International Journal of Surgery Case Reports 3 (2012) 343–345

International Journal of Surgery Case Reports

journal homepage: www.elsevier.com/locate/ijscr

Mesentery lymphoma in a patient with Crohn's disease: An extremely rare entity

Nikolaos Koronakis[a], Emmanouil Lagoudianakis[b], Dimitrios Keramidaris[a], Apostolos Pappas[c], George Gemenetzis[b], Charalampos Seretis[b,*], John Chrysikos[a], Andreas Manouras[c]

[a] Second Department of Surgery, 417 NIMTS – Nosileutiko Idrima Metohikou Tameiou Stratou (Military Veterans' Fund Hospital), Athens, Greece
[b] Second Department of Surgery, 401 General Army Hospital, Athens, Greece
[c] First Department of Propaedeutic Surgery, Hippokrateion Hospital, Athens Medical School, University of Athens, Q. Sofias 114 Avenue, 11527 Athens, Greece

ARTICLE INFO

Article history:
Received 9 March 2012
Received in revised form 19 March 2012
Accepted 6 April 2012
Available online 16 April 2012

Keywords:
Inflammatory bowel disease
Lymphoma
Immunomodulators
Crohn's disease
Mesentery

ABSTRACT

INTRODUCTION: Lymphoma is a rare complication of long-standing Crohn's disease. We report a rare case of a diffuse, B-cell non-Hodgkin's lymphoma of the mesentery in a patient receiving treatment for Crohn's disease.
PRESENTATION OF CASE: A 52 year-old patient presented with abdominal pain, anorexia and postprandial fullness. Abdominal examination revealed a firm mass, extending from the epigastrium to the right iliac fossa. CT scan showed a large intra-abdominal mass with air-fluid levels within, and soft tissue density along its walls, surrounded by distended bowel loops. The patient was scheduled for surgery due to clinical assumption of an intra-abdominal abscess. At laparotomy an ill-defined, lobulated mass with cystic areas was noted rising from the mesentery. Frozen section biopsy of the cystic mass revealed a non-Hodgkin follicle center B-cell lymphoma of the mesentery.
DISCUSSION: To the best of our knowledge, this is an extremely rare case of lymphoma development in the mesentery, in a patient receiving treatment for Crohn's disease. Although the development of abdominal lymphomas can be justified as a possible consequence of the chronic immune-modulating therapy, their location can lead to diagnostic pitfalls.
CONCLUSION: Although mesentery has scarcely been presented as a potential site of occurrence of abdominal lymphomas in the process of treatment of inflammatory bowel diseases, this rare entity should be considered in the differential diagnosis of intra-abdominal lymphomas in patients with inflammatory bowel disease. In cases where imaging techniques do not provide definitive answers, surgical intervention can safely pose the accurate diagnosis.

1. Introduction

Crohn's disease (CD) is an idiopathic, chronic, inflammatory disease of the gastrointestinal tract that primarily affects the small intestine and colon. A number of studies have reported an increased risk of lymphoma in individuals with CD, while most of the described cases have arisen from the gastrointestinal tract.[1] Various hypotheses have been proposed to explain the increased risk of lymphoma in patients with Crohn's disease, nevertheless up to date this relationship remains unclear.[2] We report an extremely rare case of a patient with CD that was complicated with a non-Hodgkin lymphoma of the mesentery.

2. Presentation of case

A 52-year-old male presented with mild, vague abdominal pain followed by anorexia and postprandial fullness of 1-week duration. Two years before, the patient was diagnosed with Crohn's disease, with the final diagnosis of CD being posed after histopathological examination of multiple intestinal biopsies, which were obtained during two sessions of lower GI endoscopies. The patient was submitted to treatment with mesalamine and prednisolone, demonstrating evident response to the therapy. At the time of admission, clinical examination showed a moderately malnourished individual, who was afebrile and in good general condition. Abdominal examination revealed a firm, and slightly tender mass, extending from the epigastrium to the right iliac fossa. Digital rectal examination was normal. Laboratory tests revealed a white blood cell count (WBC) count of 10.400/mm^3 with 85% granulocytes, hemoglobin of 10.5 g/dl, platelet count of 421,000/mm^3, creatinine level of 0.6 mg/dl and urea of 40 mg/dl, serum albumin level of 3.3 g/dl and C-reactive protein of 7.27 mg/dl. Serum levels of CA 19-9, AFP and CEA were within normal range. The patient was admitted in our department, where he was treated with intravenous steroids (prednisolone 50 mg/day) and antibiotics (Ciprofloxacin 400 mg/12 h) and oral mesalamine (4.8 mg/day). Small intestine radiography revealed a large mass dislodging and compressing the bowel toward the superior quadrants of the abdomen (Fig. 1). A computed tomography (CT) scan was also

* Corresponding author. Address: 95 Lykourgou Str, Kallithea, 17676 Athens, Greece. Tel.: +30 6937236655.
E-mail address: babismed@gmail.com (C. Seretis).

2210-2612
http://dx.doi.org/10.1016/j.ijscr.2012.04.005

Fig. 1. Small bowel radiography revealed a large intra-abdominal mass causing external compression and displacement of the small bowel loops.

performed and demonstrated a large intra-abdominal mass with air-fluid levels within, and soft tissue density along its walls surrounded by distended bowel loops (Fig. 2). The mass was thought to be an intra-abdominal abscess and considering the current clinical setting the patient was scheduled for surgery. At laparotomy, an ill-defined, lobulated mass with cystic areas was noted to arise from the mesentery. An incisional biopsy was taken from the mass and a drain was inserted, while no further surgical manipulations were deemed necessary. Due to the development of a chylous fistula in the 2nd postoperative day, with 600 ml of chylous malignant

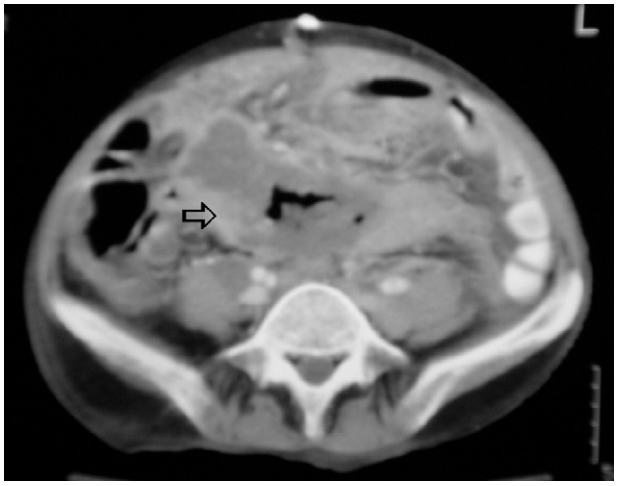

Fig. 2. Computed tomography (CT) scan shows a large intra-abdominal mass with air-fluid levels within, and soft tissue density along its walls surrounded by distended bowel loops.

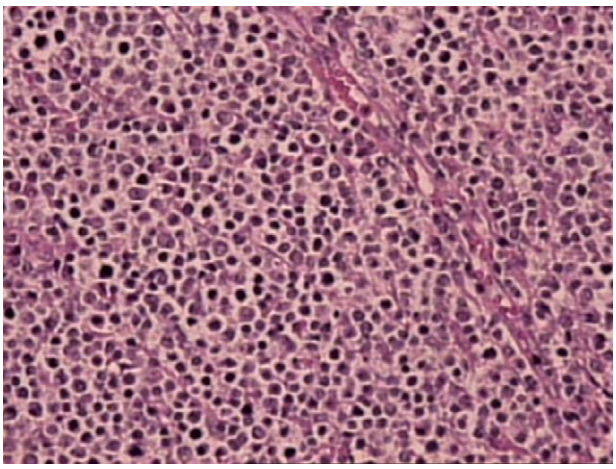

Fig. 3. Diffuse non-Hodgkin lymphoma of small and middle-sized lymphoid B-cells (hematoxylin-eosin staining 40×).

effusion draining daily, the patient also received parenteral nutrition and octreotide subcutaneously. During the following days the patient's clinical condition gradually deteriorated due to a respiratory infection and he finally succumbed to sepsis on the 14th postoperative day. Frozen section biopsy of the cystic mass showed histological features diagnostic of B non-Hodgkin follicle center cell lymphoma (Fig. 3).

3. Discussion

Lymphoma is a type of cancer involving cells of the immune system and is morphologically divided into two major categories: non-Hodgkin and Hodgkin lymphoma. Considering that the same cells are involved in the pathogenesis of autoimmune disorders, many researchers have argued upon the relationship between autoimmunity and lymphoid malignancies.[3] Driven by this notion, several studies have demonstrated an increased risk of lymphoma in patients with Sjogren's syndrome and rheumatoid arthritis.[4,5] Concerning patients with inflammatory bowel disease (IBD), studies on the incidence of lymphoma have been contradictory.[6,7] Nevertheless, a recent meta-analysis by von Roon et al. demonstrated an increased risk of lymphoma in patients with Crohn's disease.[8] Whether this finding is the result of the severity and the duration of the inflammatory processes or an effect of the immunomodulatory therapies or a combination of both, is still unknown.[9,10] A study by Kandiel et al. showed an approximate fourfold increased risk of lymphoma in IBD patients treated with azathioprine and 6-MP.[11] However, since the data used in this study were obtained from observational studies only, the authors could not exclude the possibility that the increased risk could also be associated with the severity of the disease. Furthermore, a population-based study by Bernstein et al. showed an increased risk of lymphoma for males with CD who had not received immunosuppressive therapy other than corticosteroids.[12]

After taking under consideration the aforementioned data, it is justified to consider that the deregulated interactions between the immune system and the luminal bacteria or their products, which are implicated in the pathogenesis of CD, may have contributed to the development of lymphoma.

We herein presented an extremely rare case of CD complicated with lymphoma of the mesentery. The combination of extra-intestinal lymphoma arising from the mesentery in the physical course of IBD is a fairly uncommon entity, with just one similar case presented in the literature up to date.[13] In this case, the clinical features of the patient, the area in which the mass was located and the

presence of air-fluid levels in the imaging studies mislead us and the patient was ultimately led to the operating room with the diagnosis of intra-abdominal abscess. This misjudgment can be attributed to the following: (a) intra-abdominal abscesses are common complications of CD, with CT scans being characterized by the presence of an expansive soft-tissue lesion with a central fluid collection and a thickened wall[14,15] and (b) on the other hand, lymphomas are rare complications, which usually present as shaped or irregular soft-tissue opacities rising from the gastrointestinal lumen.[16]

4. Conclusion

Extra-intestinal lymphomas are a rare complication of CD. The mesentery has scarcely been reported up to date as a site of development of lymphoma in the course of IBDs. Despite the rarity of these malignances, physicians must be alert for their existence due to the increased difficulty of diagnosis and treatment. It remains unclear whether this complication is the result of the severity and the duration of the disease or/and the use of immunomodulatory agents. It is evident that larger studies are needed in order to understand the complex relationships between inflammatory bowel diseases and lymphomas.

Conflict of interest

The authors declare that they have no competing interests.

Funding

None.

Ethical approval

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contributions

N.K., E.L., D.K., A.P. and C.S. were involved in drafting the manuscript. G.G., J.C. and A.M. were involved in collecting imaging and histopathological material, reviewing the literature and critically revising the manuscript. All authors read and approved the final paper.

Acknowledgments

Special thanks to Vilaras Georgios, Karameris Andreas, Mourouti Georgia, Pathology Department, 417 N.I.M.T.S.

References

1. Greenstein AJ, Gennuso R, Sachar DB, Heimann T, Smith H, Janowitz HD, et al. Extraintestinal cancers in inflammatory bowel disease. *Cancer* 1985;**56**:2914–21.
2. Binder V. Clinical epidemiology—how important now? *Gut* 2005;**54**:574–5.
3. Smedby KE, Askling J, Mariette X, Baecklund E. Autoimmune and inflammatory disorders and risk of malignant lymphomas—an update. *Journal of Internal Medicine* 2008;**264**:514–27.
4. Ekström Smedby K, Vajdic CM, Falster M, Engels EA, Martínez-Maza O, Turner J, et al. Autoimmune disorders and risk of non-Hodgkin lymphoma subtypes: a pooled analysis within the InterLymph Consortium. *Blood* 2008;**111**: 4029–38.
5. Hansen A, Lipsky PE, Dorner T. B-cell lymphoprolipheration in chronic inflammatory rheumatic diseases. *Nature Clinical Practice Rheumatology* 2007;**3**: 561–9.
6. Aithal GP, Mansfield JC. Review article: the risk of lymphoma associated with inflammatory bowel disease and immunosuppressive treatment. *Alimentary Pharmacology and Therapeutics* 2001;**15**:1101–8.
7. Askling J, Brandt L, Lapidus A, Karlén P, Björkholm M, Löfberg R, et al. Risk of haematopoeitic cancer in patients with inflammatory bowel disease. *Gut* 2005;**54**:617–22.
8. Von Roon AC, Reese G, Teare J, Constantinides V, Darzi AW, Tekkis PP. The risk of cancer in patients with Crohn's disease. *Diseases of the Colon and Rectum* 2007;**50**:839–55.
9. Jones JL, Loftus Jr EV. Lymphoma risk in inflammatory bowel disease: is it the disease or its treatment? *Inflammatory Bowel Diseases* 2007;**13**:1299–307.
10. Kwon JH, Farrell RJ. The risk of lymphoma in the treatment of inflammatory bowel disease with immunosuppressive agents. *Critical Reviews in Oncology/Hematology* 2005;**56**:169–78.
11. Kandiel A, Fraser AG, Korelitz BI, Brensinger C, Lewis JD. Increased risk of lymphoma among inflammatory bowel disease patients treated with azathioprine and 6-mercaptopurine. *Gut* 2005;**54**:1121–5.
12. Bernstein CN, Blanchard JF, Kliewer E, Wajda A. Cancer risk in patients with inflammatory bowel disease: a population-based study. *Cancer* 2001;**91**:854–62.
13. Charlotte F, Sriha B, Mansour G, Gabarre J. An unusual case associating ileal Crohn's disease and diffuse large B-cell lymphoma of an adjacent mesenteric lymph node. *Archives of Pathology and Laboratory Medicine* 1998;**122**:559–61.
14. Joseph AE. Imaging of abdominal abscesses. *British Medical Journal (Clinical Research Ed)* 1985;**291**:1446–7.
15. Cybulsky IJ, Tam P. Intra abdominal abscess in Crohn's disease. *American Surgeon* 1990;**56**:678–82.
16. Yenarkarn P, Thoeni RF, Hanks D. Case 107: lymphoma of the mesentery. *Radiology* 2007;**242**:628–31.