TURKISH JOURNAL OF INTERNAL MEDICINE

ISSN:2687-4245

Case Report

Persistent Stomach Pain in the Young Age Patient: A Case of Primary Gastric Burkitt's Lymphoma

Hatice HAMARAT[1], Berrin YALINBAS KAYA[2], Aktug SIMSEK[3]

[1]Department of Internal Medicine, Eskisehir City Hospital, Eskisehir, Turkey
[2]Department of Gastrointestinal Disease, Eskisehir City Hospital, Eskisehir, Turkey
[3]Department of Pathology, Eskisehir City Hospital, Eskisehir, Turkey

ABSTRACT

Lymphoma is the second most common gastric cancer, following gastric adenocarcinoma. Most gastric lymphomas are mucosa-associated lymphoid tissue lymphomas or diffuse large B-cell lymphomas. Primary gastric Burkitt's lymphoma is a subtype of non-Hodgkin lymphoma and represents an aggressive and rare malignity with the fewest cases reported globally. Primary gastrointestinal non-Hodgkin lymphoma is a rare condition. Burkitt's lymphoma is an aggressive form of B-cell lymphoma endemic to Africa while it is not endemic to the rest of the world. Here we presented a young immunocompetent male patient who had weight loss and was admitted with a stomachache. Upper gastrointestinal endoscopy and biopsy detected a large primary gastric Burkitt's lymphoma. While long-term survival rates are possible with early diagnosis and timely appropriate treatment, delay in treatment can be fatal for such patients.

Turk J Int Med 2022;4(4):195-199
DOI: 10.46310/tjim.1073581

Keywords: *Burkitt's lymphoma, stomach ache.*

Introduction

Highly malignant Burkitt's lymphoma derived from B-cells was first described by Dennis Burkitt in 1958 in a Ugandan child's jaw. Burkitt's lymphoma is a B-cell non-Hodgkin lymphoma (NHL) with a high proliferation rate. After six years, Epstein-Barr virus (EBV), a gamma herpes virus, was isolated from cultured Burkitt's lymphoma cells.[1] This tumour is mainly observed in patients in Sub-Saharan Africa.[1] It is associated with the t (8;14) (q24;q32) translocation of C-myc and IgH genes; IgH-myc fusion is characteristic.[2] Sporadic forms are rarely diagnosed in medical centres in Europe and Asia, with 4 to 5 cases annually.[3] Although Burkitt's lymphoma is regarded as a nodal lymphoma, extranodal involvement is present in 80% of the cases.[3]

Received: January 14, 2021; Accepted: June 08, 2022; Early Online: July 29, 2022; Published Online: October 29, 2022

Address for Correspondence:
Hatice Hamarat, MD

Department of Internal Medicine, Bursa Kestel State Hospital, Bursa, Turkey
E-mail: hklncal@hotmail.com

Extranodal Burkitt's lymphoma is especially common in gastrointestinal, head, and neck areas. Also, though rare, bone marrow, genitourinary system, bone, central nervous system and liver involvement have been reported.[3,4] It is a rare condition in adults. Overall survival is less than five months in more than 60% of the cases; among the negative prognostic factors are bone marrow and central nervous system involvement and late diagnosis.[3] Regarding primary sporadic Burkitt's lymphoma of the stomach, few cases with the secondary spread from retroperitoneal Burkitt's lymphoma are diagnosed due to admittance with symptoms related to gastric cancer. However, to this day, less than 200 papers have been published on gastric Burkitt's lymphoma. In this paper, we are presenting an extraordinary case of aggressive Burkitt's lymphoma.

Case Report

A 19-year-old male patient presented with a stomachache, involuntary weight loss, and persistent stomach pain history in the last three months. The patient had no associated comorbidity, family history, drug use, smoking, alcohol or drug habit. In the physical examination, the epigastric region was stiff and tender during the abdominal examination. There was not any lymphadenopathy, hepatosplenomegaly or palpable mass. The biochemistry and complete blood count of the patient were normal. He had no anemia. EBV DNA or HIV RNA tests were not detected in the patient. The patient was initiated on proton pump inhibitor and anti-acid treatments. However, the patient's complaints were not subdued. In endoscopy of the upper

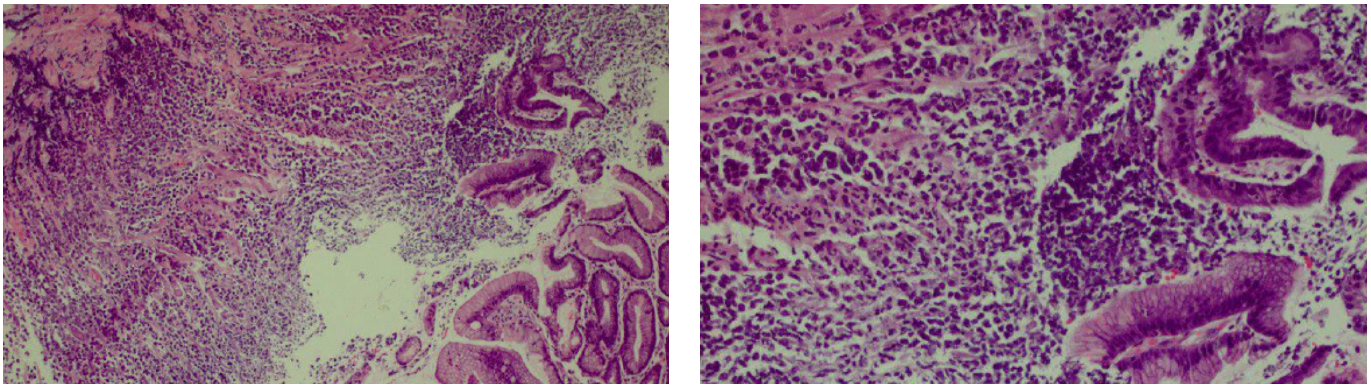

Figure 1. Microscopic findings of gastric Burkitt lymphoma. Proliferation of monomorphic medium-sized atypical lymphocytes with multiple mitotic figures and apoptotic figures in Hematoxylin-Eosin (x10).

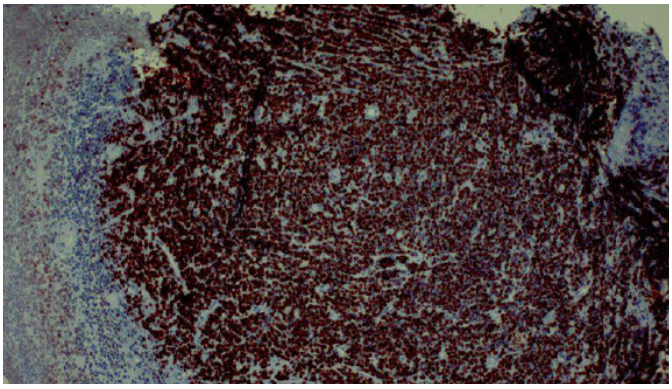

Figure 2. Ki67 staining was found close to 100% (x10).

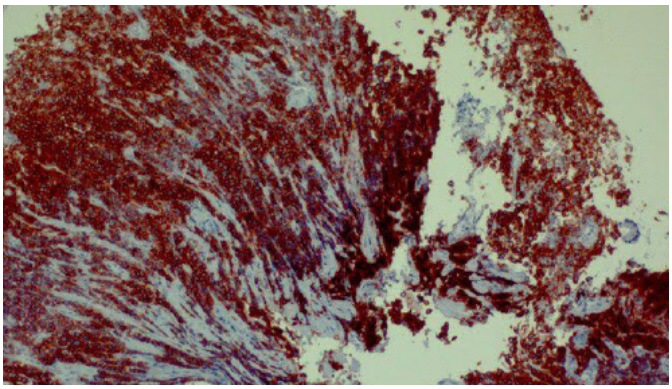

Figure 3. Immunohistochemical study revealed strong and diffusely positive neoplastic cells for LCA, c-myc, CD20 (x10).

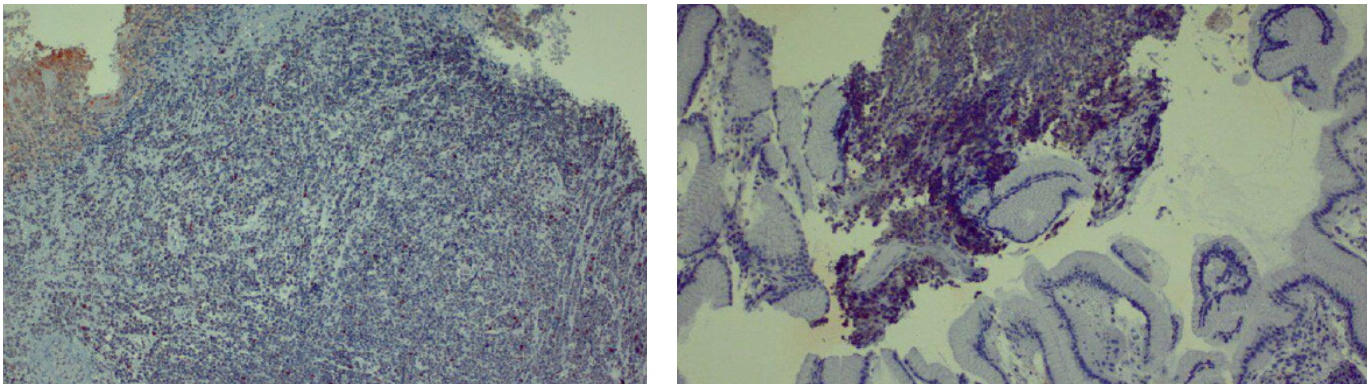

Figure 4. CD10 and bcl-2 were negative for tat, CD5, CD3 and cyclin D1.

gastrointestinal system, a giant malignant ulcer was detected in the antrum. Histological examination showed the proliferation of mid-sized monomorphic atypical lymphocytes containing many mitotic and apoptotic figures (*Figure 1*). The pathology was compatible with high-grade B-cell lymphoma. Ki67 staining was detected as near 100% (*Figure 2*). The immune histochemical study detected strong and diffuse positive neoplastic cells for LCA, c-myc, and CD20 (*Figure 3*). It was negative for CD10 and bcl-2, TdT, CD5, CD3, and cyclin D1 (*Figure 4*). MIB1 proliferation index was almost 100%. In the whole abdominal computed tomography, a filling defect with a wide opening and 42 mm diameter was observed in the gastric antrum level towards the lesser curvature. In PET-CT, in the stomach at the lesser curvature-antrum localization, increased F-18 FDG uptake (SUV max: 12.49) with approximately 4 cm diameter and soft tissue density concentrated in the region spanning towards the lumen was detected. No uptake was seen in the other areas of the body in PET-CT (*Figure 5*). The patient was referred to the hematology department for further tests and prompt treatment initiation.

Discussion

Burkitt's lymphoma is one of the most aggressive forms of B-cell NHL with a replication approaching 100% and has three clinical forms; endemic, sporadic and immunodeficiency-associated forms. An endemic variant is prevalent in Africa, while a sporadic variant is present in the USA and Western Europe, and the immunodeficiency variant is observed primarily in HIV patients. Sporadic variant includes 30% of pediatric lymphomas and less than 1% of adult NHLs.[5] The most commonly affected region, aside from lymph node involvement, is the gastrointestinal system (30-50%).[6] Primary gastrointestinal lymphoma is rare. Secondary involvement of the gastrointestinal system is common in lymphoma. Primary gastrointestinal lymphoma manifests with localized or mainly dominant symptoms in the gastrointestinal tract. Though gastric lymphomas are more common than intestinal lymphomas, primary gastric involvement is rare in Burkitt's lymphoma. For non-endemic Burkitt's lymphoma, the gastrointestinal system is the most common region, followed by retroperitoneal, kidney, ovary and testicular involvement.[7] Incidence of Burkitt's gastric lymphoma in adults is exceptionally rare. Burkitt's lymphoma is a highly aggressive malignancy and is one of the fastest-growing malignancies among human malignancies.[5] It requires immediate and aggressive treatment. Fortunately, regardless of being a rapidly growing malignancy, it responds to aggressive chemotherapy.[8]

In the literature, few cases related to this issue were found in the childhood age group. Our case is in the young age group. No new cases were found in the literature in the last ten years. Published patients present with mass effect, vomiting and abdominal pain. In our case, however, obstruction due to the mass did not occur yet, and only a persistent stomach was present. The presence of alarm symptoms is a priority in upper gastrointestinal endoscopy patients.[9] Our patient did not have any alarm symptoms. Duodenal ulcer, gastritis, and gastroesophageal reflux are most common in younger patients.[9]

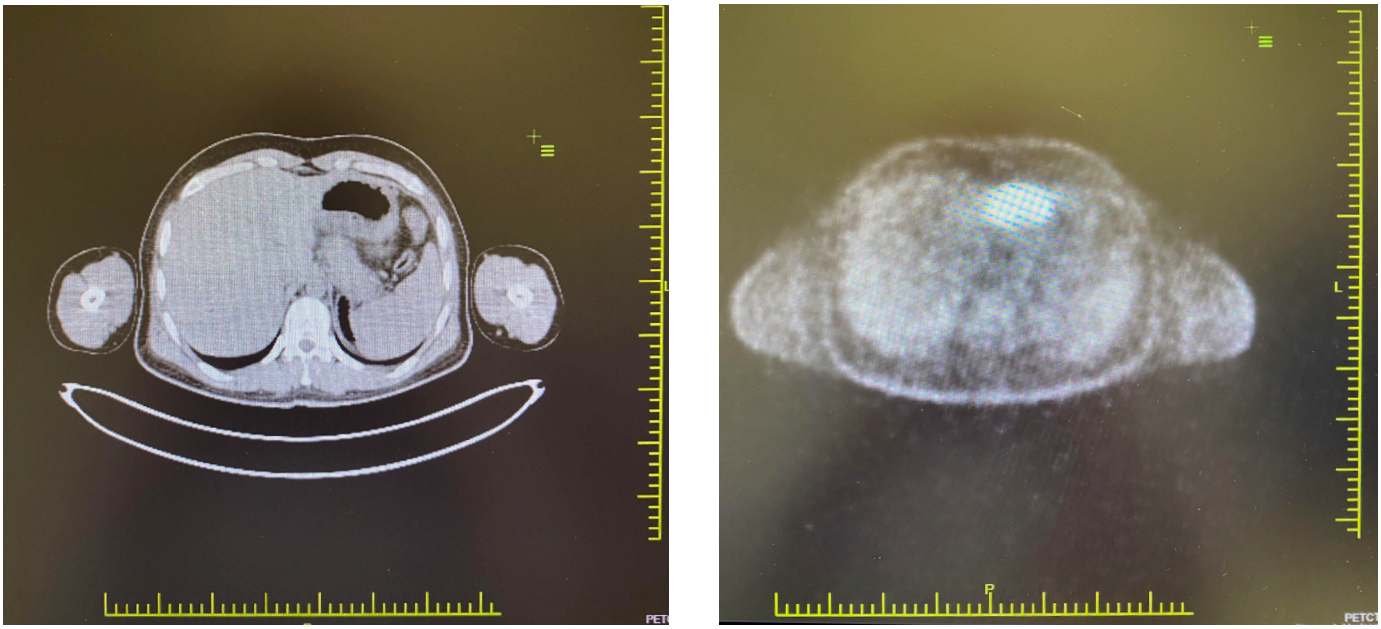

Figure 5. In PET-CT, intensely increased F-18 FDG uptake (SUV max: 12.49) was detected in the area extending to the lumen with a soft tissue density of approximately 4 cm in the small curvature-antrum localization of the stomach.

Conclusions

Gastric Burkitt's lymphoma is a rare form of non-Hodgkin lymphoma with a high rate of proliferation, aggressive nature, and poor prognosis in adult patients. Optimum treatment for this disease is still unknown. Aggressive chemotherapy should be a part of all treatment regimens. The advanced stage is associated with poorer outcomes in older age groups. While long-term survival rates of 70-80% may be possible with early diagnosis and timely appropriate treatment, delay in treatment might be fatal for these patients. Although it is not an alarming symptom, it should be kept in mind that gastric lymphoma can be seen in the young age group, and upper gastrointestinal endoscopy should be performed without delay. Early diagnosis of gastric lymphoma prolongs the life of the patient.

Ethical Approval

Our institution does not require ethical approval to publish an anonymous case report. Informed consent was obtained from the patient for the use and publication of data and images in the case report.

Conflict of Interests

The authors declare that they have no competing interests. The funders had no role in the design, conduct, analysis, or interpretation of data or in writing the manuscript.

Authors' Contribution

HH wrote the first draft of this paper. All authors approved the final version.

The case report has written in an anonymous characteristic. Thus secret and detailed data about the patient has been removed. Editor and reviewers can know and see these detailed data. These data are backed up by editors and by reviewers.

References

1. Young LS, Rickinson AB. Epstein-Barr virus: 40 years on. Nat Rev Cancer. 2004 Oct;4(10):757-68. doi: 10.1038/nrc1452.
2. Krugmann J, Tzankov A, Fiegl M, Dirnhofer S, Siebert R, Erdel M. Burkitt's lymphoma of the stomach: a case report with molecular cytogenetic analysis. Leuk Lymphoma. 2004 May;45(5):1055-9. doi: 10.1080/10428190310001623847.
3. Jang SJ, Yoon DH, Kim S, Yoon S, Kim DY, Park CS, Huh J, Lee SW, Lee DH, Ryu JS, Suh C. A unique pattern of extranodal involvement in Korean adults with sporadic Burkitt lymphoma: a single center experience. Ann

Hematol. 2012 Dec;91(12):1917-22. doi: 10.1007/s00277-012-1531-1.
4. Sekiguchi Y, Yoshikawa H, Shimada A, Imai H, Wakabayashi M, Sugimoto K, Nakamura N, Sawada T, Takeuchi K, Ohta Y, Komatsu N, Noguchi M. Primary hepatic circumscribed Burkitt's lymphoma that developed after acute hepatitis B: report of a case with a review of the literature. J Clin Exp Hematop. 2013;53(2):167-73. doi: 10.3960/jslrt.53.167.
5. Erkan G, Çoban M, Çalıskan A, Ataç GK, Gulpınar K, Degertekin B, Korkmaz A. A Burkitt's lymphoma case mimicking Crohn's disease:a case report. Case Reports Med. 2011;2011 doi:10.1155/2011/685273.
6. Koch P, del Valle F, Berdel WE, Willich NA, Reers B, Hiddemann W, Grothaus-Pinke B, Reinartz G, Brockmann J, Temmesfeld A, Schmitz R, Rübe C, Probst A, Jaenke G, Bodenstein H, Junker A, Pott C, Schultze J, Heinecke A, Parwaresch R, Tiemann M; German Multicenter Study Group. Primary gastrointestinal non-Hodgkin's lymphoma: I. Anatomic and histologic distribution, clinical features, and survival data of 371 patients registered in the German Multicenter Study GIT NHL 01/92. J Clin Oncol. 2001;19(18):3861-73. doi:10.1200/JCO.2001.19.18.3861.
7. Magrath IT, Shiramizu B. Biology and treatment of small non-cleaved cell lymphoma. Oncology (Williston Park). 1989 Nov;3(11):41-53.
8. Ferry JA. Burkitt's lymphoma: Clinicopathologic features and differential diagnosis. Oncologist. 2006;11(4):375-83. doi:10.1634/theoncologist.11-4-375.
9. Lúquez Mindiola A, Otero Regino W, Schmulson M. Diagnostic and therapeutic approach to dyspepsia and functional dyspepsia: what's new in 2019. Rev Gastroenterol Peru. 2019 Apr-Jun;39(2):141-52 (in Spanish).