Case report

The use of interventional radiology in the diagnosis and treatment of gastric bleeding in Burkitt lymphoma

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ABSTRACT

The paper presents a case of Burkitt lymphoma manifested by bleeding from a gastric ulcer. The control of bleeding required the use of diagnostic and therapeutic methods of interventional radiology.

Key words: Burkitt lymphoma, bleeding, interventional radiology

INTRODUCTION

Burkitt lymphoma (BL) is a tumor of the lymphatic system classified as highly aggressive non-Hodgkin lymphoma originating from B cells. Its rapid clinical course results from a 26-hour doubling of the tumor mass (the proliferative index Ki-67/MIB1 is 100% in the majority of cases). It is divided into 3 subtypes: endemic, sporadic and immunodeficiency-related. It mainly occurs in children and young adults [1]. In about 78% of cases, it involves the extranodal location (the central nervous system, gastrointestinal tract and bone marrow), in 11%, it involves the gastrointestinal tract, mainly the stomach. Sporadic BL has no specific geographical preference and occurs worldwide. It accounts for 1% to 2% of lymphomas in the adult population, mainly in men [2].

CASE REPORT

A 44-year-old patient was hospitalized in the surgery ward of the our center due to the third episode of bleeding from the upper gastrointestinal tract within a month. The patient underwent urgent gastroscopy which showed that the stomach contained blood and there was an extensive infiltration of the distal part of the stomach body with the presence of an ulcer covered with blood clots located at the bottom of the stomach. No signs of active bleeding were found. Histopathological specimens were collected from the lesion. The patient was qualified for conservative treatment until the above mentioned result was obtained. The obtained histopathological examination showed the presence of an infiltration with lymphatic cells corresponding to BL malignant lymphoma [CD20+PAX5+c-MYC+(100%)MUM1+CD10+bcl6+bcl2-CD5-Cd3-Cd23-CD3-EBV-ALK-Ki67 98–100%].

In order to complete the diagnosis and start antineoplastic therapy, the patient was transferred to the Clinical Department of Hematology of our center. During hospitalization, the patient was subjected to full diagnostics to assess the severity of the disease and prognostic factors. We observed no myeloid infiltration with cells corresponding to BL [CD45+CD19+CD20+ CD10+Cd22+CD38+CD43+Cd5-CD23-CD71+ phenotype]. No metaphases were found in cytogenetic testing. The FISH method revealed no *BCL6* gene rearrangement, *11q22.3* (ATM) deletion, *MYC* gene (8q24) rearrangement, *IGH/BCL2* gene fusion [t(14;18) (q32;q21)], or *17p* (p13.1) (TP53) deletion. Cerebrospinal fluid involvement (the presence of 4% of Burkitt-phenotype lymphoma cells) was confirmed. Positron emission tomography (PET) combined with computed tomography (CT), i.e., PET-CT, revealed an infiltration of the stomach, pylorus, and duodenum, as well as the surrounding fat and peritoneum reaching the pelvis. The involvement of the tumour process in the lymph nodes of the abdomen and chest was confirmed, which corresponded to the stage IV according to Lugano. Ultimately, the following diagnosis was made: malignant B-cell Burkitt lymphoma, stage IV B, IPI – high risk.

During hospitalization in the Clinical Department of Hematology, the patient experienced another episode of bleeding from the upper gastrointestinal tract. Gastroscopy was performed and, similarly to the previous examination, extensive infiltration and ulceration covered with clots were found at the previously reported location. The actual site of active bleeding was not visible again. Due to the suspicion of clinically significant vascular malformations associated with rapid tumor proliferation, a multiphase CT scan was ordered. One of the findings of the examination was an anatomical variant of the visceral trunk, occurring in about 30% of patients, consisting in an independent branching of the common hepatic artery from the aorta and a joint branching of the splenic artery and the left gastric artery from the aorta.

Moreover, the examination revealed a diffuse pathological infiltration of the gastric body with wall thickening of up to approx. 43 mm, and the presence of streaky densities in the surrounding fatty stroma with enlarged, quite numerous, oval lymph nodes up to 16 mm in size in the short axis view (fig. 1, 2). No active bleeding was noted in the arterial or venous phases. In order to create a more optimal environment for the healing of gastric ulcer as a potential bleeding site, it was decided to try embolization of selected gastric vessels. Under local anesthesia, the femoral artery was punctured using the Seldinger technique and a vascular sheath was inserted. The visceral trunk was catheterized with

Figure 1. CT examination showed an extensive marked thickening of the stomach wall in the distal part of the body in the pyloric area. No active bleeding was seen in either phase of the study.

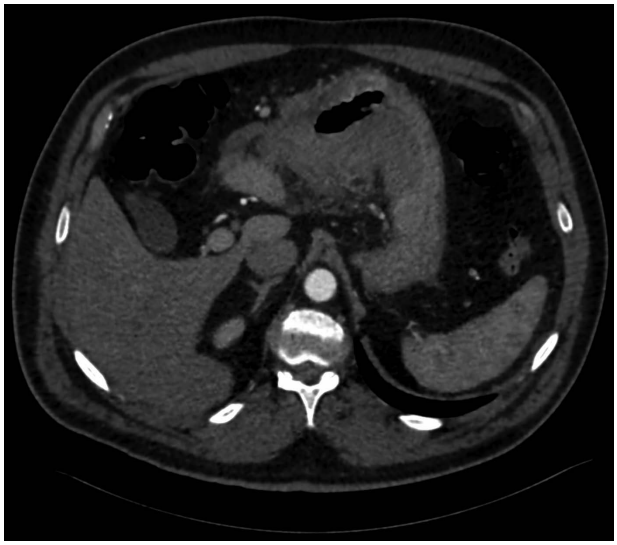
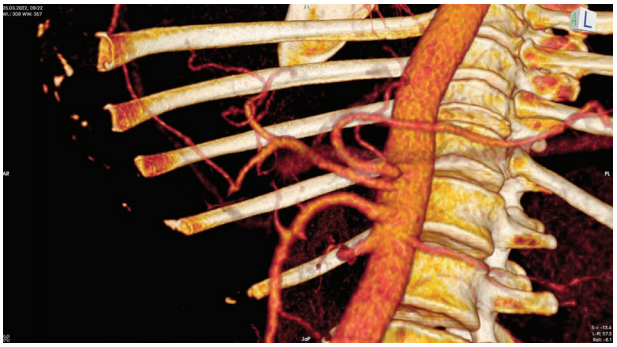


Figure 2. CT – volumetric reconstruction reveals the anatomical variant of the visceral trunk: independent branching of the common hepatic artery. The visceral trunk is divided into the splenic artery and left gastric artery.



a cobra catheter (Terumo C2). The anatomical variant visible in the CT was confirmed. The division of the common hepatic artery into the left hepatic artery and the common trunk of the right hepatic artery and the gastroduodenal artery was another anatomical variant found during the examination. The branching of the right gastric artery from the left hepatic artery was yet another atypical anatomical variant. Arteriography also showed an abnormal enhancement of the affected gastric wall supplied by the right gastro-omental artery (dominant) and, to a lesser extent, by the right gastric artery, without signs of active bleeding (fig. 3, 4). Using the Progreat 2.7Fr microcatheter (Terumo), the gastric branches supplying the infiltrates were selectively catheterized. The subsequent step involved temporary embolization with the spongo-

Figure 3. Arteriography of the common hepatic artery. Contrast enhancement was obtained in the visibly thickened stomach wall supplied by the right gastro-omental artery, without obvious extravasation of the contrast.

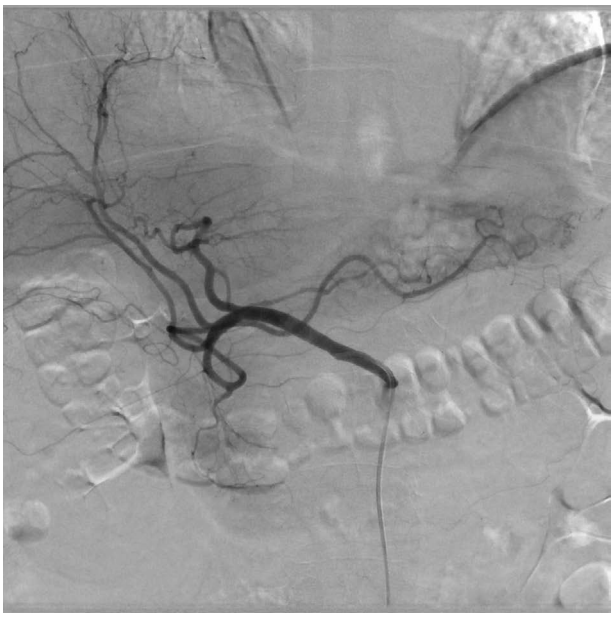
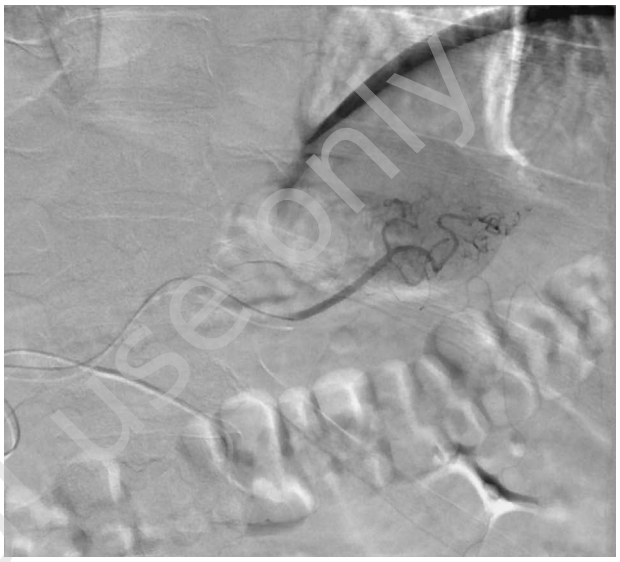
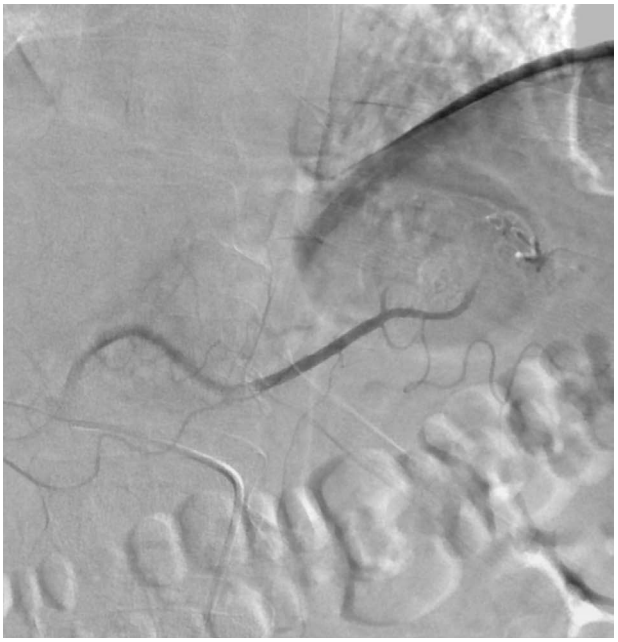


Figure 4. Selective arteriography of the right gastro-omental artery. Visible tortuous pathological vessels and contrast enhancement of the thickened stomach wall.



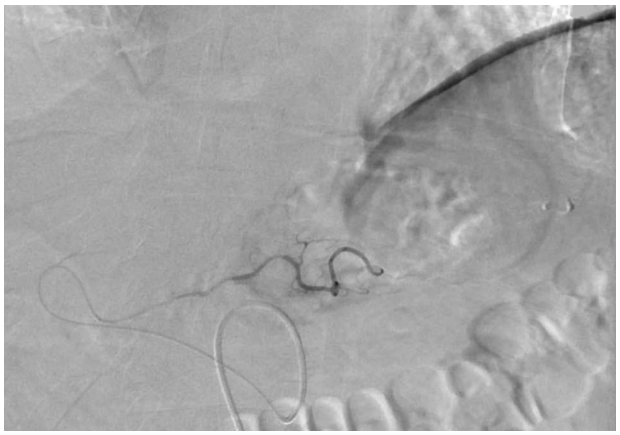
stan gel. Follow-up arteriography showed an effective reduction of infiltration vasculature (fig. 5, 6). After stabilizing the patient's clinical condition, antineoplastic treatment was started according

Figure 5. Post-embolization control – no pathological contrast enhancement of the stomach wall is visible.



to the Hyper-CVAD/MTX-Cytarabine regimen (rituximab, cyclophosphamide, vincristine, doxorubicin, dexamethasone, methotrexate, cytarabine), obtaining the regression of cancerous lesions and the resolution of disease symptoms. A PET-CT follow-up ex-

Figure 6. Selective right gastric arteriography – normal image. No pathological enhancement or extravasation of contrast.



amination, performed approximately 7 months after initiating the therapy, revealed the resumption of the neoplastic process. The patient was qualified for second-line treatment according to the R-DA-EPOCH regimen (rituximab, doxorubicin, etoposide, prednisone, vincristine, cyclophosphamide). During the treatment, the patient additionally received intrathecal chemotherapy as a prophylaxis of central nervous system involvement. After the completion of second-line immunochemotherapy, a complete regression of neoplastic lesions was obtained in the patient as confirmed with a PET-CT examination. Due to the high risk according to the IPI, the patient was qualified for the autotransplantation of hematopoietic cells as a consolidation treatment of remissions. The treatment was performed on April 20, 2023. The patient is currently in complete remission confirmed by the result of the PET-CT examination of July 27, 2023.

DISCUSSION

According to the National Cancer Registry, 937 patients with BL were registered in Poland in the years 1999–2017, which confirms the very rare occurrence of this type of cancer. In the years 2019–2023, 7 patients were under the care of our center, which is the leading hematology department in the province (the population of the province is 1.4 million). According to observations conducted in the world and in Poland, the survival rate of patients with this type of cancer has been systematically improving over the past two decades. This applies to both 3-year and 5-year survival (an increase from about 50% to about 70%) [3]. 3-year overall survival rates increased to approximately 90% after the inclusion of rituximab into the previously used regimens [4, 5].

CONCLUSIONS

The aim of the study was to present a case of rare, aggressive lymphoma with an unusual clinical course. The first problem associated with the presence of BL in the patient included recurrent coffee ground vomiting as a symptom of bleeding from the upper gastrointestinal tract, which is rare in the course of this disease. The implementation of emergency endoscopic diagnostics facilitated a rapid diagnosis of the underlying disease, which is particularly important in case of this type of cancer. Intensive systemic immunochemotherapy is the treatment of choice for BL as long as the patient's condition is stable [5, 6]. In the present case, the classic methods of the endoscopic treatment of upper gastrointestinal (stomach) bleeding were ineffective. The use of interventional radiology procedures proved to be a highly effective method of tumor vascular occlusion. Therefore, a rapid stabilization of the clinical status was achieved, which allowed for the efficient implementation of effective antineoplastic therapy and achieving complete remission of the disease.

References

1. Swerdlow SH, Campo E, Pileri SA et al. The 2016 revision of the World Health Organization classification of lymphoid neoplasms. Blood. 2016; 19: 127(20): 2375-90. http://doi.org/10.1182/blood-2016-01-643569.
2. Harne PS, Macklin J, Muniraj T. Disseminated Burkitt lymphoma presenting as massive gastrointestinal bleed. Proc (Bayl Univ Med Cent). 2020; 33(3): 433-5. http://doi.org/10.1080/08998280.2020.1747835.
3. Caetano Dos Santos FL, Michalek IM, Wojciechowska U et al. Improved survival of Burkitt lymphoma/leukemia patients: observations from Poland, 1999-2020. Ann Hematol. 2022; 101(5): 1059-65. http://doi.org/10.1007/s00277-022-04758-2.
4. Thomas DA, Faderl S, O'Brien S et al. Chemoimmunotherapy with hyper-CVAD plus rituximab for the treatment of adult Burkitt and Burkitt-type lymphoma or acute lymphoblastic leukemia. Cancer. 2006; 106(7): 1569-80. http://doi.org/10.1002/cncr.21776.
5. Roschewski M, Staudt LM, Wilson WH. Burkitt's Lymphoma. N Engl J Med. 2022; 387(12): 1111-22. http://doi.org/10.1056/NEJMra2025746.
6. Crombie J, LaCasce A. The treatment of Burkitt lymphoma in adults. Blood. 2021; 137(6): 743-50. http://doi.org/10.1182/blood.2019004099.

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