</td>
<td>2011</td>
<td>800</td>
<td>11.5 y</td>
<td></td>
<td></td>
<td>Conventional</td>
<td>30</td>
<td>Rh: 7 (23.3%)</td>
<td>E: 6 (20%)</td>
<td>C: 5 (16.7%)</td>
<td>Single</td>
</tr>
<tr>
<td>Th. Spanosa et al.</td>
<td>Greece</td>
<td>2013</td>
<td>1200</td>
<td>11.5 y</td>
<td></td>
<td></td>
<td>Conventional</td>
<td>220</td>
<td>Rh: 22.6%</td>
<td>K: 134</td>
<td>C: 66 (14%)</td>
<td>Single: 51.8%</td>
</tr>
</tbody>
</table>

*calculated
COMPARISON BETWEEN ALLOIMMUNIZATION IN IRAN WITH OTHER COUNTRIES

Similar to Iran the most common antibodies against transfused red blood cells were anti-Rh and anti-Kell antibodies in other countries [14–22]. The rate of alloimmunization varies between these countries from ~4% in India and Pakistan to ~75% in Kuwait [16, 21, 22]. Similar to Iran, conventional tube method is the most commonly used method for antibody detection and identification in other countries [23–26]. Although as low as 0.7% of autoantibody was reported in other countries, most studies reported a significantly higher incidence of autoimmunization in other countries [18, 22–25]. In Kell blood group system, the most commonly alloantibodies directed against K antigen [14, 18–20]. In Rh blood group system, majority of antibody directed against D, E and C antigens (Table 3). With regards to these studies it seems that alloimmunization and even autoimmunization are a major concern in transfusion dependent thalassemia patients [5, 7, 22]. In Iranian patients with β-thalassemia major, transfusion related reaction was reported with a prevalence of about 15% in some studies [15, 20]. Sometime, in patients with alloimmunization is significant and can have life-threatening consequences. In emergency situations, appropriate blood selection is really difficult and required sophisticated laboratory investigations that only can be performed in specialized laboratories. In addition to this condition, clinically significant alloantibodies and autoantibodies can affect quality of life of these patients and affect overall survival of patients with β-thalassemia major. To prevent such conditions, its appropriate to use more suitable preventable strategies such as phenotyping of patients prior to beginning blood transfusion and used of relatively complete matched pRBC. Another way to prevent, is application of direct donation instead of random pRBC transfusion that increase the rate of alloimmunization and related consequences.

Keywords: Alloimmunization, Blood transfusion, Kell, Rh, β-thalassemia major

How to cite this article


Article ID: Z01201706ED10008AD

Acknowledgements

We appreciate all patients with thalassemia that taking part in our researches and improved our knowledge about different aspects of thalassemia.

Author Contributions

Akbar Dorgalaleh – 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
Mohammad Saeed Gholami – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published
Mohammad Shokuhiyan – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published
Mohsen Valikhani – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published
Esmaei Saneei Moghaddam – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published
Majid Naderi – 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 Akbar Dorgalaleh 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

of erythrocyte alloantibodies in patients with thalassemia major referred to Ahvaz Shafa hospital. Feyz Journals of Kashan University of Medical Sciences 2013;17(2).


6. Rahgozar S, Moafi A, Yavari F, Hourfar H. Alloantibody detection in major beta thalassemia patients transfused within less-than-20-day intervals. 2005. [Available at: h t t p : / / b l o o d j o u r n a l . i r / b r o w s e . p h p ? a _ i d = 1 0 & s i d = 1 & s l c _ l a n g = e n]


Impact of collagen hydrolysate in middle-aged athletes with knee and ankle osteochondral lesions: A case series

Fábio Krebs Gonçalves

ABSTRACT

Introduction: Osteochondral lesions (OCL) are a significant issue among professional and amateur athletes in several sports modalities. Lack of treatment or inadequate therapy may aggravate the symptoms and jeopardize the chance of a complete functional recovery. Multidisciplinary treatment is the mainstay of OCL, including physiotherapy, physical rehabilitation with aerobic and anaerobic exercises and lately, the use of nutritional supplementation with collagen hydrolysate (CH). Case Series: We report three cases of OCL in athletes, which were diagnosed and followed with magnetic resonance imaging (MRI) scan after multidisciplinary treatment including nutritional supplementation with Fortigel® a specific collagen hydrolysate that contains bioactive collagen peptides. A literature review on the role of collagen hydrolysate and its nutritional aspect in supporting the regeneration of articular cartilage is also presented. Conclusion: MRI images of patients assessed in this study depict the recovery of the articular surfaces after the treatment. Also, studies in athletes presenting joint pain but otherwise not diagnosed with osteoarthritis or other osteoarticular diseases have demonstrated the beneficial effects of CH intake.

Keywords: Athletes, Collagen hydrolysate, Nutritional supplementation, Osteochondral lesions

INTRODUCTION

Osteochondral lesions (OCL) are a significant issue among professional and amateur athletes in several sports modalities. Lack of treatment or inadequate therapy may worsen the symptoms and jeopardize the chance of a complete functional recovery. Osteochondral lesions can be induced by several factors, such as ischemia, repetitive microtrauma and acute trauma. These lesions may affect any joint, but they are most commonly seen in the weight-bearing area of the lateral femoral condyle, the inferomedial pole of the patella and the talar dome [1].

Multidisciplinary treatment is the mainstay of OCL, despite being surgical or conservative. It involves physiotherapy, physical rehabilitation with aerobic and anaerobic exercises and lately, the use of nutritional supplementation with collagen hydrolysate.
Collagen is the main insoluble fibrous protein in the extracellular matrix and is commonly found in the skin, tendons, cartilage, bone and cornea. This complex protein consists of repeated amino-acid sequences (especially glycine, proline and hydroxyproline) packed together and forming fibrils [2, 3]. There are over 20 known different types of collagen, but types I, II and III comprise about 90% of what is found in the human body.

Type II collagen is the principal protein found in cartilage, and its small fibrils disposed in a random fashion on the proteoglycan matrix confer traits of strength and compressibility that allows absorption of impacts [2, 3]. There are four distinct zones (superficial, transitional, deep and calcified) found in the articular cartilage, which are defined by the morphological differences in density, cellular disposition (chondrocytes), nature, content and distribution of proteoglycans and the special organization of the collagen fibrils. The percentage of collagen found in the superficial layers of the articular cartilage reaches 80%, while the deeper layers have around 65% of that protein in their structure [4].

Sanctioned as a safe food ingredient by regulatory agencies, collagen hydrolysate is obtained by enzymatic hydrolysis of animal collagenous tissues (mainly skin) and has an identical amino acid composition to type II collagen with high levels of glycine and proline, which are a prerequisite for a healthy cartilage [5–7].

We aim to report three cases of OCL in athletes, which were diagnosed and followed with magnetic resonance imaging (MRI) scan after multidisciplinary treatment that included nutritional supplementation with Fortigel®, a specific collagen hydrolysate, developed by Gelita Company (Heidelberg, Germany), which contains bioactive collagen peptides (BCP). A literature review on the role of collagen hydrolysate in supporting the benefits from a specific nutritional approach focused on cartilage is also presented.

**CASE SERIES**

**Case 1**

A 42-year-old male football player who plays seven-a-side indoor soccer twice a week and jogs three times a week. He sought medical attention due to pain in the right knee and difficulty in walking following a sprain whilst playing indoor soccer. Patient’s body measurements were as follows: 1.75 m, 70 kg and 22.86 kg/m² of body mass index (BMI). Physical examination showed neutral alignment of lower limbs, pain on the medial joint line and medial femoral condyle during palpation. Limitation of final knee flexion and pain during the McMurray test for the medial meniscus were also detected. All ligament tests were normal, with motion ranges of 0–140 degrees on left knee and 0–115° on the right one. An MRI of the right knee performed a month after his lesion showed deep ulceration on the medial femoral condyle’s weight-bearing cartilage, area with intense reactional bone edema in the spongy section of the femoral condyle, slight strain on the medial collateral ligament, and edema on adjacent soft tissues (Figure 1A).

Final diagnosis was right knee pain due to osteochondral ulceration on the medial femoral condyle. Treatment included physiotherapy and the daily oral intake of 10 grams of collagen hydrolysate Fortigel® dissolved in a glass of water. The patient returned six months later, having undergone 20 sessions of physiotherapy and reporting significant pain reduction. He was counseled to maintain the physiotherapy program, continue the intake of collagen hydrolysate and initiate physical activities to ensure muscle strengthening. In his following consultation, after nine months, the patient was asymptomatic. Physical examination showed a slight weight loss (67 kg), absence of pain, edema or effusion on the joints, and normal range of mobility (0–140°) in both knees. A follow-up MRI scan performed 17 months after his lesion (Figure 1B) showed small, superficial chondral erosions on the medial femoral condyle’s weight-bearing area with significant reduction of the ulcerations formerly detected and without reactional subchondral osteitis.

The patient was advised to maintain the ingestion of the collagen hydrolysate Fortigel® and continue with the physical training in the local academy, once-a-week soccer and treadmill running three times a week. With the subsequent functional recovery achieved, the patient was cleared to fully return to his sports practice while maintaining the therapeutic measures prescribed.

**Case 2**

A 43-year-old salesman, who plays indoor soccer and runs twice a week. He suffered a sprained ankle during a soccer game. The attending physician ordered an X-ray
that showed soft tissue edema on the periarticular area with no evidence of fractures or other bone lesions. The patient had the left ankle immobilized for 8 days and was prescribed non-steroidal anti-inflammatory drugs (NSAID). He came to a consultation almost a month after the lesion, reporting persisting ankle pains and difficulty to stand-up. Physical examination showed neutral alignment of lower limbs, edema and pain to the touch of the anterolateral surface of the left ankle. Mobility tests showed limitations on the left ankle with apparent ligament stability. Range of motion for plantar flexion was 50° on the right ankle and 35° on the left and for dorsiflexion 12° and 4° respectively. Patient’s body measurements were 1.81 m, 80 kg and BMI of 24.42 kg/m². He was diagnosed with a left ankle sprain with ligament lesion and prescribed the use of removable cast walker boots, as well as 20 sessions of physiotherapy. After concluding the treatment, the patient returned to his usual sports practice (indoor soccer and running). The patient was treated with removable immobilization and physical therapy and in a consultation, six years after his lesion, he informed recurring pains on the left ankle after exercising, with subsequent limitation of mobility. Physical examination showed pain during palpation of the anterior surface of the left ankle on the region of the tibiotalar joint. Mobility ranges for plantar flexion were 50° on the right ankle and 30° on the left one and dorsiflexion ranges were 12° and 4°, respectively. The MRI scan ordered on this consultation (Figure 2A) showed a significant osteochondral lesion on the medial talar dome with a strain on the anterior talofibular ligament. The patient was then recommended the daily oral intake of 10 g collagen hydrolysate (Fortigel®), advised to avoid high-impact activities and referred to the physiotherapist for treatment. On his next consultation, after 16 months, the patient reported to be pain-free. Physical examination showed a 50° plantar flexion on the right ankle and 45° on the left one, while dorsiflexion ranges were 12° and 10° respectively. Palpation on the anterior region of the left ankle was normal. A new MRI scan, conducted on the same period, showed preservation of the articular surface, without progression of the OCL on the talar dome or signs of instability (Figure 2B). Due to his functional recovery, the patient was advised to maintain the oral intake of the collagen hydrolysate Fortigel®, continue with physical training in the academy, start jogging and progressively return to running.

Case 3

A 30-year-old male civil engineer, and a professional yachting (soling class) athlete. He sought medical attention reporting pain on the right knee following a game of padel tennis and sailing. He also complained of discomfort while walking and ascending/descending stairs. Physical examination showed neutral alignment of lower limbs, pain and crepitation during mobilization of the femoropatellar joint. Ligament and meniscal tests were normal. Flexion motion ranges were 130° on the right knee and 145° on the left one. Extension was complete bilaterally. Patient’s body measurements were 1.81 m, 80 kg and BMI of 24.69 kg/m². An MRI scan of the right knee performed on the same day of his medical consultation showed patellar chondropathy with chondral fissures in the apex and medial facet, reaching into the deeper layers (Figure 3A). The images demonstrated stress areas on
the subchondral structures of the facets and the patellar ridge. Final diagnosis was knee pain due to patellar chondropathy. Treatment included daily oral intake of 10 g collagen hydrolysate (Fortigel®), physiotherapy and interruption of high-impact activities (such as sailing). On his return one month later the patient reported improvement of symptoms and decrease in the right knee pain. This patient presented a clinical and radiological picture indicative of osteochondral pathology at the level of the left ankle and a sequela of sprain with joint trauma, which caused him to withdraw from soccer practice and from races. He presented limitation of the joint mobility of the affected ankle, which recovered after interdisciplinary treatment and use of bioactive collagen peptide. With the functional recovery obtained clinically and radiologically, he was able to return to the partial progressive practice of his sports activities, with orientation to maintain the prescribed therapeutic measures. He was counseled to maintain the intake of collagen hydrolysate and reinitiate his training program since he would participate on an International Yachting Event (Soling Class) in one month. On his next consultation, after two months, the patient reported being asymptomatic and pain-free and also that he had been classified among the first six finalists on the yachting event. Physical examination showed normal mobility range on both knees, with 145° flexion and complete bilateral extension. A new MRI scan, performed on the same period, revealed signs of patellar chondropathy on the apex and medial facet, reaching only the superficial layers (Figure 3B). The exam showed reduction of the OCL on the facets and patellar ridge, in comparison to the previous one. The patient was advised to continue the use of the collagen hydrolysate Fortigel®, maintain his anaerobic activities (such as weight lifting) and also the aerobic training. The clinical and radiologic functional recovery allowed the patient to keep his sports activities and he became champion of the local boat race (XXI Copa da Cidade de Porto Alegre) in March 2015.

DISCUSSION

The use of collagen hydrolysate as a nutraceutical or nutritional supplement has been studied on the last decades, not only for skin and hair diseases, but also in researches focusing on degenerative conditions of the musculoskeletal system, including nutritional aspects and in particular on joint cartilage health. Chondrocytes control the maintenance of the extracellular matrix and consequently the functional integrity of the articular cartilage. These cells are continuously placed in a dynamic mechanical environment, combining compression, variations in hydrostatic pressure and tension, which create a host of signals transmitted through the extracellular and pericellular matrixes and result in altered gene expressions and changing metabolism [8].

In vitro studies have shown that the addition of collagen hydrolysate in the culture medium of chondrocytes increased the secretion of type II collagen in a dose-dependent fashion, suggesting a likely feedback mechanism to regulate the collagen turnover in cartilage tissue [9]. Chondrocytes also responded to collagen hydrolysate addition with a significant increase in proteoglycan synthesis, aggrecan expression and 1.5 fold increment in type II collagen biosynthesis [10].

Previous analysis of radiolabeled collagen hydrolysate determined that the orally administered substance was absorbed from the intestine and accumulated preferentially on the cartilage tissue [11]. A randomized placebo control study on animal osteoarthritis (OA) model demonstrated that the oral ingestion of collagen hydrolysate over a Three-month period led to a significant decrease in cartilage tissue degeneration in the knee joints when compared with non-treated subjects [12].

Clinical studies also pointed to a beneficial effect of collagen hydrolysate in patients with osteoarthritis of the knee and hip, with significant reduction in the consumption of analgesics after two months of intake [13]. Recently, Kumar et al. published a randomized, double blind, placebo controlled trial evaluating the effects of daily ingestion of 5 g collagen hydrolysate (two types) on the control of knee osteoarthritis in 30 patients [14]. The endpoints were measured by the Western Ontario and McMaster Universities Osteoarthritis Index (WOMAC), used to assess pain, stiffness, and physical function in patients with hip and/or knee osteoarthritis; a visual analog scale (VAS) and quality-of-life (QoL) parameters. Patients in the collagen hydrolysate group presented a statistically significant improvement in all endpoints (p < 0.01) compared with those in the placebo arm.

Articular traumas are everyday occurrences among professional and recreational sports players. Several studies reported the prevalence of injuries among amateur athletes in various modalities. A prospective study with 231 amateur soccer players recorded 213 injuries (79% due to trauma) in 57% of the players during one season. Knee (20.2%) and ankle (19.2%) were often affected sites with strain and/or ligament lesions in 11.3% and 17.3%, respectively [15]. A retrospective analysis of 123 recreational athletes in several activities found that 74% reported injuries due to soccer practice (indoor, futsal, sand, and turf) [16]. Ankle sprains responded to almost half (49%) of the injuries in this study.

Sailing related injuries were surveyed, among others, by Fontoura and Oliveira, who reported 18.8% of foot and ankle lesions and 16.36% of knee traumas among 165 subjects and Moraes, who revealed that up to 32% of sailors in the Brazilian Olympic Team had knee pain [17, 18].

In fact, physical activity itself directly affects cartilage metabolism, as seen by the imbalance of proteoglycan degradation versus synthesis in the extracellular matrix after joint immobilization or by the direct damaging effects of overtraining [6].

We presented the case reports of three healthy athletes, two of them amateurs and one professional, engaged in
a wide range of sports practice (indoor soccer, running, padel tennis and sailing) that sought medical attention due to painful exercise-related osteoarticular lesions.

Studies in athletes presenting joint pain but otherwise not diagnosed with osteoarthritis or other osteoarticular diseases have demonstrated the beneficial effects of daily collagen hydrolysate oral intake.

An observational trial including one hundred athletes with exercise-related joint pain analyzed the effect of 10 g daily oral intake of collagen hydrolysate for 12 weeks on pain reduction [19]. The results showed that 78% of subjects reported reduction in pain levels on movement at the end of the observation period. Objective decrease in pain levels, measured on a 1–10 scale, was also confirmed by the attending physician.

A 24-week prospective, randomized, placebo-controlled, double blind study was conducted with 147 healthy college athletes complaining of arthralgia [20]. The students were randomized to receive either placebo or 10 g of collagen hydrolysate daily for the duration of the study. The level of pain on each joint at rest and during movement was recorded by students and physicians through a VAS in five visits. Data available for 97 subjects showed statistically significant changes in joint pain for the treatment group in six parameters: at rest recorded by the physician and by the subject (p = 0.025 and p = 0.039, respectively), when walking (p = 0.007), when standing (p = 0.011), when carrying objects (p = 0.014) and when lifting (p = 0.018).

All the cases reported in the present paper were followed with MRI scan, documenting the improvement or stabilization of the OCL after the physiotherapy treatment and collagen hydrolysate supplementation. Magnetic resonance imaging (MRI) scan has been increasingly used to confirm the diagnosis, define the stages and document the evolution of OCL [21, 22]. The reliability of the method to recognize cartilage changes after collagen hydrolysate (Fortigel®) intake was demonstrated by McAlindon [23]. A single center, prospective, randomized, placebo-controlled, double blind trial included 30 patients with mild knee osteoarthritis, whose cartilage changes were followed by two MRI techniques (delayed gadolinium enhanced magnetic resonance imaging of cartilage - dGEMRIC or T2 mapping) before and after collagen hydrolysate nutritional supplementation. Results of this pilot trial showed an increase in the dGEMRIC score in two regions of interest in patients on the collagen hydrolysate group, suggesting an increment in the proteoglycan content in the cartilage. The results in the placebo group presented a decrease of the score on these regions.

Magnetic resonance imaging scan of our patients clearly represent the recovery of the articular surfaces after the treatment. Of course, case reports or series do not possess the adequate design to confirm hypotheses, and large, controlled, randomized trials would be necessary to measure how much of such recovery is due to the intake of collagen hydrolysate. However, the collecting of data and the recording of evidence are fundamental to increase the body of knowledge on the treatment of OCL.

CONCLUSION

The beneficial effects of collagen hydrolysate intake was observed in the series of the three cases reported in our study and is also demonstrated in several studies in athletes presenting joint pain but otherwise not diagnosed with osteoarthritis or other osteoarticular diseases. As more information becomes available on the nutritional effects of collagen hydrolysate in osteoarticular recovery and maintenance, we will likely have a broader range of tools at hand to ensure the best possible treatment to injured athletes.

*********

Acknowledgements
We would like to thank Evidências, a Kantar Health company, for the editorial support in this paper.

Author Contribution
Fábio Krebs Gonçalves – 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 Fábio Krebs Gonçalves. 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


Abdominal menstruation: A dilemma for the gynecologist

Seema Singhal, Sunesh Kumar, Yamini Kansal, Deepika Gupta, Mohit Joshi

ABSTRACT

Introduction: Menstrual fistulae are rare. They have been reported after pelvic inflammatory disease, pelvic radiation therapy, trauma, pelvic surgery, endometriosis, tuberculosis, gossypiboma, Crohn’s disease, sepsis, migration of intrauterine contraceptive device and other pelvic pathologies. We report two rare cases of menstrual fistula. Case Series: Case 1: A 27-year-old nulliparous female presented with complaint of cyclical bleeding from the abdomen since three years. There was previous history of hypomenorrhea and cyclical abdominal pain since menarche. There is history of laparotomy five years back and laparoscopy four years back in view of pelvic mass. Soon after she began to have blood mixed discharge from scar site which coincided with her menstruation. She was diagnosed to have a vertical fusion defect with communicating left hypoplastic horn and non-communicating right horn on imaging. Laparotomy with excision of fistula and removal of right hematosalpinx was done. Case 2: 25-year-old female presented with history of lower segment cesarean section (LSCS) and burst abdomen, underwent laparotomy and loop ileostomy. Thereafter patient developed cyclical bleeding from scar site. Laparotomy with excision of fistulous tract and closure of uterine rent was done. Conclusion: Clinical suspicion and imaging help to clinch the diagnosis. There is no recommended treatment modality. Surgery is the mainstay of management. Complete excision of fistulous tract is mandatory for good long-term outcomes.

Keywords: Abdominal menstruation, Excision of fistulous tract, Menstrual fistula, Salpingocutaneous fistula, Uterocutaneous fistula

How to cite this article


Article ID: Z01201706CS10088SS

doi:10.5348/ijcri-201709-CS-10088

INTRODUCTION

Gynecologists are familiar with vesicovaginal, ureterovaginal or rectovaginal fistula. Menstrual fistula is defined as communication between the reproductive organs and skin, is a rare condition and only few case reports are published [1]. They are characterized by periodic discharge of blood from laparotomy scar site that coincides with normal menstruation [1]. Various types
of such fistula viz tubocutaneous, tuboenterocutaneous, uterocutaneous, cervicocutaneous and salpingo-ureteric fistula are described. This is a highly morbid condition as it usually affects young women and is a source of anxiety to patient as well as clinician because of persistence of symptoms despite treatment. It is essential to be aware of this entity because clinical suspicion is the key for successful management. Herein, we report two rare cases of menstrual fistula and discuss the management options of this condition.

CASE SERIES

Case 1

A 27-year-old nulliparous female with history of primary infertility presented with complaint of cyclical bleeding from the abdomen since three years. She had hypomenorrhea and cyclical abdominal pain since menarche. History of laparotomy five years back elsewhere because of mass in abdomen. However, intraoperatively no intervention could be done due to adhesions. After one year she underwent laparoscopy outside but again nothing was done except adhesiolysis. Few months after the second surgery, patient started having blood tinged purulent discharge from anterior abdominal wall scar site. Discharge coincided with menstruation. She had to use 1-2 pads per day at the abdominal site. On examination her built was average, breasts were Tanner stage IV, axillary hair were present and spine was normal. A midline vertical scar was present on her abdomen. Discharge was seen from a small pinkish area on the scar site along with mild induration (Figure 1). A tender cystic mass of 18 weeks size uterus, with restricted mobility was felt towards right.

On local examination pubic hair were present, Tanner stage V. On speculum examination cervix was normal but deviated to left. On bimanual examination same mass was felt from right fornix and small size uterus from left fornix. Ultrasonography revealed bicornuate uterus with collection within endometrial cavity of right horn likely non-communicating. Bulky right ovary with a tract noted from it to the scar site on anterior abdominal wall likely scar endometriosis with solitary left kidney. Magnetic resonance imaging scan revealed two uterine cavities with a single cervix and vagina and a left communicating horn and a right non-communicating uterine horn with hematometra and right hematosalpinx (Figure 2). Blood filled tract was extending from the hematosalpinx to the skin opening on the anterior abdominal wall (Figure 3). Patient and her family were counseled and prognosticated about poor fertility potential. She was given the option of excision of the horn. Patient wished to retain her menstrual and child bearing function. She was taken up for laparotomy. Hysterotomy along with drainage of collection, excision of right hematosalpinx and creation of neovagina was done. A small left uterine horn communicating with the cervix was identified. A new opening was created in the vagina followed by stent insertion and previously non-communicating horn was connected with the vagina. Histopathology showed ulcerated fistulous tract with inflammatory granulation tissue with chronic salpingitis and foci of endometriosis. Patient received oral hormonal pills for three months. Later, there was resumption of normal menstruation with good flow postoperatively along with relief of pain.

Figure 1: Fistulous opening at abdominal scar site.

Figure 2: Magnetic resonance imaging scan showing two uterine cavities with a single cervix and vagina with a left communicating horn and a right non-communicating uterine horn with hematometra and right hematosalpinx.
Case 2

A 25-year-old P1L1 presented with complaint of bleeding from scar site during periods since six months. She had an LSCS one year back at some hospital, followed by burst abdomen. She was then referred to us and underwent a laparotomy. Intraoperatively an injury was detected in ileum and a loop ileostomy was done. Patient recovered well but subsequently she started having monthly bleeding from LSCS scar site for which she was using 1–2 pads per day. On per abdomen examination midline puckered scar with a small central defect was seen. On speculum examination cervix was not visualized and vaginal examination revealed six weeks size uterus with restricted mobility. Ultrasound showed bulky uterus with multiple fibroids, largest measuring 35 mm, bilateral ovaries were normal with ill-defined tissue (1 cm) at scar site anterior to uterine fundus. Contrast enhanced magnetic resonance imaging (MRI) showed divagination of recti with mid line defect. There was defect in the lower uterine segment communicating with anterior abdominal wall with endometrial fluid extending up to the anterior abdominal wall (Figure 5). There were two foci of T2 hyperintensity at superior and inferior margins of the mid line abdominal wall scar respectively. Retroverted distorted uterus with multiple uterine fibroids with adherent small bowel loop in right adnexal region. Patient was taken up for laparotomy. Intraoperatively bladder was adherent to previous cesarean scar on uterus, sinus tract extending from previous cesarean scar site to peritoneum, to rectus sheath and to subcutaneous tissue (Figure 6). Adhesiolysis (Figure 7) with excision of...
fistulous tract (Figure 8) was done. A probe was inserted to confirm communication with uterine cavity (Figure 9) and closure of uterine defect was done (Figure 10) along with ileostomy closure. Postoperative period was uneventful.

Both patients are under follow-up and are keeping well with no recurrence of symptoms.

**DISCUSSION**

Menstrual fistula have been reported after pelvic surgery especially after incomplete removal of inflammatory tissue, pelvic inflammatory disease, trauma, endometriosis, tuberculosis, gossypiboma, pelvic radiation therapy, Crohn’s disease and other pelvic pathologies [2]. Most tubal fistulas arise as a result of inflammation of the fallopian tube, bowel or as a complication of gynecological surgery (cesarean section, salpingectomy or myomectomy) [2]. In most cases infection is an additional complicating factor. Salpingocutaneous fistula has been described in case of pelvic abscess as a result of induced septic abortion [3]. Yadav et al. described the utero-cutaneous fistula as a complication of laparotomy performed due to paraovarian cyst and intraperitoneal adhesions [4]. Alina et al. described a case of salpingocutaneous fistula in a case of peripartum hysterectomy where the fistula was formed during removal of the surgical drains when the fallopian tube moved under the skin [5]. Tubocutaneous fistula is a rare presentation of uterine malformations. In our first case patient had a vertical fusion defect (class U4aCoVo) with hematometra, hematosalpinx in non-communicating horn with abdominal menstruation and resultant adhesions [6]. With subsequent surgeries there was formation of fistulous tract between fallopian tube and skin.

The diagnosis of menstrual fistula is based on high index of clinical suspicion. One should suspect this entity in a postoperative case if there is persistent discharge from scar site despite treatment, and later with regularization of periods cyclical discharge from the scar site corresponding to menstrual bleeding [1–5]. Vellanki et al. reported a case where patient presented with groin sinus, recurrent painful swelling over edge of scar site after LSCS and was managed on lines of suture granuloma, scar endometrioma, underwent local drainage and excision but there was no relief in symptoms [7]. The diagnosis was made only after she was taken up for laparotomy. To confirm the diagnosis various bedside tests are available,
methylene blue dye test after inserting Foley catheter into uterus has been used to confirm the diagnosis [8]. With wide spread availability of contrast MRI scan and hysteroscopy complete delineation of fistulous tract and demonstration of its opening in uterus is used to confirm the diagnosis [9]. However, other modalities such as fistulography, hysterosalpingography, CT scan have been used for confirming the diagnosis and delineating the fistulous tract [7–9].

Due to rarity of this condition there is no recommended treatment modality. Medical, surgical or combined medical and surgical treatment have been tried [9–13]. Treatment depends on the age of the patient and the desire to retain fertility. There are case reports of successful medical management of uterocutaneous fistula with six months therapy of GnRH agonists [11, 12]. The mechanisms of action of GnRH agonists in the management of fistula are not clear. However, cessation of menstruation, endometrial atrophy, and decreasing uterine size may cause cicatrization and contracture of the fistulous tract, leading to fibrosis and closure [11–12]. Patients should be reviewed after six months for failure of medical management because of limitations of prolonged treatment with GnRH [9]. Surgery remains the mainstay of treatment. The actual demonstration of the fistulous tract and identification of its underlying cause is quite tedious and difficult. However, the type of resection should be individualized according to the needs of the patient [14]. For salpingocutaneous fistula one may advocate fistula resection and salpingectomy as the only feasible method of treating this rare disease to prevent the occurrence of ectopic pregnancy subsequently [13]. For uterocutaneous fistula, excision of fistulous tract with or without hysterectomy has been attempted. Maintaining the postoperative uterine drainage is essential to prevent recurrence.

CONCLUSION

Adopting good surgical techniques and prevention of postoperative infection can prevent this highly morbid condition. High index of clinical suspicion and prompt excision of the tract along with maintenance of good drainage of uterus post operatively is the key to successful management.

**********

Author Contributions

Seema Singhal – Substantial contributions to conception and design, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published
Sunesh Kumar – Analysis and interpretation of data, Critical revision of the article, Final approval of the version to be published
Yamini Kansal – Substantial contributions to conception and design, Acquisition of data, Drafting the article, Final approval of the version to be published
Deepika Gupta – Substantial contributions to conception and design, Acquisition of data, Drafting the article, Final approval of the version to be published
Mohit Joshi – Analysis and interpretation of data, Critical revision of 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 Seema Singhal 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

Pulmonary embolism and atrial fibrillation: A complicated relationship

Saroj Lohani, Niranjan Tachamo, Bidhya Timilsina, Salik Nazir

ABSTRACT

Introduction: Atrial fibrillation (AF) and pulmonary embolism (PE) share a complicated relationship. Pulmonary embolism has been associated with increased incidence and prevalence of AF. On the other hand, AF has also been associated with increased risk of PE in multiple cohort studies. What complicates the relationship further is that in some cases as in our case, patients presented with AF and were found to have PE. Case Report: A 72-year-old female presented to the emergency department with respiratory distress and was found to have atrial fibrillation with rapid ventricular response. She worsened despite appropriate treatment for atrial fibrillation. She was found to have massive pulmonary embolism after she underwent echocardiogram and CT scan of chest. However, she deteriorated quickly and died within five hours of presentation to the emergency department. Conclusion: Pulmonary embolism can precede, coexist or occur after atrial fibrillation. Clinicians should be alert about the complicated relationship of AF and PE.

Keywords: Atrial fibrillation, Anticoagulation, Cardioversion pulmonary embolism

INTRODUCTION

We present a case of 72-year-old female presenting to hospital with atrial fibrillation (AF) with rapid ventricular response and was diagnosed with massive pulmonary embolism (PE) after she deteriorated despite cardioversion and rate controlling agents. The relation between atrial fibrillation and pulmonary embolism has been extensively studied. Pulmonary embolism has been associated with increased incidence and prevalence of AF [1]. The main mechanism for AF in these patients has been proposed as right ventricular dilation and strain. On the other hand, AF has been associated with increased risk of pulmonary embolism in multiple cohort studies [2–4]. What complicates the relationship further is that in some cases [5, 6] and in our case, patients presented with atrial fibrillation and were found to have pulmonary embolism. Thus clinicians should be alert about the complicated relationship of AF and PE.

CASE REPORT

A 72-year-old female was presented to the emergency department after she was found gasping for breath. She had come from Florida few days back and was
complaining of symptoms suggestive of upper respiratory tract infection for the past one day. Past medical history was significant for deep venous thrombosis, paroxysmal atrial fibrillation not on anticoagulation, non-ischemic cardiomyopathy and hypertension. She did not smoke or drink alcohol. She was non-compliant with her medication for the past one and half years.

On arrival at the scene, she was found to have undetectable blood pressure by Emergency Medical System. On the spot electrocardiogram (EKG) revealed atrial fibrillation with rapid ventricular response (Figure 1). She received three electrical cardioversions with shock of 100 J, 150 J and 200 J en route to the emergency department but was unsuccessful in restoring normal sinus rhythm. On initial evaluation in the emergency department, she was sedated and on bag and mask ventilation (BMV). Her initial vital signs included blood pressure of 107/48 mmHg, pulse 160 s, respiratory rate 15/minute, and oxygen saturation 99% on BMV. Physical examination revealed irregularly irregular heart rate in 170 s but clear bilateral breath sounds. No jugular venous distension was noted. She was intubated in the emergency department and started on diltiazem drip for her rapid atrial fibrillation, following which her blood pressure dropped to 54/43 mmHg and she was started on pressor support with norepinephrine and epinephrine. Arterial blood gas on 100% oxygen on AC/CMV mode of ventilation revealed pH 7.03 (normal: 7.35–7.45), pCO2 69.2 mmHg (normal: 35–48 mmHg), pO2 157.4 mmHg (normal: 83–108 mmHg), bicarbonate 18.6 mEq/L (normal 21–28 mEq/L).

Patient’s labs included troponin I 0.04 ng/mL (normal <0.03 ng/mL), white blood cell count 7700/\(\text{mm}^3\) (normal: 4800–10800/\(\text{mm}^3\)), hemoglobin 14 g/dL (normal: 12–16 g/dL), platelets 183000/\(\text{mm}^3\) (normal: 130000-400000/\(\text{mm}^3\)), blood glucose 327 mg/dL (normal: 70–99 mg/dL), D-dimer 11809 ng/ml (normal: <500 ng/mL), Brain natriuretic peptide (BNP) 200 pg/mL (normal: 0–200 pg/mL) and INR 1.2 (normal: 0.9–1.1). Transthoracic echocardiogram revealed severely dilated right ventricle with severely decreased systolic function and moderate to severe tricuspid regurgitation. Computed tomography (CT) scan of chest showed large volume diffuse bilateral pulmonary emboli with a large saddle embolus (Figure 2). Computed tomography scan of chest also revealed reflex of contrast into the inferior vena cava and hepatic veins and bowing of the interventricular septum, suggestive of right heart strain. Shortly afterwards, she developed multiple episodes of cardiac arrest with pulseless electrical activity (PEA) and was successfully resuscitated each time. She was started on tissue plasminogen activator (tPA) infusion and transferred to intensive care unit. Cardiosurgery consultation deemed surgical embolectomy to be futile. Her family opted for no more resuscitation efforts and she passed away from subsequent PEA within five hours of presentation to the emergency department.

**DISCUSSION**

There are many ways in which we can define the relationship between atrial fibrillation (AF) and pulmonary embolism (PE). Pulmonary embolism has been associated with increased prevalence and subsequent incidence of AF. The prevalence of atrial fibrillation in PE patients is 18126 per 100,000 persons. The possible predictors of subsequent AF in PE patients include age, diabetes mellitus, obstructive sleep apnea, congestive heart failure, and admission sodium (hypernatremia) [1]. It has been proposed that the right ventricular dilation and strain associated with pulmonary embolism is the main mechanism for atrial fibrillation in these patients [7]. History of atrial fibrillation or presence of atrial fibrillation at admission in patients with pulmonary embolism also has prognostic implications. In a study done by Barra et al. [8], history of atrial fibrillation or atrial fibrillation at admission in patients with PE was associated with higher one month and six months mortality risk.

On the other hand, atrial fibrillation has been associated with increased risk of pulmonary embolism in multiple cohort studies [2–4]. In a study by Enga et al. [2], atrial fibrillation was associated with increased risk of pulmonary embolism during the first six months of diagnosis of atrial fibrillation and after six months. Higher
CHADS\textsubscript{2}VASc score is associated with increased incidence of pulmonary embolism [9]. Multiple mechanisms have been proposed for increased risk of pulmonary embolism in patients with atrial fibrillation. One of them is clot formation in right atrium. Spontaneous echo contrast has been noted in right atrium in patients with atrial fibrillation [10, 11]. The echo contrast is believed to proceed to frank clotting. Another mechanism is that atrial fibrillation is associated with hypercoagulability that might lead to clot formation in right atrium and subsequently lead to pulmonary embolism [12, 13].

It is not entirely clear whether atrial fibrillation precedes pulmonary embolism or vice versa. It is not clear if clarifying this relationship carries any clinical significance. Any patients with atrial fibrillation with CHADS\textsubscript{2}VASc score of 2 should be anticoagulated to prevent stroke and other ischemic events. The increased risk of pulmonary embolism in atrial fibrillation and relation of pulmonary embolism with CHADS\textsubscript{2}VASc score carries no implication for prescribing anticoagulation in atrial fibrillation. Chwan et al. suggested that patients presenting with pulmonary embolism should be screened for atrial fibrillation post pulmonary embolism as the cardiovascular cause specific deaths were higher in patients who developed atrial fibrillation during or after hospitalization [1]. The screening criteria however were not clearly defined. Kenneth M Flegel suggested that if further studies prove that atrial fibrillation is associated with a particular pulmonary embolus then it might be worthwhile intervening the atrial fibrillation [7].

Many of the clinical features like palpitations, shortness of breath, syncope, hypoxia, chest pain can be present in both atrial fibrillation and pulmonary embolism and focusing the treatment only on atrial fibrillation can mask the hidden pulmonary embolus as in our patient and lead to delay in diagnosis. Pulmonary embolism has been diagnosed during workup of patients presenting with new onset atrial fibrillation [14] or in patients with prior history of atrial fibrillation [15] as in our patient. The presence of AF in patients with acute PE independently predicts mortality [8]. Hence, it is important for clinicians to be aware of the association between atrial fibrillation and pulmonary embolism as it has clinical and prognostic implications.

**CONCLUSION**

Atrial fibrillation and pulmonary embolism have a complicated relationship. The risk of pulmonary embolism should be considered in patients presenting with new onset atrial fibrillation or with history of atrial fibrillation. Pulmonary embolism should also be kept in mind if patient presenting with atrial fibrillation with rapid ventricular response worsen despite appropriate treatment. Clinicians should be aware of increased incidence and prevalence of atrial fibrillation in patients with pulmonary embolism.


An indirect sinus floor elevation by using piezoelectric surgery with platelet-rich fibrin for sinus augmentation: A short surgical practice

Ali H. Neamat, Shakhawan M. Ali, Saman W. Boskani, Payman Kh. Mahmud

ABSTRACT

Introduction: The aim of this surgical practice is to present a case report to describe a technique for sinus floor augmentation of an atrophic posterior maxilla by platelet-rich fibrin (PRF) has been used as a graft material with a one-step crestal approach (indirect sinus lift) where the residual bone is less than 7 mm in the atrophic posterior maxilla. Sinus floor elevation was performed with a crestal approach by using piezosurgery. Case Report: A 38-year-old female with an atrophic right posterior maxilla was treated with sinus floor augmentation and immediate implant placement using PRF as the sole graft material in our implant clinic, maxillofacial department, Sulaimany teaching hospital prior to surgery the residual bone height was approximately 2–4 mm but six months after surgery, orthopantomogram show that the use of PRF as a graft material during sinus floor augmentation induces natural bone regeneration. Conclusion: Herein, illustrate a minimally invasive procedure aimed at sinus floor elevation with immediate implant placement by crestal approach, PRF only is a safer and easy available simpler technique than the protocol graft materials and piezosurgery device to prevent Schneiderian membrane perforation.

Keywords: Immediate implants, Indirect sinus lift, Piezosurgery, Platelet-rich fibrin

INTRODUCTION

Dental implants are used to replace both the form and the function of missing teeth but an insufficient vertical height of the alveolar bone in the posterior maxillary area due to the presence of the maxillary sinus, poor quality and quantity of alveolar bone and post-extraction bone resorption may limit implant placement [1]. In such cases, several sinus augmentation procedures for implant placement have been introduced since the 1980s [2, 3]. The most widely used approaches for sinus lifting are...
direct sinus lifting approach and indirect sinus lifting approach. The indirect sinus lift procedure presents the advantage of being less invasive and less time to consume compared to direct sinus lift. Using piezoelectric ultrasonic vibration (25–30 kHz), the piezosurgery device precisely drills the bone without cutting soft tissues, which remain undamaged even in a case of accidental contact (Figure 1) [4]. The piezosurgery device provides a clear surgical site, as it maintains a blood-free surgical field this allows improved visualization of the surgical area during bone cutting, due to the air–water cavitation effect of the ultrasonic instrument. The main advantage of the osteotome technique is that it improves bone density, which allows greater initial stability of implants also it is a less invasive procedure than lateral antrostomy [5]. After progressive preparation of the bone then the elevation of the sinus floor is obtained with a reduced operative time compared with other sinus graft procedures. The movement of the piezosurgery knife is very small, so the cutting precision is greater and causes less discomfort for the patient. The absence of macro vibrations makes the instrument more manageable and allows greater intraoperative control, with a consequent safer action in anatomically difficult situations. When using this instrument the clinician applies a very small amount of pressure which allows a very precise cut [6]. The techniques create space between the maxillary alveolar process and the elevated Schneiderian membrane, which is filled with various graft materials to maintain adequate space for new bone formation. Many graft materials have been applied to these techniques such as autogenous bone, allograft, xenograft, alloplastic bone, or combinations thereof [7, 8]. Although autogenous bone is considered to be the gold standard, but it creates another wound at the donor site. Thus, autogenous bone is not widely used in clinical practice. The other graft materials also have limitations, including the risk of infection, insufficient bone regeneration and increased overall cost. Accordingly, no graft material appears to be superior to the others.

Several studies have recently reported the application of PRF in dental implant surgery such as PRF mixed bone substitute or PRF has so far solely been used as a graft material for sinus augmentation using both the lateral and crestal approach [9]. Platelet-rich fibrin (PRF) was first reported in 2001 by Choukroun et al. as a second-generation platelet concentrate [10]. The application of PRF for sinus augmentation is a relatively easy surgical procedure and the clinical and radiological findings have been shown to have a good effect regarding new bone formation.

Platelet concentrates have been shown to be a promising scaffold for tissue regeneration. Platelet concentrates are autologous, easy to prepare at the chair side, and full of high concentrations of growth factors. In vitro studies have proved the effects of these signaling molecules on cell proliferation, migration, differentiation and matrix synthesis. In recent years, platelet concentrates have been applied in sinus floor elevation and bone grafting. Platelet-rich plasma (PRP), which was among the first generation of platelet concentrates, has been used with autogenous bone or a bone substitute in sinus augmentation but without significantly positive effects. Platelet-rich fibrin (PRF) is from the second generation of platelet concentrate products. Platelet-rich fibrin has many advantages over Platelet-rich plasma, including osteogenic ability, a simple preparation process, absence of extrinsic biological agents, and sustained release of growth factors. Previous studies have revealed the potential of PRF to promote endo-sinus bone regeneration and to reduce healing time after sinus floor elevation [11].

CASE REPORT

A 38-year-old female presented to our implant clinic, maxillofacial department, Sulaimany Teaching Hospital with multiple missing teeth. A clinical examination was done followed by the radiographic examination using OPG (Figure 2). On evaluation, it was found that the right premolar and molar region had only 2–4 mm bone and an indirect sinus lift procedure with immediate implant placement was planned. The anterior region had adequate bone height. Laboratory blood investigation was done and antibiotic prophylaxis was given 1 hour prior to the surgery.

Surgical phase

• Start by putting a butterfly cannula in cephalic vein of the patient and blood samples approximately 50 ml is collected put in 10 ml blood collection tubes centrifuged immediately in a table-top centrifuge at a rate of 3000 rpm for 10 minutes then in the test tube three layers are obtained after centrifugation (Figure 3). Platelet-poor plasma is a top most layer consisting of cellular PPP, Clot in the middle are PRF and RBCs at the bottom of the test tube. PRF in middle layer clot is then removed with sterile tweezers and separated from the underlying RBC layer using scissors and then put on a sterile dish (Figure 4).

  • Gave Local anesthesia and by no 15 blade crestal incision was made.
  • Envelop flap was reflected and it was found that the bone width was sufficient.
  • Initial depth was achieved using a marker drill 2 mm at a speed of 700 RPM with saline irrigation.
  • Then that piezoelectric device was used. The osteotomy began to continue osteotomy was preparation and come into contact with the sinus membrane (Figure 1).
  • The sinus was lifted indirectly by using sinus elevation instruments. After that, the PRF material was inserted.
  • A final osteotomy was done then W&H Implant motor driven at 40 rpm and 20 Ncm torque
• After selecting a proper size and diameter three fixture were placed 2 of them D 5 mm* L 6 mm, While the other D 3 mm* L 13 mm.
• Put Cover screw and suturing was done using multifilament 3-0 absorbable polyglactin sutures.
• Orthopantomogram (OPG) was taken directly postoperative (Figure 1B). Also, the patient was instructed to the cold application for two hours after the operation, maintain the oral hygiene, mouthwash with antibiotic, analgesic and good follow-up.
• Implant loaded after four months (Figure 5).
• After six months OPG showed more than 3 mm bone regeneration by PRF around the implant (Figure 6).

DISCUSSION

Advancement in dentistry continues, placement of implant has become the most preferred means of replacement of missing teeth. In posterior maxilla because of the maxillary sinus in most of the cases presents a problem. Solution to these cases it is necessary to elevate the sinus floor and increase the bone height before implant placement. Direct sinus lift and indirect sinus lift are the most commonly used techniques. In the direct technique, the sinus is approached through the lateral window.

Figure 1: Ultrasonic piezoelectric device. This device works in hard tissue, not in the soft tissue. Therefore, membrane perforation is rare when breaking sinus floor (Courtesy Pr. Dong-Seok Sohn) [12].

Figure 2: (A) Preoperative image showing atrophic right maxillary bone less than 2–4 mm in the molar region. (B) Directly postoperation implant in situ after sinus left.

Figure 3: Platelet-rich fibrin procedure (Courtesy Pr. Dong-Seok Sohn) [12].

Figure 4: Platelet-rich fibrin on sterile dish after separation ready for use.

Figure 5: Operation outcome successfully after four months.

Figure 6: Showing amount of bone regeneration by PRF in two different times. (A) Three months after the operation, and (B) Six months after the operation.
technique most of the time whereas indirect technique follows a crestal approach. Herein this article the indirect sinus lifting approach using sinus elevation instruments has been followed by increasing the bone height and also piezosurgery has advantages of micrometric bone cut to provide precise bone cut and of working in only hard tissue, not in the soft tissue easily getting tactile sense of the sinus membrane during osteotomy to the sinus floor, and reducing the risk of perforation of sinus membrane.

The crestal approach for sinus augmentation provides 97% success rate, by minimizing Schneiderian membrane perforation and the bone would form around the implant in four months and could be loaded [13]. But the only disadvantage of this technique is that it is a blind procedure and only 3–4 mm sinus elevation can be achieved.

The present case showed a bone height of 2–4 mm before operation radiographically and the sinus floor was elevated up to 3 mm for the placement of an implant of 6 mm length and 5 mm diameter. But according to the study conducted by Ahn et al. the mean height of the residual alveolar process was 5.8 (0.9) mm, whereas mean elevation of the sinus floor was 6.2 (0.4) mm using indirect sinus lift. The study by Fornell et al. [14] showed a mean bone gain of 3 mm without any marginal bone loss after 3–12 months which was similar to the present case report.

CONCLUSION

Really most of the time placement of implants can be difficult due to compromised situations especially in the posterior maxillary region, by good evaluation and use the various techniques available it is possible to provide a good treatment outcome. The indirect sinus lift procedure and platelet-rich fibrin used as the sole graft material for sinus augmentation along with immediate implant placement helps to reduce the treatment time in patients. Herein by keeping the treatment procedure, simple successful rehabilitation can be achieved. The piezosurgery technique is a predictable sinus augmentation method without having to use the striking of a mallet. It may be a good choice at an implant site that has at least 3-mm residual bone under the maxillary sinus floor because it reduces the possibility of membrane perforation. It also reduces surgical time and discomfort to the patient.

*********

Acknowledgements

We would like to thank ministry of health Kurdistan regional government and implant department at a Sulaiman teaching hospital for made a facility to this operation.

Author Contributions

Ali H. Neamat – 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

Shakhawan M. Ali – 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

Saman W. Boskani – 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

Payman Kh. Mahmud – 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

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

Copyright

© 2017 Ali H. Neamat 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


Access full text article on other devices

Access PDF of article on other devices
A rare case of primary squamous cell carcinoma of the stomach

Xu Chen, Chengxin Luo, Hu Zhang

ABSTRACT

Introduction: Given that stomach is lined by glandular epithelium, the most common type of gastric malignancy is adenocarcinoma, which accounts for more than 90% of primary gastric carcinomas. Primary squamous cell carcinoma of the stomach is an extremely rare entity with an incidence of 0.04–0.07% among all gastric carcinomas. In this study, we report a case of a 66-year-old male and review literature about this uncommon disease. Case Report: A 66-year-old male presented to our outpatient department and complained about epigastric pain. Gastroscopy found a protruding mass (about 3.0x4.0 cm) with central ulceration on the greater curvature of the middle gastric body. He was finally diagnosed with primary squamous cell carcinoma of the stomach based on pathological examination of biopsies. The patient declined further examination and therapy, lost to follow-up after discharge. Conclusion: Primary squamous cell carcinoma of the stomach is an extremely rare entity. This type of tumor is aggressive and prone to metastasize to lymph nodes and liver, it is critical to make a correct diagnosis at early stage and place the patients on appropriate management.

Keywords: Adenocarcinoma, Gastric carcinoma, Primary squamous cell carcinoma of the stomach, Squamous metaplasia

INTRODUCTION

The most common type of gastric malignancy is adenocarcinoma and primary squamous cell carcinoma of the stomach is extremely rare [1, 2]. The pathogenesis is still not well understood for the latter. It exhibits non-specific symptoms such as epigastric pain, etc. The prognosis is poor since it is usually diagnosed at an advanced stage and prone to metastasize to lymph nodes and liver. No general consensus on the management of this disease is available. This study presents a case of a 66-year-old male who presented as epigastric pain and protruding mass on the gastric body. Epidemiological characteristics, hypotheses regarding pathogenesis, available diagnostic criteria, management and prognosis are also discussed based on literature review.

CASE REPORT

A 66-year-old male was presented to our outpatient department with a complaint of epigastric pain for four months. He denied any other symptoms like dysphagia,
hematemesis, and melena. His past medical history was unremarkable. Mild tenderness was elicited on the epigastric area. Other systems were normal on physical examination. No data of laboratory tests was available. An upper gastrointestinal endoscopy was performed, which revealed a protruding mass (about 3.0x4.0 cm) with central ulceration on the greater curvature of the middle gastric body. The mass was friable and covered by yellow greasy fur (Figure 1). Endoscopic observation was normal for the esophagus, gastroesophageal junction and cardia. Multiple endoscopic biopsies were taken for histopathological examination, identifying a moderately differentiated squamous cell carcinoma with keratinized cell masses and keratin pearl formation under hematoxylin and eosin staining (Figure 2). No evidence of adenocarcinoma was found. Finally, a diagnosis of primary squamous cell carcinoma (SCC) of the stomach was confirmed. Unfortunately, the patient declined further examination and therapy, lost to follow-up after discharge.

DISCUSSION

It is well known that the stomach is lined by glandular epithelium, the most common type of gastric malignancy is adenocarcinoma, which accounts for more than 90% of primary gastric carcinomas [1, 2]. Primary SCC of the stomach is an extremely rare entity with an incidence of 0.04–0.07% among all gastric carcinomas [3]. It was first described by Rörig et al. in 1895 and less than 100 cases have been reported in English literature [3]. There is a male predominance in the incidence of primary gastric SCC, with a male and female ratio 5:1 [4]. The peak incidence is in the sixth decade of life, however, a case as young as 17 years old has been reported by Schwab et al. [3, 5]. Wakabayashi et al. reviewed 56 cases based on Japanese literature and reported a median age of 64.7±1.7, the ratio of male/female was 3.7 (44/12) [6]. A retrospective analysis included 21 patients diagnosed in China reported a higher median age of 67 years old, a male-female ratio of 6:1 [7]. The most common location of tumor is the upper third of the stomach (57.1%, 66.7%) [6, 7]. The common clinical manifestations were identical with other type of gastric tumors, included abdominal pain, nausea, vomiting, melena, hematemesis, and weight loss [4, 7]. Symptoms of paraneoplastic syndrome like hypercalcemia and leukocytosis also have been reported [8, 9]. In addition, there was a patient of SCC of the stomach that presented as a huge retroperitoneal tumor revealed by CT scan and showed no significant results on endoscopic examination [10]. Here we reported a 66-year-old male patient with primary SCC of the stomach, who presented with upper abdominal pain, in line with the epidemiology characteristics of this uncommon disease.

The pathogenesis of primary SCC of the stomach are still not well understood and various theories regarding its histogenesis have been proposed, including

- the presence of nests of ectopic squamous cells in gastric mucosa;
- squamous metaplasia of the gastric mucosa before malignant transformation;
- squamous differentiation in a preexisting adenocarcinoma;
- totipotent stem cells in the gastric mucosa which is capable of differentiating into any cell type [1, 11].

Islands of squamous epithelium have been identified in the gastric mucosa of individuals without SCC, supporting the theory that the primary SCC of the stomach may arise from ectopic squamous cells [12–14]. Squamous metaplasia may occur in the margin of a gastric peptic ulcer [11]. Corrosive acid burns, infection with syphilis, chemotherapy for lymphocytic lymphoma,
chronic inflammation and foveolar hyperplasia in Menetrier’s disease were also reported to be associated with squamous metaplasia of the stomach, which was further followed by development of squamous cell carcinoma [15–18]. In addition to squamous metaplasia, SCC was also reported to arise in the context of chronic atrophic gastritis with intestinal metaplasia [3, 17]. Another theory suggested SCC may origin from the overgrowth of a squamous epithelium element in a primary adenocarcinoma, since the reexamination of previously diagnosed pure squamous cell carcinoma revealed components of adenocarcinoma [3, 19]. Mori et al. proposed the hypothesis that multipotential stem cells first turn into adenocarcinoma, followed by the occurrence of squamous metaplasia, which finally turn into SCC [19].

Besides the above etiological theories, some other risk factors have also been suggested for the pathogenesis of primary gastric SCC. Takita et al. proposed that Epstein-Barr virus (EBV) infection may play a role in the development of gastric SCC given that the evidence of EBV infection was witnessed in surgical specimens of the tumor [20]. However, in situ hybridization of other patients failed to confirm evidence of EBV infection [9, 21]. The long-term use of cyclophosphamide has ever been suggested as a risk factor for the development of gastric SCC in patients with multiple myeloma and lupus erythematosus [22]. In addition, Chen et al. reported that 61.9% of patients with gastric SCC had a long history of smoking [7]. It is well known that smoking plays an important role in the pathogenesis of squamous carcinoma of lung and esophagus. Taken together with Chen’s finding, it is suggested that smoking may be a risk factor for this disease.

Although primary gastric SCC is very rare, some diagnostic criteria have been suggested for it. According to the Japanese Classification of Gastric Carcinoma, the diagnostic criteria of primary SCC of the stomach include:

1. all tumor cells are SCC cells, without components of adenocarcinoma in any sections
2. there is distinct evidence that SCC arises directly from the gastric mucosa [3, 23].

To exclude SCC that extended from esophageal carcinoma and other primary sources, Parks proposed three diagnostic criteria:

1. the tumor should not be located in the cardia;
2. the tumor must not extend into the esophagus;
3. there must be no evidence of SCC in any other part of the body [1, 3].

The histopathological criteria for primary SCC of the stomach were identified by Boswell and Helwig as followings:

1. keratinized cell masses with typical keratin pearls formation;
2. a mosaic pattern of cell arrangement with sharp borders;
3. the presence of intercellular bridges;
4. high concentrations of sulphydryl or disulphide bonds which indicates the presence of keratin [14].

Immunohistochemistry analysis for indicators of squamous cell carcinoma (p63 and CK5/6), is widely performed to confirm the diagnosis, with a high specificity of 99% and a sensitivity of 98% [7, 24]. CK7, the indicator for adenocarcinoma, is negative in pure squamous cell carcinoma [3].

As our case is concerned, the tumor was located in the greater curvature of the middle gastric body. The possibility of involvement of cardia and esophagus was ruled out through endoscopic examination. No evidence of tumors in any other organs including skin was observed on physical examination. Thoracic computed tomography was carried out to exclude tumors originating from lung. Histologically, the tumor showed moderately differentiated squamous cell carcinoma without glandular components. Above all, our case meets the diagnostic criteria of primary SCC of the stomach.

Due to the rarity of primary SCC of the stomach, there is no general consensus on the management of this disease. Radical surgical resection remains the mainstay of the treatment for localized disease. Surgery to achieve R0 (no residual tumor) resection can substantially improve outcome. A retrospective analysis reported that patients could achieve a median survival time of 46 months in surgery group and 4.5 months in non-surgery group [7]. For advanced-stage SCC of the stomach, survival after surgical resection is poor and adjuvant chemotherapy may improve the prognosis [6]. Combined postoperative radiotherapy and chemotherapy with aggressive surgical resection have been reported to result in a recurrence-free survival time of five years [25]. However, no standard chemotherapy regimen has been established. The most commonly used chemotherapy regimens were based on 5-fluorouracil [7]. Intrahepatic administration with 5-fluorouracil plus mitomycin was reported to obtain complete response in metastatic SCC in liver [4]. A patient on 5-fluorouracil plus cisplatin chemotherapy after surgery has survived at least 45 months [7]. Amazing effectiveness of neoadjuvant chemotherapy with low-dose 5-fluorouracil plus was first demonstrated by Marubashi et al. in a 70-year-old male patient [26]. Neoadjuvant chemotherapy with carboplatin and paclitaxel followed by successful surgical resection was also reported to improve a clinical outcome [27].

It is difficult to predict the prognosis of primary SCC of the stomach. Generally speaking, its prognosis is better than gastric adenocarcinoma [7]. An overall survival time ranging from 7 months to 8 years has been reported by Gao et al. [4], but it has also been reported that a patient died within three months of admission in Turkey [11]. However, primary gastric SCC is usually diagnosed at an advanced stage. Its marked local infiltration and distant metastasis often means a poor outcome [6]. Given that this uncommon tumor is aggressive and prone to metastasize to lymph nodes and liver, it is critical to make
CONCLUSION

Primary squamous cell carcinoma of the stomach is an extremely rare entity which has a peak incidence in the sixth decade of life and non-specific symptoms identical with other type of gastric tumors. It is usually diagnosed at an advanced stage and the outcome turns out to be poor. So prompt diagnosis and appropriate management are critical since it is aggressive and prone to metastasize to lymph nodes and liver.

Author Contributions
Xu Chen – Substantial contributions to conception and design, Drafting the article, Final approval of the version to be published
Chengxin Luo – Substantial contributions to conception and design, Drafting the article, Final approval of the version to be published
Hu Zhang – 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 Xu Chen 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

Live *Ascaris* in anterior chamber causing hypertensive uveitis

Anadi Khatri, Bal Kumar Khatri

**ABSTRACT**

Introduction: Human ocular parasitosis is common in the parts of the world—especially in the developing and the underdeveloped nations where there is poor knowledge regarding hygiene and the devastating consequences that may arise due to its disregard. Case Report: Herein, we describe a 42-year-old male presenting with decreased vision with painful red eye since five days. Best corrected Visual Acuity (BCVA) was 1/60 and 6/6. The right eye was congested; hypopyon and vitreous exudates present. Single live freely motile worm noted in anterior chamber. Flat retina with hyperechoic vitreous shadows observed in ocular ultrasonography. Intraocular pressure was 38 and 17 mmHg. No relevant systemic association identified. Surgical retrieval of live worm was achieved via corneal incision. The worm was identified as adult male *Ascaris lumbricoides* (26x2 mm) by parasitologist. Blood eosinophil count was raised and adult worms and ova of *Ascaris lumbricoides* was also seen in the stool. The eye was treated with topical steroid, antiglaucoma and cycloplegic agents supplemented by oral anthelmintic and corticosteroid (1 mg/kg). Visual recovery was achieved. Conclusion: In patients presenting early with intraocular parasite, surgical removal of the live adult worm along with oral steroids is recommended to treat and preserve vision.

Keywords: *Ascaris*, Parasite, Uveitis, Viscoexpression

**INTRODUCTION**

Intraocular invasion by *Ascaris lumbricoides* is an extremely rare condition. It can present suddenly and with an adult worm with no obvious posterior segment pathology (except uveitis). Also, commonly known as the roundworm, it infects humans when an fertilized egg of the organism is ingested. It then transforms into a larval worm which penetrates the wall of the duodenum and enters the blood stream. It is then transported to the liver and heart from where it enters pulmonary circulation to reach the alveoli. In three weeks, the larva passes from the respiratory system to be coughed up, swallowed, and thus returned to the small intestine, where it matures to an adult male or female worm [1].

*A. lumbricoides* is characterized by its great size. Males are 2–4 mm in diameter and 15–31 cm long. The male’s posterior end is curved ventrally and has a bluntly pointed tail. Females are 3–6 mm wide and 20–49 cm long [2]. More than 1 billion people are affected by this infection [3]. Ocular infestation by this parasite has
been described earlier in nasolacrimal passage and an intraocular invasion by its larval form dating back to 1937 [2].

Surgical removal is essential and best done with no touch technique to prevent capsule of the worm peeling off and for prevention of both vision and life-threatening allergic reaction.

CASE REPORT

A 42-year-old male presented to our outpatient department complaining of dull aching pain with the sensation of ‘something moving’ in the right eye. It was associated with redness, watering and photophobia. He denied previous ocular or systemic symptoms. General physical examination was unremarkable. He gives no history of fever, vomiting, abnormal/involuntary body movements, pruritus.

The patient has no history of diabetes, hypertension or any other chronic illness for which he has been treated or is receiving treatment. He is a non-vegetarian – consumes fish, chicken and egg. He denies intake of raw meat. He is a farmer by occupation and has no similar complains/problem in the past.

The patient’s vision in right eye was 1/60 and 6/6 in left eye. On slit lamp examination, there was diffuse congestion of the conjunctiva of the right eye with circumcorneal congestion. The cornea was clear. Hypopyon of 1 mm was present. A moving worm was seen in the anterior chamber of the right eye. Pilocarpine was added to prevent the worm from escaping from anterior chamber and shifted to operating room. Posterior segment findings of the right eye could not be appreciated and the findings of the left eye were normal.

Investigations

Intraocular pressure (IOP) was 38 mmHg in right eye and 17 mmHg in left eye. B-scan was performed (longitudinal) which showed flat retina with hyperechoic vitreous shadows, Blood and stool was collected from the patient and sent for microbiological investigation. Blood examination revealed eosinophilia (6%). The other lineage were within normal limit. Stool examination revealed adult worms and ova in the stool of A. lumbricoides.

Differential diagnosis

Right eye Pan Uveitis secondary to live intraocular parasite (Phylum)

Treatment

Patient was planned for right eye live intraocular foreign body removal (worm) under subconjunctival anesthesia. Under subconjunctival anesthesia, a clear corneal incision was made at temporal and nasal side measuring approx 2.8 mm. Viscoelastics—hydroxypropyl methylcellulose— was injected from the nasal port to expel the worm with no touch technique to prevent compromise in its capsule. Upon expulsion, AC wash was done and subconjunctival injection of vancomycin and dexamethasone was given. The worm was placed in saline (later transferred to ethanol 70%) and sent for a specialized parasitology department. The worm was identified as an adult male A. lumbricoides measuring 26 mm long and 2 mm wide. The patient was prescribed eye drop ofloxacin, prednisolone, timolol, systemic acetazolamide, tropicamide along with systemic prednisolone (1 mg/kg) and albendazole stat (Figures 1–4).

Outcome and follow-up

On subsequent follow-ups, his visual acuity improved to 6/18 at third week with no further improvement in later follow-ups (At week 6 and 8) even after refraction.

Fundus examinations including ultrasonography (A/B scan) revealed normal nomenclature bilaterally. Intraocular pressure was 14 and 16 mmHg respectively.

DISCUSSION

Intraocular invasion by A. lumbricoides is an extremely rare condition. It can present suddenly and with an adult worm with no obvious posterior segment pathology (except uveitis). Surgical removal is essential and best done with no touch technique to prevent capsule of the worm peeling off and for prevention of both vision and life-threatening allergic reaction.

A. lumbricoides is the largest intestinal nematode parasitizing man. Man is the only definitive host of A. lumbricoides. This parasite is unique in that it passes its entire life cycle in one host. Continuance of the species is ensured by transference from one host

Figure 1: Live A. can be seen resting over the iris from around 9–12 o’clock.
to another. The female is capable of laying both fertilized and unfertilized eggs. The mature female worm has an enormous egg laying capacity, liberating about 200,000 eggs daily [4]. Kean et al. have reported that eggs of *Ascaris* behave like an inert material [5]. It is the larva produces an irritant fluid due to the presence of Ascarase, resulting in allergic manifestation. and when absorbed, is toxic and may damage the tissues. The worm is known to cause subconjunctival mass, granulomatous iridocyclitis, choroiditis (macular or paramacular), recurrent vitreous hemorrhage, periphlebitis, papilloedema, chronic dacryocystitis and invasion into the subretinal space [5]. *Ascaris* larvae do not normally develop in the eye [4] as shown by animal experiments with intraocular injection of *Ascaris* ova.

**CONCLUSION**

Management of live ascariasis in anterior chamber surgically has never been described in any literature. The use of viscoelastics caused restricted movements and also prevented it outer coating materials to be directly come in contact with the surrounding tissue. The technique of viscoexpulsion could be helpful in future cases in this regard. The definitive diagnosis of the type of worm should be definitely done by morphological evaluation of the adult worm. Nonetheless, some patients may not have them detectable in the blood due to the following reasons i) infestation by a sole male or female adult and/ or ii) low parasitaemia load. The worms can incubate for months and even years before they start to migrate. Surgical removal of the live adult worm is recommended.

********

**Author Contributions**

Anadi Khatri – 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

Bal Kumar Khatri – 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 Anadi Khatri 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

Acute colonic pseudoobstruction (Ogilvie’s syndrome) as a postoperative complication: A case report and literature review

Alaa Sedik, Mufid Maaly, Salwa ElHoushy

ABSTRACT

Introduction: Acute colonic pseudoobstruction, also known as Ogilvie’s syndrome, is an acute clinical condition with clinical and radiological features of an acute large bowel obstruction in the absence of any mechanical cause; yet a medical or surgical predisposing factors are present. Case Report: We describe a middle aged male victim of road traffic accident (RTA), who presented lately with acute colonic pseudoobstruction without any apparent cause following recent lumbar spine surgery for trauma. Conclusion: This case report highlights the rare, but potentially dangerous, diagnosis of Ogilvie’s syndrome after spinal fracture fixation. Rapid diagnosis is the key to avoid serious complications.

Keywords: Ogilvie’ syndrome, Spine, Surgery

INTRODUCTION

Acute colonic pseudoobstruction (Ogilvie’s syndrome) is characterized by abdominal distension and massive colonic dilatation without any mechanical cause of obstruction [1]. Ogilvie first described this syndrome in 1948 [2]. The pathogenesis remains unknown but likely involves imbalance between sympathetic and parasympathetic colon innervation [3]. Trauma, sepsis, and intrapelvic, neurosurgical or orthopedic surgery have been reported as etiological factors [4, 5]. As regards surgical patients, some have been reported after cervical discectomy [1], after cesarean section [6], after spinal anesthesia, trauma or surgery [4, 5]. And after laparoscopic surgery [6]. We report a case of Ogilvie’s syndrome after lumbar vertebral surgery. The pathophysiology and treatment are discussed based on a review of literature.

CASE REPORT

A 44-year-old Egyptian male; not known to have any medical problems before; was admitted as RTA victim. He had recently underwent operative fixation for unstable lumbar spinal fracture five days prior to surgery consultation. He had also a plaster cast applied to a fracture of left ankle. Patient had three-day history of progressive abdominal distension with diffuse pain, nausea and occasional nonbilious non bloody vomiting. Patient did not passed stools or flatus. On general examination, a bedridden obese male having stable vital signs and normal temperature. He was in pain, mildly dehydrated. Locally, the abdomen was diffusely
distended with diffuse tenderness maximally over right lower quadrant with rigidity and positive rebound. A tympanic note was noted with sluggish bowel sounds. The rectum was empty. Labworks showed mild leukocytosis with elevated serum creatinine and urea. X-rays of the abdomen showed hugely distended colon. Volvulus then contrast enhanced abdominal and pelvic CT scan confirms the possibility of volvulus of cecum (Figure 1).

Resuscitation was started to prepare for surgery and a nasogastric tube was inserted. The situation was fully discussed and informed consent obtained about the need for urgent laparotomy as its late to ask for colonoscopy. Patient was taken to the theatre and the abdomen explored through a long midline incision. Exploration showed intraperitoneal serosanguineous fluid and a hugely distended ischemic right colon up to the right half of the transverse colon with extensive serosal tears and edema of the wall. The cecal diameter exceeded 15 cm. The rest of the colon was distended down the rectum but looked viable. No masses or volvulus were found along the entire colon. The patient was offered right hemicolectomy with right quadrant stoma fashioned (ileostomy and mucus fistula) (Figure 2). The abdomen was closed over a pelvic drain. Postoperatively, he made uneventful recovery and tolerating gradually oral feeding with viable functioning stomas. He was discharged in good condition for possible outpatient department follow-up.

**DISCUSSION**

Colon in the absence of an anatomic or mechanical lesion that obstructs the flow of intestinal contents. This form of adynamic ileus is also named Ogilvie's syndrome. It carries the name of the British surgeon Sir William Heneage Ogilvie (1887–1971), who first reported it, in 1948 [2]. The most serious complication of Ogilvie’s syndrome is perforation of the cecum. Early recognition and treatment of pseudo-obstruction of the colon may prevent cecal perforation, which reportedly carries a mortality rate of 25–60% [4]. Signs and symptoms resemble those of paralytic ileus. It affects mainly elderly, bedridden patients. The pathogenesis remains unknown but likely involves imbalance between sympathetic and parasympathetic colon innervation [6]. The fact that Ogilvie’s syndrome may occur after manipulation of vertebral structures (spinal anesthesia, lumbar surgery) could support this hypothesis. It is known that the key is early recognition and rapid medical approach is the most appropriate treatment in patients presenting without complications. Medical treatment in the form of nasogastric suction, rectal tubes, keep Nulla per os, correction of fluid and electrolyte imbalance, and parasympathetic agent neostigmine administration if the diagnosis is made early and abdominal radiograph shows a diameter of the distended cecum less than 9 cm [7, 8]. If cecum overdistention occurs, then colonoscopic decompression can be proposed with possible passing a rectal tube [9]. Surgical management is indicated if cecal diameter is greater than 9 cm or conservative treatment is ineffective after or there is evidence of cecal perforation. In absence of perforation or bowel ischemia, cecostomy is the procedure of choice but in cases of bowel ischemia and perforation resection with or without primary anastomosis should be performed [10, 11]. The complications of Ogilvie syndrome include perforation, peritonitis and shock. The mortality rates in cases of colonic perforation are 43–50%. In our case, no medical treatment or endoscopic decompression was attempted because the clinical and radiological evidences warranted urgent surgery.

**CONCLUSION**

Ogilvie’s syndrome is a rare condition and should be differentiated from toxic megacolon where urgent surgery is recommended. Vertebral surgery or trauma probably acts as predisposing factor. Complications following pseudo-obstruction may be avoided if pseudo-obstruction is recognized early and treated with optimal conservative treatment including para-sympathetic drugs. In case of failure of medical treatment or complications, surgery remains the treatment of choice.

***********
Author Contributions
Alaa Sedik – 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
Mufid Maaly – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published
Salwa ElHoushy – 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 Alaa Sedik 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
A misdiagnosis of benign abdominal mass as a non-Hodgkin lymphoma

Luma Haj Kassem, Ahmad Ghazal, Khawla Mayoh, Najat Mehio Sailam

ABSTRACT

Introduction: Vascular transformation of sinuses (VTS) is a rare reactive process. The lymph node sinuses got converted into a complex anastomosing network of vascular channels. Case Report: We report a unique case of VTS in a healthy 13-year-old female who complained only of abdominal pain with no other findings. She was misdiagnosed as non-Hodgkin lymphoma by FNA on a retrocaval mass and administered with chemotherapy before performing an excisional biopsy. Her pain recurred after a year; another course of chemotherapy was determined, but the patient and her parents refused it. Another clinician was consulted and he decided to perform an excisional biopsy which revealed VTS with no malignancy. Conclusion: By reviewing literature, the majority of VTS cases were reported in adults and associated with neoplasia, but the case described here may be the first case to be reported about VTS in a child, who has been healthy to date. We also report this case to emphasize the role of diagnosis before making any therapeutic procedure, in particular chemotherapy.

Keywords: Lymph node, Lymphoma, Misdiagnosis, Vascular transformation

INTRODUCTION

Vascular transformation of sinuses (VTS) is a rare condition in which the lymph node sinuses are characterized by vascular proliferations. In this condition, capsule, parenchyma and perinodal fibroadipose tissues are not involved [1, 2]. It is often found incidentally in surgical specimens of lymph nodes, but it can also be present as lymphadenopathy [3].

Vascular transformation of sinuses (VTS) has variant histological features such as vasodilation with minimal changes and vascular proliferations, which resemble Kaposi’s sarcoma [4, 5]. These histological features, result of the duration of regional lymphatic and/or venous obstruction [3, 5, 6].

We report a unique case of VTS in a healthy young female, who complained only of abdominal pain. She was misdiagnosed as a non-Hodgkin lymphoma and administered with chemotherapy before performing an excisional biopsy. To our knowledge, the case described here may be the first case to be reported about VTS in a child in literature.
CASE REPORT

A 13-year-old female presented to the hospital complaining of intermittent, moderate and vague abdominal pain for several months. The pain has a sudden onset, without any radiation or relieving/exacerbating factors. She denied any fever, vomiting, nausea or weight loss. Her medical and family history was unremarkable. Her vital signs were completely normal. On physical examination, there were not any visceral enlargements or masses, any axillary or inguinal lymph nodes could be palpated; there were not any positive findings. All laboratory tests were almost normal except of erythrocyte sedimentation rate (ESR) which was raised 1st hour 25 mm.

An abdominal ultrasound was performed and showed a 4.5x3 cm mass (Figures 1), which was confirmed on abdominal computed tomography (CT) scan. Abdominal pelvic multi slice CT scan before and after contrast in sagittal and coronal sections demonstrated that the mass was retro caval adjacent to the right adrenal gland (Figures 2–4).

The mass was oval, well defined, homogeneous without fatty components and no calcification or necrosis. The mass was high density (35 HU) before contrast and clearly homogeneously enhancing mass in arterial time.

Fine needle aspiration (FNA) was performed and the specimen revealed proliferation of lymphoid cells, part of them were small and had indistinct cytoplasm, round and inconspicuous nuclei.

The FNA was followed by immune chemical stains, which revealed positivity for CD3; as a result, it was diagnosed as T cell lymphoma. Unfortunately, the FNA slides are not available.

Our patient underwent a chemical course with (CHOP) for five months; her pain improved. On follow-up, ultrasound and CT demonstrated no change of the mass.

Unfortunately, after a year, our patient reported that her pain had recurred; the clinician decided that it is a recurrence of lymphoma, so another course of chemotherapy was determined. Nevertheless, the patient and her parents refused it because she was unable to tolerate the side effects of chemotherapy and they requested a consultation of another clinician, who reviewed the whole case and suspected the diagnosis.

Therefore, the decision was taken to perform an excisional biopsy of the mass. The pathology revealed vascular transformation of the lymph node with no malignancy (Figure 5).

The patient tolerated the procedure well and had uneventful course. At the time of writing, our patient is 22-year-old. She has been completely healthy.

DISCUSSION

Vascular transformation of sinuses (VTS) is a reactive process. The sinuses of lymph node were converted into a complex anastomosing network of vascular channels, which vary from capillaries to cavernous spaces [1, 3, 5–7].

Haferkamp et al. used the term vascular transformation of sinuses (VTS) in 1971 for the first time. He described the sinuses of lymph node, which resembled blood capillaries [1, 2, 5, 7].

Most of the VTS cases were diagnosed incidentally in excised lymph nodes during a variety of surgical procedures mostly for neoplastic lesions [1, 3, 4]. VTS can occur in any age group regardless of sex and in lymph nodes of any location [8].

Figure 1: (A, B) Abdominal ultrasound showing homogeneous mass located at right supra-median renal region behind inferior vena cava.

Figure 2: Abdominal axial multi-slice computed tomography scan at arterial phase, (A) Venous phase, (B) Showing a retro caval mass at the region of right adrenal gland.

Figure 3: (A) Abdominal axial multi-slice computed tomography reveals an oval, well-defined and homogeneous mass which is located above right renal region, no calcifications or necrosis, and (B) Abdominal coronal multi-slice computed tomography with contrast (arterial phase) reveals the above mass uptaking contrast homogeneously.
The frequency of VTS may be dependent upon the anatomic location of lymph nodes and determined in part by the mode of nodal blood supply [6].

Intra-abdominal lymph nodes are the most common to be affected by VTS, cervical being relatively rare [1, 2, 8]. Vascular thrombosis, severe congestive heart failure, constrictive pericarditis and previous operation or radiotherapy may also contribute to the pathogenesis of this condition [8].

The unique aspects of our case are: First, on review of literature, most of case reports have described VTS in adult patients, whereas we report a unique case of VTS in a child.

Secondly, it is known that the majority of VTS cases, which were reported, associated with neoplasia, in contrast to our patient who had VTS without any apparent cause or cancer.

Treatment is not indicated, but when vascular transformation is found, a search for occult cancer causing lymph node obstruction is warranted [2, 3, 5].

In 1995, Cook et al. reported 18 cases of VTS. In 15 of these cases, the lesion was identified in retroperitoneal lymph nodes which were removed as part of radical nephrectomies for renal cell carcinoma, all of the tumors were high histologic grade, and one case was exhibiting sarcomatoid features.

In these cases, there seems to be a pathogenetic relationship between the two entities, the changes in the lymph nodes probably are representing a reaction to the renal carcinoma. Renal cell carcinomas are known for their rich vasculature. It is thought that it is due to the production of angiogenic factors by the tumor cells. Theoretically, these angiogenic factors could drain into the regional lymph nodes and elicit the changes of VTS [9].

Thirdly, of rather interest of this case is the misdiagnosing followed by mismanagement. When we seriously doubt of non-Hodgkin lymphoma, there are many topics which must be considered before making decision of chemotherapy. These topics include the best type of biopsy for diagnosis, genetic testing, the role of fluorodeoxyglucose (FDG) positron emission tomography (PET) computed tomography (CT) scan in staging, patient information needs and survivorship but biopsy is an essential one. According to NICE guideline (National Institute for health and Care Excellence), it is considered that an excision biopsy as the first diagnostic procedure for people with suspected non-Hodgkin lymphoma at first presentation [10].

By contrast, our patient was diagnosed as lymphoma by performing an FNA not by an excisional biopsy; furthermore, she was administered with course of chemotherapy.

The reason for not performing an excisional biopsy was the high risk of a surgical procedure which outweighs the potential benefits of an excision biopsy. The retrocaval lymph node and its adjustment to the right adrenal gland need special instruments and experts for excisional biopsy; unfortunately, these instruments were not available in our country at that time.

Kaposi’s sarcoma and all vasoproliferative lesions of lymph node can be included in the differential diagnosis of VTS. Reactive vascular proliferations which include proliferation of high endothelial venules such as peripheral T cell lymphoma, Hodgkin’s disease and reactive paracortical hyperplasia are also included [1–5, 7–9].

From histological point of view, the spindle cells of VTS lack well-formed fascicles and are always accompanied by an appreciable number of irregular narrow vascular channels, the cellular areas of VTS display maturation into well-formed vascular channels toward the capsular aspect [1, 9].

Important limitations in understanding the misdiagnosis of VTS are the significant time lapse from the diagnosis to reporting of this case and unavailability of the FNA slides.
We can interpret the misdiagnosis of this rare condition because VTS is a structural change of lymph node sinuses, which needs a histological assessment to recognize. An FNA procedure provides cytological samples and no microarchitecture of lymph node is visualized.

In our case, the FNA cytology sample which was followed by immune chemical stains revealed positivity for CD3 which is restricted to the T cell lineage and occurs virtually throughout T cell differentiation.

CD3 is the most sensitive and specific marker of T cell lineage available for the immunophenotyping of lymph proliferative disorders. Consequently, the pathologist considered T cell lymphoma as the main diagnosis [11].

Actually, this is an important aspect of our case; in spite of rarity of VTS, which could be unthinkable; the misunderstanding of lymphoma approach leads to mismanagement of this case; hence, the whole path of this case was changed, which was ended by in giving chemotherapy to a healthy young girl, who suffered all its adverse effects.

CONCLUSION

Nevertheless, vascular transformation of sinuses (VTS) is incidentally found in surgical specimens and often associated with malignancy; doctors should consider VTS in the differential diagnosis especially when the clinical picture of malignancy is incomplete. Misdiagnosis of other conditions could occur when fine needle aspiration is performed instead of excisional biopsy to diagnosis non-Hodgkin lymphoma.

**********

Acknowledgements
We are thankful to Dr. Mohamed Imad Eddin Mouhandes (MD), Consulting Pathologist, Dr. Rama Faour (MD), University of Aleppo faculty of Medicine, Ms. Manal Mehio Sailam and Ms. Nour Ajam, University of Aleppo, faculty of Arts and Humanities, Aleppo, Syria, for their contribution.

Author Contributions
Luma Haj Kassem – 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

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

Khawla Mayoh – 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

Najat Mehio Sailam – 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 Luma Haj Kassem 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

10. National institute for health and care excellence. [Available at: www.nice.org.uk/guidance/ng52]
Spontaneous splenic rupture without trauma: A case report

Mürşit Dincer, Ahmet Kocakuşak, Gamze Çitlak, Ekrem Ferlengez, Muzaffer Akinci

ABSTRACT

Spontaneous splenic rupture is a rare ill-defined clinicopathological entity and occurs in only 1% of all splenic ruptures. It occurs usually as a result of splenic infiltration by infectious or hematological diseases. We present a case of 36-year-old female who was admitted to our emergency department with a three-hour history of acute onset abdominal pain and dyspnea. There was no history of trauma and infectious or hematological diseases. Considering the hemodynamic instability an emergent laparotomy was performed. During laparotomy, a 5-cm splenic laceration was found and a splenectomy was performed. The histology report confirmed that there was no pathological cause of splenic rupture.

Keywords: Acute abdomen, Atraumatic splenic rupture, Splenectomy

INTRODUCTION

Spontaneous splenic rupture is a life-threatening abdominal emergency. Spontaneous splenic rupture without a history of trauma is very uncommon [1]. It occurs in only 1% of all splenic ruptures. Atraumatic splenic rupture usually occurs due to infectious, hematological, or malignant infiltration of spleen [2]. Its etiology and management are unclear [3]. Nonetheless, emergency splenectomy is the standard treatment for patient with spontaneous splenic rupture [4].

CASE REPORT

A 36-year-old female was admitted to our emergency department with a three-hour history of acute onset abdominal pain and dyspnea. There was no history of trauma and infectious or hematological diseases. On examination, her pulse was 88/minutes with systolic blood pressure 85/60 mmHg, oxygen saturation of 95%. There was guarding over the left hypochondrium and epigastrium. Initial blood tests showed hemoglobin of 7.8 g/dl, hematocrit of 23.9% with normal coagulation profile. Abdominal sonography showed presence of free fluid around the spleen. Computed tomography scan confirmed the peri-splenic hematoma (Figure 1). The...
patient was monitored in our clinic. Intravenous fluids and empirical antibiotics were given. A total of two units of red blood cells were given. Repeat blood tests showed hemoglobin of 7.5 g/dl, hematocrit of 23.3%. Considering the hemodynamic instability and persistent pain, surgical intervention was decided. Laparotomy revealed 3000 cc of blood in the abdomen and a 5-cm splenic laceration were found. During the surgery a total of three units of red blood cells and three units of fresh frozen plasma were given. A splenectomy was performed. She was discharged with no problem on day-4. The histology report confirmed that there was no pathological cause of splenic rupture.

DISCUSSION

Spontaneous splenic rupture occurs mostly in a spleen due to infectious hematological or malignant infiltration. Spontaneous splenic rupture without a history of trauma is an uncommon entity. This serious clinical event which warrants immediate intervention is mostly in the form of operative surgery to save life [2, 5]. In spite of that, it has been reported in literature that a less invasive approach as proximal splenic artery embolization may be a safe, successful therapeutic alternative in selected patients [6].

CONCLUSION

In conclusion, spontaneous splenic rupture without a history of trauma is an uncommon life-threatening abdominal emergency. The pathogenesis of the disease remains unclear. In patients with atraumatic left hypochondrial pain and low hemoglobin, splenic rupture should be kept in mind.

*******

Author Contributions
Mürşit Dincer – 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
Ahmet Kocakuşak – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published
Gamze Çitlak – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published
Ekrem Ferlengez – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published
Muzaffer Akinci – 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 Mürşit Dincer 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
5. Mohammed AM, Majid ZI, Villatoro EA. Spontaneous rupture of the spleen as a result of primary splenic

A diagnostic dilemma: Sclerosing encapsulated peritonitis

Mürşit Dincer, Gamze Citlak, Zehra Zeynep Keklikkiran, Ahmet Kocakusak, Muzaffer Akinci

ABSTRACT

Sclerosing encapsulated peritonitis is a rare entity and it is interpreted as the total or partial encasement of the abdominal organs within a thick fibrocollagenous membrane. The underlying conditions of sclerosing encapsulated peritonitis are multifactorial. Most of the cases are unfortunately diagnosed at laparotomy. It may lead to diagnostic laparotomy because of the acute abdominal signs and symptoms. Adhesiolysis of the sac is enough for the surgical treatment of sclerosing encapsulated peritonitis, unless a non-vital intestinal segment is present which requires resection. Herein, we report a case of sclerosing encapsulated peritonitis with internal herniation findings detected in the preoperative abdominal computed tomography scan and signs of acute abdomen.

Keywords: Acute abdomen, Mechanical intestinal obstruction, Sclerosing encapsulated peritonitis

INTRODUCTION

Sclerosing encapsulated peritonitis is a rare benign cause of acute or subacute small bowel obstruction. It causes total or partial encasement of the abdominal organs within a thick fibrocollagenous membrane [1]. It may lead to diagnostic laparotomy because of acute abdominal signs and symptoms. Sclerosing encapsulated peritonitis may cause obstructive symptoms with negative laparotomies. Herein, we report a case of sclerosing encapsulated peritonitis with internal herniation findings detected in preoperative abdominal computed tomography scan and signs of acute abdomen.

CASE REPORT

A 60-year-old female patient was admitted to the emergency service and presented with abdominal pain. Symptoms such as total constipation, nausea and vomiting were present for three days. She had had similar history of these symptoms previously and she had been hospitalized because of those symptoms. At that time, laboratory and imaging studies revealed no pathological finding one year ago. She had history of hypertension. Abdominal examination revealed abdominal distension
and a palpable mass in her upper left quadrant. Laboratory studies were normal. Plain abdominal X-ray showed that there are air-fluid levels in small intestines (Figure 1). Abdominal CT revealed dilatation of small intestines and internal herniation (Figure 2). With these results explorative laparotomy was performed which showed a fixated membranous structure encapsulating all of the intraabdominal organs and not allowing to explore intraabdominal organs. The diagnosis was sclerosing encapsulated peritonitis according to explorative laparotomy. Adhesiotomy was partially applied. Physical examination was normal with spontaneous intestinal motility and normal defecation during the postoperative follow-up period. The patient was discharged from the hospital following uneventful course and appeared well in the outpatient visits.

DISCUSSION

Sclerosing encapsulated peritonitis is a rare clinical condition and its etiology is obscure. The underlying conditions of sclerosing encapsulated peritonitis are multifactorial [2]. It is classified into two categories as idiopathic and secondary. Generally, there are colicky abdominal pain, nausea, vomiting and sometimes a palpable mass in the midline of the abdomen. Preoperative diagnosis requires a high index of clinical suspicion. Generally, diagnosis of sclerosing encapsulated peritonitis is made with laparotomy [3, 4]. In the present case, there was a patient with abdominal pain, left upper quadrant mass and obstructive findings which resulted in explorative laparotomy. In the surgical treatment of sclerosing encapsulated peritonitis, adhesiolysis or adhesiotomy is enough, unless a nonvital intestinal segment is present which requires resection [5]. The retrospective evaluation of the same radiologist revealed a sac encapsulating intestines in the postoperative period. The preoperative mass was in fact the sac enveloping the intestines to the side.

CONCLUSION

In conclusion, sclerosing encapsulated peritonitis may cause acute abdominal signs and most cases are diagnosed at laparotomy. The preoperative diagnosis of this entity may be helpful for proper treatment of patients. Clinicians must rigorously pursue a preoperative diagnosis as it may prevent unnecessary laparotomies, since obstructive findings may be relieved with conservative treatment of sclerosing encapsulated peritonitis to decrease the incidence of negative laparotomies. A better awareness of this entity and the imaging techniques may facilitate preoperative diagnosis.

*********

Author Contributions

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

Gamze Citlak – Substantial contributions to conception and design, Acquisition of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published
Zehra Zeynep Keklikkiran – Substantial contributions to conception and design, Acquisition of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

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

Muzaffer Akinci – Substantial contributions to conception and design, Acquisition 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 Mürşit Dincer 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
CASE REPORT

Congenital epulis: A rare case report

Jaya Naidu, Shreya Banerjee, Sapna Jyoti, Pavanalakshmi GP, Kirthana Satish

ABSTRACT

Introduction: Congenital epulis, also known as congenital granular cell tumor, is a rare benign intraoral tumor which occurs along the gingiva of alveolar ridges of the jaws. It has a female predilection with a female and male ratio of 8:1. It usually occurs as a single mass, however, multiple epulis have also been reported. It has the potential to interfere with feeding and cause airway obstruction. Case Report: A two-year-old girl presented with the chief complaint of swelling in upper left back tooth region. The parent also reported bleeding from the swelling while brushing. The mass was soft in consistency with an irregular surface, approximately 3x2 cm in size, and caused vestibular obliteration. The parents reported that the mass had slowly increased in size from when they had first noticed it, nine months back. Based on the clinical appearance a provisional diagnosis of peripheral giant cell granuloma was given. Complete surgical excision of the mass under general anesthesia was performed and the specimen was sent for histopathology. Conclusion: Pedodontists may be consulted initially regarding such cases and should be aware of the potential for complications like feeding difficulties and airway compromise.

Keywords: Congenital epulis, Congenital granular cell lesion, Intraoral tumor, Neumann’s tumor

How to cite this article


Article ID: Z01201706CR10798JN

doi:10.5348/ijcri-201759-CR-10798

INTRODUCTION

Congenital epulis is a rare benign intra-oral tumor, which mostly occurs along the gingiva of the alveolar ridges of the newborn. Also known as Neumann’s tumor (Newman, 1871), it has varyingly been referred to as congenital gingival granular cell tumor (CGCT) of the
newborn, congenital granular cell lesion (CGCL) [1–3] and congenital myoblastoma (historically) [3]. Though numerous theories, which varyingly consider these lesions as embryonal hamartomas, or of fibroblastic, histiocytic, myogenic and neurogenic origin, have been postulated, the etiology and histogenesis of these lesions remains unclear [1, 3, 4]. Ultrastructural studies strongly support a mesenchymal histogenesis [1, 4], while a hormonal factor cannot be discounted considering the marked female predilection of the lesion [1, 5]. Although mostly occurring as single entities, multiple epulis have also been reported [1–3]. The maxilla is more commonly affected with a ratio of 2:1 [1–3]. Usually occurring as a nodular, sessile or pedunculated mass with a smooth, normal colored surface, that appears to arise from the alveolar ridge or process, it varies considerably in size from only a few millimeters in diameter to several centimeters [1–6]. Females show a greater predilection with a ratio of 8:1 [1, 3, 7, 8]. It has the potential to interfere with feeding, cause airway obstruction [1–3] and prevent adequate closure of the mouth [7]. No recurrence or metastasis has been reported in literature [1–3], while spontaneous regression of the lesion has been known to occur [5, 6]. It has been incurred that cases where the lesion has been reported to seemingly increase in size, inflammation and edema as a result of traumatic factors could be responsible [5]. The management of congenital epulis may involve conservative surgical excision if feeding or respiratory problems exist [1–3].

To date relatively few cases of congenital epulis have been reported. This case report describes the clinical features and management of an atypical case.

CASE REPORT

A two-year-old girl presented with a swelling in upper left back tooth region. The swelling was noticed by the child’s parent, nine months before, and was observed to be slowly increasing in size. A history of bleeding on brushing and a slight discomfort on mastication were also reported. The patient’s medical, dental and family histories were non-contributive. Extraoral examination revealed a slight left sided facial asymmetry. All deciduous teeth were present on intraoral inspection. A swelling in the maxillary left posterior segment, which appeared to be growing from the alveolar ridge, was observed. The swelling extended from the distal aspect of first primary molar to the first permanent molar region and from one centimeter above the gingival margin to the level of the occlusal plane and 0.5 cm palatally. Vestibular obliteration was present. The lesion was erythematous, with an irregular surface. On palpation, it was soft in consistency and appeared to be pedunculated (Figure 1).

Three-dimensional reconstructed computed tomography scan revealed a bilateral asymmetry along with displacement of the first permanent molar (Figure 2). Based on the clinical examination, a differential diagnosis of fibroma or peripheral giant cell granuloma was made.

Patient was referred to the Department of Oral and Maxillofacial surgery for complete surgical excision of the mass under general anesthesia. Complete excision of the lesion along with the removal of second primary molar and displaced first permanent molar was carried out (Figure 3). Post excision, the specimen was sent for histopathology (Figure 4). Intraoperative course and postoperative recovery of the patient was uneventful. The patient was prescribed perioperative antibiotics and postoperative analgesics. Oral feedings were commenced postoperatively and the patient was discharged from the hospital after four days of admission. Regular follow-up and review was advised.

Hematoxylin and eosin stained sections showed homogenized collagen fibers along with sheets of cells with granular cytoplasm and basophilic nuclei with prominent nucleoli. Plenty of blood vessels including capillaries,
venules and arterioles with RBC’s could be appreciated in the stroma. Hemorrhagic areas along with inflammatory cell infiltrate consisting of lymphocytes, neutrophils and plasma cell and dilated capillaries were also observed (Figure 5). Based on these findings, a histopathological diagnosis of congenital epulis was established.

**DISCUSSION**

Congenital epulis is usually diagnosed at birth and if the lesion is large, it may be diagnosed in utero by 3D ultrasound and MRI scan [5, 6]. In this case, the diagnosis was delayed as the tumor went unnoticed due to its small size. It usually presents as a pedunculated mass with a smooth surface and normal color. In the present case, it was atypical with an erythematous and irregular surface. Seen mainly in the alveolar maxillary process, lateral to the midline in the canine and lateral incisor region [3], it was observed in the maxillary posterior region in the present case. The treatment of choice for congenital epulis is surgical excision, if the lesion is interfering with feeding. If there is no interference with feeding, regular monitoring of the lesion has been advocated as case reports on spontaneous regression have been documented [5, 6]. As the lesion interfered with feeding in the present case, it was excised to minimize further complications.

Although additional congenital or underlying bony defects or dental anomalies are not usually present [3], in the present case an underlying bony defect was encountered along with displacement of the maxillary first permanent molar.

It is essential for pedodontists and pediatricians to familiarize themselves with the clinical differential diagnosis of growths in the oral cavities of newborns as the treatment modalities may vary. The differential diagnosis for congenital epulis includes granular cell tumor, fibroma, granuloma, rhabdomyosarcoma, chondrogenic and osteogenic sarcomas, hemangiomas and lymphangiomas [6, 7].

**CONCLUSION**

Pedodontists and pediatricians can be consulted initially in such cases, necessitating the need for awareness regarding the clinical features, differential diagnosis, treatment and management of congenital epulis. An atypical case of congenital epulis such as the one being reported also demonstrates that it is essential to keep in mind the clinical, radiographic and histological characteristics of the lesion and not just the epidemiological characteristics to establish the correct diagnosis.
Acknowledgements
We like to thank Dr (Col) Suresh Menon and Dr Srihari of the Department of Oral Maxillofacial Surgery, Dr Jayalakshmi K of the Department of Oral Pathology and Microbiology and the Department of Oral Medicine and Radiology for their support.

Author Contributions
Jaya Naidu – 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
Shreya Banerjee – 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
Sapna Jyoti – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published
Pavanalakshmi GP – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published
Kirthana Satish – 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 Jaya Naidu 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
CASE REPORT  PEER REVIEWED | OPEN ACCESS

Complex esophageal reconstruction after esophagogastrectomy with non-supercharged right colon interposition for the treatment of lye ingestion

Paige Finkelstein, Omar Picado, Elizabeth Paulus, Janeth Ng, Gabriel Ruiz, Danny Yakoub

ABSTRACT

Introduction: Lye injury to the esophagus often produces necrotic injury, but gastric injury is less common; subsequently, gastric pull-up surgery can be performed to reconstruct the esophagus. Herein, we present a case where the stomach was destroyed and had to be removed, forcing alternative reconstruction to be performed. Case Report: A 67-year-old male requiring total esophagogastrectomy after lye ingestion presented for reconstruction where gastric pull-up surgery was no longer a viable option. We performed a novel right-colonic interposition leaving the cecum in place, combined with a Roux-en-Y procedure to replace both the esophagus and stomach. The patient made a full recovery, experiencing few complications after the reconstructive surgery. Conclusion: Complex esophageal reconstruction using a non-supercharged colon bypass in the retrosternal route can be used in esophageal replacement in cases with esophagogastrectomy for lye ingestion or otherwise. This can be done with leaving the ileocecal valve in place.

Keywords: Caustic injury, Colon interposition, Colonic interposition, Esophagogastrectomy, Lye ingestion

How to cite this article

Article ID: Z01201706CR10799PF
doi:10.5348/ijcri-201760-CR-10799

INTRODUCTION

Lye injury to the esophagus is well recognized, producing rapid and extensive liquefaction necrosis due to active trans-cellular Na+ transport that is both time-dependent and concentration-dependent when above pH 11.5 [1]. Gastric injury from lye ingestion is less common, due to the inherent gastric acidity that counteracts the mostly alkaline pH of caustic agents [2]. The management of digestive caustic injuries is challenging; prevention of stricture formation on the stable patient is a primary goal, but often surgery is necessary if severe burns have been identified [3]. For esophagectomy, if the stomach is viable, gastric pull-up is preferred for reconstruction [4].

Colon interposition is another attractive treatment option for total esophageal replacement due to its length and functional capabilities; nonetheless, it has been accustomed to be attached to the stomach. Replacing
the esophagus with a segment of the colon was first recorded in 1911 [5], and since, it has become a standard reconstruction method for patients who have previously undergone esophagectomy [6]. Here, we report the utilization of colon interposition surgery for successful delayed reconstruction of excised necrotic esophagus and stomach following lye ingestion. A segment of the right and transverse colon was used without resecting the cecum, and colocolic anastomosis was done. The distal part of the colon conduit was anastomosed to a Roux-en-Y jejunal loop as the stomach had been previously excised due to severe burn.

CASE REPORT

A 67-year-old male was brought to the hospital via emergency medical services in mild distress with the inability to speak, sore throat, congestion, and hematemesis. Past medical history was unclear due to altered mental status. It was reported that the patient had ingested lye just prior to his arrival to the hospital.

Initial examination showed stable vital signs, mild confusion and a moderately-tender, non-distended abdomen, with normal intestinal sounds and no organomegaly; at this point he did not develop peritoneal signs yet. Initial fluid resuscitation was started, and proton pump inhibitors were given followed by steroids. Computed tomography scan of the head without contrast was negative for acute cerebrovascular accidents. Findings from X-ray, followed by CT scan of the chest (Figure 1) indicated a thickened esophagus consistent with esophagitis, and moderate left pleural fluid. Computed tomography scan of the abdomen and pelvis (Figure 2) without contrast revealed pneumatosis of the gastric wall, free intraperitoneal air and ascites consistent with suspected perforation.

About four hours after admission, the patient rapidly deteriorated and developed severe abdominal pain/rigidity, tachycardia, anuria, and hypotension despite adequate fluid resuscitation. The decision to operate was made, and exploration of the peritoneal cavity showed a necrotic stomach with evident necrosis of the distal esophagus up into the lower mediastinum. Furthermore, a large gastric perforation with full thickness necrosis of the entire body of the stomach was found, and there were large amounts of caustic fluid in the lesser sack. The whole gastric mucosa was grey and non-viable, with signs of ulceration and general sloughing. At this point, a total gastrectomy and transhiatal esophagectomy was performed with a diverting cervical esphagogastomy. The abdominal cavity was washed out with 13 L of normal saline. A feeding jejunostomy was inserted. The patient tolerated the procedure well and was admitted to the ICU.

The patient remained in the hospital for six months, during which he had multiple complications including pneumonia with pleural effusion and development of left upper quadrant colocutaneous fistula (resulting from the original caustic injury damaging a small area the colon), which eventually healed. He also had complications of malnutrition, where subsequently he recovered his nutritional status after months of appropriate enteral tube feeding. Finally, he was deemed ready for esophageal reconstruction. Preoperative preparation included mechanical bowel preparation and CT angiography of the abdomen to ensure integrity of mesenteric and pericolic vessels. Colonoscopy was done and showed no other abnormalities other than the site of the healed fistula by the splenic flexure.

At the time of the reconstruction procedure, the abdomen was opened and adhesions were noted and carefully divided. Inflammation was noted in the area of the splenic flexure as a result of the previous fistula that had developed there. Trials for mobilization of the
splenic flexure failed. The decision was made to use the right colon as the conduit to reconstruct the esophagus. First, the right colon was mobilized with identification and preservation of the right ureter. A Cattell–Braasch maneuver was performed, as well as a Kocher maneuver with division of adhesions between the transverse mesocolon and the inferior aspect of the pancreas to achieve full mobilization of the right and transverse colon. The middle colic vessels were identified at their origin from the superior mesenteric vessels just below the neck of the pancreas. The middle colic pedicle would provide the blood supply to the interpositioned colon; test bulldog clamps were placed on the ileocolic and right colic vessels (planned to be divided) to confirm ability of middle colic vessels to supply the whole length of conduit via the artery of Drummond. Umbilical tape was used to measure the length of the colon, and determine how much would need to be mobilized to bring it up into the neck. The esophagostomy and proximal cervical esophagus were then mobilized, and its edge trimmed to normal esophagus. Owing to the fact the posterior mediastinum was obliterated by fibrosis due to previous inflammation, a retrosternal tunnel was created from the neck above and hiatus below using blunt finger dissection. To widen the thoracic inlet allowing easy passage of the colon conduit, the left half of the manubrium sterni and the head of the left clavicle were removed with a bone saw, avoiding injury to the underlying vessels.

When the colon vessel clamps were rechecked, the colon remained pink and viable. The colon was divided just distal to the cecum at the beginning of the ascending colon. The ileocolic and right colic arteries were divided; the marginal artery was left intact. The proximal end of the right colon was then brought up to the neck in an isoperistaltic manner by placing the colon segment in a laparoscopic sleeve bag, guiding a Foley catheter through the retrosternal tunnel, securing the Foley into the bag containing the conduit then pulling it into the neck to avoid excessive handling of the conduit or its twisting along its longitudinal axis. The conduit was brought into the neck; pulse in the vessels of the conduit was demonstrated by Doppler examination of the mesocolon in the neck. The proximal esophageocolic anastomosis was performed using a stapled triangulated technique [7].

The transverse colon was then subsequently divided, taking care to preserve the marginal artery of Drummond. The middle colic vessels to supply the whole length of conduit via the artery of Drummond. Umbilical tape was used to measure the length of the conduit, and determine how much would need to be mobilized to bring it up into the neck. The previous jejunostomy feeding tube was disassembled.

When the colon vessel clamps were rechecked, the colon remained pink and viable. The colon was divided just distal to the cecum at the beginning of the ascending colon. The ileocolic and right colic arteries were divided; the marginal artery was left intact. The proximal end of the right colon was then brought up to the neck in an isoperistaltic manner by placing the colon segment in a laparoscopic sleeve bag, guiding a Foley catheter through the retrosternal tunnel, securing the Foley into the bag containing the conduit then pulling it into the neck to avoid excessive handling of the conduit or its twisting along its longitudinal axis. The conduit was brought into the neck; pulse in the vessels of the conduit was demonstrated by Doppler examination of the mesocolon in the neck. The proximal esophageocolic anastomosis was performed using a stapled triangulated technique [7].

The transverse colon was then subsequently divided, taking care to preserve the marginal artery of Drummond. The previous jejunostomy feeding tube was disassembled. A Roux-en-Y jejunostomy loop was brought up to the conduit to create the colojejunal, stapled anastomosis. A stapled colocolonic anastomosis was performed between the cecum and the distal transverse colon. Appendectomy was also performed to avoid potential future issues.

Finally, a new feeding jejunostomy tube was placed distal to the jejunoojejunojejunostomy anastomosis via Witzel technique [8] and secured to abdominal wall. Nasogastric (Nasocolic in this case) tube was left in place. Drains were placed in the neck and in the upper abdomen near the colo-jejunal anastomosis. The neck and abdomen were inspected to ensure adequate hemostasis and then closed in layers in the standard fashion. Overall, the patient tolerated the procedure well with no intraoperative complications. Operative time was 436 min, estimated blood loss was 200 cc.

The patient’s postoperative course was unexpectedly smooth without any major complications. DVT prophylaxis was started on day-2, oral diet was resumed on day-5 after a barium swallow confirmed absence of leakage. The patient experienced minor diarrhea which subsided, and was discharged home day-9. At 4th week postoperative, the feeding tube was removed as he had adequate oral intake. The patient was subsequently seen at 6th and 12th months with no reported dysphagia or other complications. Artistic rendition of the reconstructive surgery is seen in Figure 3.

Figure 3: The left image demonstrates the status of the gastrointestinal tract after esophagogastrectomy with spit and feeding jejunostomy, but before any reconstruction. The right image demonstrates the gastrointestinal tract after reconstruction with reversed colon bypass and Roux-en-Y colojejunojejunostomy with feeding jejunostomy.

DISCUSSION

To avoid post-surgical reconstruction stenosis of the esophagus, it is recommended to delay the date of surgery up to three months in the absence of pharyngeal injury and up to six months in case of pharyngeal injury [9]. Additionally, the selection of right versus left colon for interposition has been controversial [10]. The choice of right colon versus left colon usually depends on the preference of the surgical team [6]. Right colic interposition benefits include prevention of pharyngeal reflux with preservation of the ileocecal valve, congruence of the esophagus and the ileum at the cervical level, ease of anastomosis, and ability to use the left colon for rescue.
CONCLUSION

Complex esophageal reconstruction using a non-supercharged colon bypass in the retrosternal route can be used in esophageal replacement in cases with esophagogastronomy for lye ingestion or otherwise. This can be done with leaving the ileocecal valve in place.

*********

Author Contributions

Paige Finkelstein – 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

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

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

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

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

Danny Yakoub – 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 Paige Finkelstein 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


Reversible stomatocytosis

Akanksha Agrawal, Deepanshu Jain, Mitchell Goldstein

CASE REPORT

Stomatocytosis is a rare morphological condition of the red blood cells in which the normal central zone of pallor is replaced by a mouth-like or slit-like pattern. Stomatocytosis can be either hereditary or acquired in the setting of acute alcohol consumption, chronic liver disease [1]. It may be asymptomatic or lead to hemolysis presenting as anemia. We present a case of a young lady with chronic alcoholism who presented with symptomatic anemia. Alcoholism is known to cause hypophosphatemia [2], which can cause stomatocytosis. Repletion of phosphorus and abstinence of alcohol improved the patient’s anemia, improving the stomatocytosis on the peripheral smear. The use of an easily available, rapid and cheap test like peripheral smear in guiding the therapy in such a patient reflects the importance of peripheral smear and its cost-effectiveness.

A 38-year-old African-American female with history of chronic pancreatitis, cholelithiasis, diabetes mellitus and alcohol abuse presented with severe anemia and light-headedness. She was consuming about 80 grams of alcohol on a daily basis. On admission, she had hemoglobin of 5.6 g/dL, mean corpuscular volume (MCV) 90 fl (normal), iron level 92 µg/dL (normal) and ferritin 1188 ng/mL. Her vitamin B12 and folate levels were normal and reticulocyte count corrected for anemia was 1.2%. Her phosphorus was 1.0 mg/dL (critically low). Abdominal ultrasound was consistent with hepatic steatosis and distended gallbladder with possible gallstones. On peripheral smear, she had stomatocytosis (Figure 1A). Numerous RBCs were described with fish mouth appearance, lacking the typical central pallor. After three days of phosphorus repletion, the phosphorus level improved to 2.7 mg/dL and peripheral smear showed marked improvement with negligible stomatocytes (Figure 1B). Patient’s anemia improved significantly. She was counseled to abstain from alcohol use, and discharged home with adequate follow-up.

DISCUSSION

Stomatocytosis can be hereditary or acquired secondary to medications (vincristine, vinblastine, chlorpromazine, etc.), acute alcohol intoxication or chronic liver disease [1, 3, 4]. Our patient had hypophosphatemia due to alcohol ingestion, which further led to stomatocytosis of the blood cells. In a study by Rauchenzauner et al., 3.4% of patients with acute alcohol intoxication had hypophosphatemia [5]. This results from decreased phosphate absorption and increased urinary phosphate excretion. In addition, chronic alcoholism can induce cellular phosphate depletion [6]. A study done in

Figure 1: The peripheral smear on the left showing stomatocytes in the setting of hypophosphatemia due to chronic alcohol consumption. The subsequent smear on the right side reflects the improvement in the blood picture with almost no stomatocytes after phosphorus repletion.
1979 by Wisloff et al. studied the peripheral blood smears in 100 alcoholic patients. They reported 15% patients manifesting marked stomatocytosis [1]. Low serum phosphorus level depletes the ATP level in RBC affecting the RBC pliability. The complex metabolic sequelae of hypophosphatemia also include 2,3-diphosphoglyceric acid depletion, a shift to the left in the oxygen dissociation curve, decreased glucose utilization, and increased lactate production, resulting in rigid and non-yielding RBCs.

In the review of recent literature, not many studies have commented on the association of hypophosphatemia with stomatocytosis. This case illustrates the presence of hypophosphatemia with chronic alcohol use and its effect on peripheral smear in the form of stomatocytosis. It also reports the prompt improvement of stomatocytosis with repletion of phosphorus. Peripheral smear is an easily available, rapid and a cheap test. The presence of a rare finding in a common scenario like chronic alcohol consumption highlights the clinical importance of peripheral smear.

CONCLUSION

This case depicts the reflection of significant laboratory value on the peripheral smear in a patient with severe hypophosphatemia in the setting of chronic alcohol consumption leading to stomatocytosis. This rare occurrence in a common clinical scenario highlights the importance of peripheral smear as a clinical tool.

Keywords: Alcoholism, Phosphorus, Stomatocytosis

REFERENCES

CASE REPORT

An 80-year-old female with past medical history of hypertension, hyperlipidemia, diabetes mellitus type 2, and history of remote smoking presented to her primary care doctor for evaluation of ten days history of cough. She did not report any chest pain, shortness of breath, or fever. Her physical examination was unremarkable. Two view chest X-ray revealed a clear lung parenchyma with normal heart size, calcific thoracic aorta, and an interesting incidental finding of possible splenic artery aneurysm (Figures 1 and 2). The patient was asymptomatic; she denied left upper quadrant pain, nausea, or vomiting. Computed tomography (CT) scan of the abdomen demonstrated significant atherosclerotic changes of the aorta without aneurysmal dilation, normal renal and mesenteric arteries, a partially calcified splenic artery aneurysm measuring maximum diameter of 2.0 cm, and there is also an area of the splenic artery ectasia where the vessel diameter reaches 0.8 cm (Figures 3 and 4). After a detailed discussion with vascular surgery and interventional cardiology; the multidisciplinary team decided to monitor the patient every six months. In event of aneurysmal enlargement more than 2.5 cm, the patient will be a candidate for elective splenic artery aneurysm procedure.

DISCUSSION

Splenic artery aneurysm is common visceral aneurysm; it is the third most common aneurysm after abdominal aortic and iliac aneurysms. Most of the time patients are asymptomatic, found incidentally during routine investigations. It can cause left upper quadrant abdominal pain, nausea, and vomiting. In rare cases, it could rupture and leads to shock [1].

Risk factors include hypertension, atherosclerosis, and iatrogenic during abdominal surgeries. There is no consensus when to operate, but it has been suggested that pregnant women, symptomatic patients should undergo repair procedure [2]. It is often difficult to correctly diagnose splenic artery aneurysm in asymptomatic patients who present with only chest X-ray or abdominal ultrasound; differential diagnosis included pancreatic...
cyst, pancreatic tumor, calcified left adrenal hematoma. Computed tomography and or magnetic resonance angiography can easily recognize the splenic artery aneurysm [3]. There is no specific guidelines on how to treat asymptomatic splenic artery aneurysm, though it is widely accepted to evaluate any aneurysm size more than 2.0 cm. for possible intervention. Treatment modality can be either endovascular or surgical; percutaneous intervention with either coil embolization or covered stent have been associated with a better short-term results compared to open repair, while open repair has less long-term complications including less re-exploratory procedures. The main indications for either types of the intervention are symptomatic patients, pregnancy, and pseudoaneurysm. Splenic artery anatomy is the main factor to decide which procedural approach the doctors should take [4, 5]. Timing for repair in asymptomatic patients is not standardized, but asymptomatic aneurysm of size 1–2 cm can be monitored safely every six months with an imaging method [6].

CONCLUSION

Splenic artery aneurysm is the third common abdominal aneurysm yet it is still rare, we present this nice chest X-ray. Splenic artery aneurysm is not just a radiographic diagnosis alone; it requires confirmation with other either computed tomography angiography and/or Doppler ultrasound. In this case, we will do a follow-up computed tomography scan in six months.

Keywords: Aneurysm, Asymptomatic, Imaging, Spleen, Splenic artery

Figure 2: Lateral view chest X-ray showing clear lung parenchyma.

Figure 3: Computed tomography angiography scan coronal plane. The arrow indicates the calcific splenic artery.

Figure 4: Computed tomography angiography, axial plane. The arrow indicates the calcific splenic artery.

How to cite this article


Article ID: Z01201706CL10125SA

doi:10.5348/ijcri-201715-CL-10125
Author Contributions
Sayf Altabaqchali – 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
Mohanad Hasan – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published
Ahmed Altabaqchali – 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 Sayf Altabaqchali 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

Access full text article on other devices

Access PDF of article on other devices
Kienböck’s disease mimicking gouty monoarthritis of the wrist

Ingo Schmidt

To the Editor,

A 57-year-old male presented with increasing pain and swelling in his left wrist over a period of two years. There was no history of any trauma, and additionally, a systemic inflammatory disease was unknown to the patient. On examination, there were no inflammatory clinical signs locally, and all (non-)specific serum inflammatory markers (including uric acid) were not increased. With the use of radiographs and computed tomography (CT) scans, an advanced stage of Kienböck’s disease with peripheral stress fractures of the lunate bone and secondary arthritic signs of articular surface in the lunate fossa was primarily diagnosed (Figure 1A), that was confirmed by the radiologist as stage IIIB in magnetic resonance imaging. The motion-preserving total wrist arthroplasty (TWA) using the relatively new angle-stable Maestro™ Wrist Reconstructive System (WRS, Biomet, Warsaw, Indiana / USA) was indicated. Intraoperatively, there were multiple calcification deposits on and around the lunate, associated with the primarily diagnosed peripheral ‘stress fractures’ of the lunate (Figure 1B). Gouty monoarthritis of the wrist involving the lunate was confirmed on histological examination by the pathologist. Two years postoperatively, there was unchanged correct positioning of TWA without any signs of loosening or subsidence (Figure 1C). Pain in visual analogue score (0–10 points) and function in patient-rated wrist evaluation score (0–100 points) with 8 and 73 preoperatively had improved to 2 and 19. The patient reported that he would have the same procedure again if it would be necessary.

Kienböck’s disease is defined as avascular osteonecrosis of part or all of the lunate, which progresses through several stages if not treated or treated with preservation of the lunate [1, 2]. It was first described in 1843 by Peste [3]; and in 1910, Kienböck [4] was the first who recommended to use the term lunatomalacia, and correctly hypothesized that the disease was precipitated by an interruption of the nutrition of the bone as a result of a traumatic insult. However, the etiology is still widely unknown, and many risk factors are discussed such as anatomical features (shape of lunate or distal radius, difference between distal end of radius and ulna, coverage of lunate by radius, arterial factors) and/or repetitive microtrauma potentially leading to subchondral stress fracture [1, 5]. Lichtman et al. [6] introduced in 1977 a modified classification with four broad stages which remains the most commonly used today, and stage IIIB, such as in our primarily suggested diagnosis in case presentation, is associated with lunate collapse and fixed scaphoid rotation. The secondary effects of the collapsing lunate are ‘compromised’ wrists including deformity and collapse of the central column, degeneration of the perilunate articulations, proximal row instability, and degeneration of the radial column [5]. For surgical treatment of stage IIIB, the preferred procedures are proximal row carpectomy with a portion ranging from 42–62%, followed by intercarpal fusions with a portion ranging from 11–19%, total wrist fusion with a portion ranging from 2–16%, radial shortening osteotomy with a portion of 12%, vascularized bone grafting with a portion of 6%, trial of splinting with a portion of 4%, and lunate arthroplasty with a portion of 1% [7, 8]. An option for surgical treatment of stage IIIA (lunate collapse without scaphoid rotation) is callotasis lengthening of the capitate bone [9]. Another option for treatment of advanced stage of Kienböck’s disease is motion-preserving TWA (Figure 2A–B) with a portion of 2% of all TWAs performed by surgeons who have published their experiences with this procedure [10], and the relatively new angle-stable Maestro™ WRS that was used in our case presentation,
is one of the modern biaxial-anatomical third generation type that is currently in use [11–14].

Gout (i.e., hyperuricemia) is a systemic disease often firstly presented as a monoarthritis. Its prevalence in western population was reported to be 1.4% in the 90th, associated with a overall male-to-female ratio of 3.6:1, and the prevalence peaked in men between the age of 75 and 84 years (7.3%), while in women its prevalence continued to rise beyond the age of 85 years (being about 2.8%) [15, 16]. Gouty monoarthritis, caused by crystal induced synovitis, is usually associated with agonizing pain, swelling, erythema, warmth, tenderness of the affected joint, fever and increased (non-)specific serum inflammatory markers [17], however, these local and/or systemic inflammatory clinical signs were not all present in our case presentation preoperatively. Such as in our case presentation, it can be a diagnostic challenge in the wrist if gout is unknown in the history and only pain and swelling are present preoperatively [17]. When gouty arthritis of wrist and hand is primarily diagnosed, 5% of patients may not respond to medical treatment and surgical treatment become necessary [16].

In literature, it has been reported only in few cases that Kienböck's disease can mimic a giant cell tumor of the lunate preoperatively [18], and the preoperative finding of scapholunate ligament disruption with or without erosions of carpal bones also can mimic a gouty monoarthritis of the wrist [19–21]. Additionally, first presentation of gouty arthritis of the wrist can be in 0.6% of cases a carpal tunnel syndrome [22].

**Keywords:** Gouty monoarthritis wrist, Kienböck's disease, Total wrist arthroplasty

---

**Figure 1 (Case Presentation):**

(A) Posteroanterior radiograph and sagittal CT scan demonstrating primarily suggested advanced stage of Kienböck's disease with bony destruction, peripheral stress fractures and collapse of the lunate associated with secondary arthritic signs of the articular surface in the lunate fossa, (B) Clinical photograph intraoperatively showing synovial calcification deposits on and around the lunate (white arrow), and fracture of lunate (blue arrow), (C) Posteroanterior and lateral radiographs two years postoperatively showing correct positioning and alignment of TWA without any signs of loosening or subsidence.

---

**Figure 2:**

(B) Example for Kienböck's disease stage IIIB with a collapsing lunate in a 54-year-old woman (left wrist), (B) Same patient, treated with another third generation TWA (RE-MOTION® Total Wrist).
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

ABOUT THE AUTHOR


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.