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Case Report

Burkitt's lymphoma revealed by intestinal obstruction: A case report ☆

*Eric Michel Kessi**, *Yousra Guelzim*, *Amine Bennani*, *Fatim Zahra Laamrani*, *Laila Jroundi*, *Omar El Aoufir*

Emergency radiology department, UHC Ibn Sina Mohamed V University, Rabat, Morocco

ARTICLE INFO

Article history:
Received 29 May 2024
Accepted 5 June 2024

Keywords:
Burkitt's lymphoma
Small intestine
Intestinal obstruction

ABSTRACT

Intestinal obstruction is a surgical emergency frequently encountered in routine practice, usually caused by abdominal adhesions. Although extra nodal lymphoma is most often localized in the gastrointestinal tract and may be responsible for intestinal obstruction, Burkitt's lymphoma is a very rare cause in adults.

We report a case of Burkitt's lymphoma mimicking an intestinal obstruction in a 48-year-old adult who presented with an obstructive syndrome and altered general condition.

Imaging and anatomopathological examination after immunohistochemical analysis concluded to a multi-systemic Burkitt's lymphoma.

Chemotherapy was immediately started with complete remission.

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Introduction

Small bowel occlusive syndromes are a common reason for admission to the emergency department [1]. Indeed, it is one of the most frequent surgical emergencies, accounting for up to 15% of hospital admissions for acute abdominal pain [2]. Postoperative adhesions remain the predominant cause, accounting for around two-thirds of all obstructive events [3]. Intraluminal tumors play a significant role, including lymphomas, which account for around 24% of neoplasia-induced intestinal obstructions [4], with a few cases attributed to Burkitt's lymphoma (BL) in adults.

Imaging, in particular computed tomography (CT), can be used to identify the tumor burden, its extent and the thickness of the intestinal wall, but only histological and immunohistochemical studies can confirm the diagnosis [5].

The authors report this case to emphasize the rare and nonspecific presentation both clinically and radiologically, and at the same time show the importance of the radiological role in the diagnosis and management of an occlusive syndrome due to BL in order to avoid further complications such as perforation or intra-abdominal sepsis in an area considered sporadic.

Case report

A 48-year-old man with no specific medical history presented with an occlusive syndrome 1 month prior to his admission to the emergency room, for which he was initially put on symp-

☆ Competing Interests: The authors declare no conflicts of interest.

* Corresponding author.
E-mail address: ericmichelkessi@gmail.com (E.M. Kessi).

https://doi.org/10.1016/j.radcr.2024.06.019

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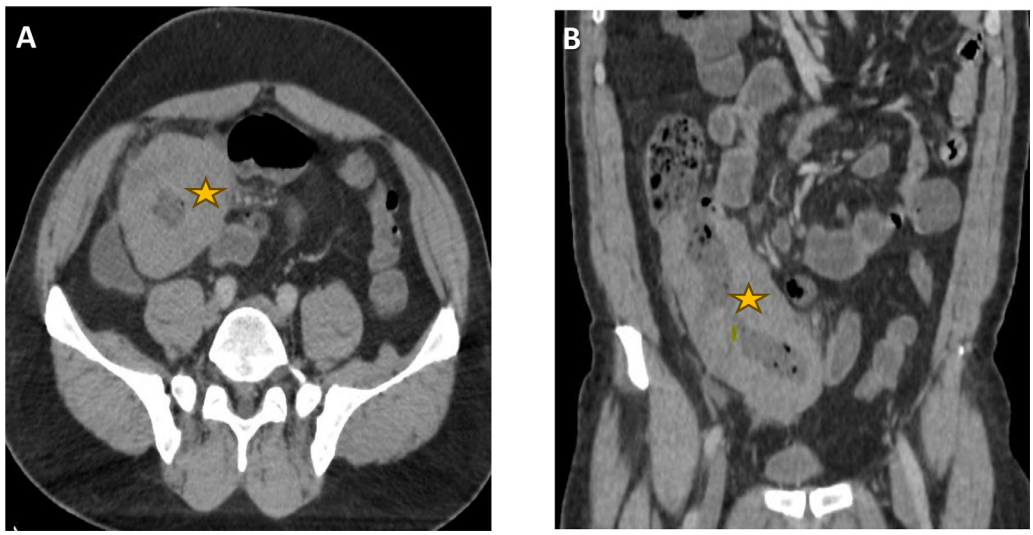

Fig. 1 – Abdominal CT with PDC injection in axial (A) and coronal (B) sections showing pseudo-stenotic ileo-caecal parietal circumferential tumour thickening (orange star) with discrete infiltration of adjacent fat.

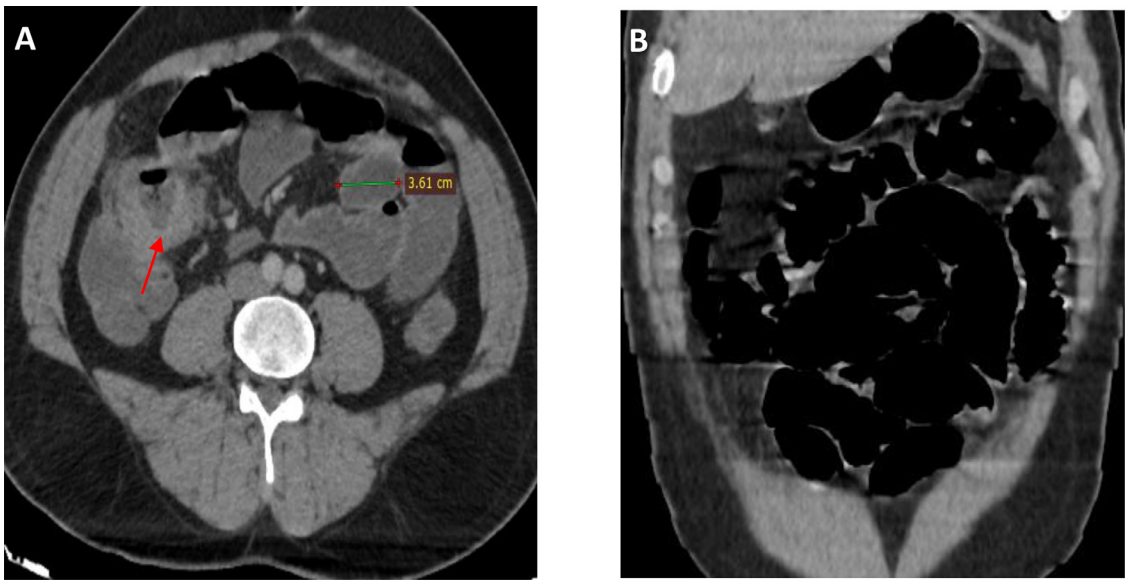

Fig. 2 – Abdominal CT scan after injection in axial (A) and coronal (B) sections: circumferential ileo-caecal mass (red arrow) enhanced after injection, associated with diffuse distension of the small intestines and hydro-aeric levels in relation to an occlusive syndrome.

tomatic treatment by his physician with slight clinical improvement. The evolution was marked by worsening abdominal pain in a context of altered general condition (weight loss and asthenia). Clinically, the patient was conscious, apyretic and hemodynamically stable; physical examination revealed tenderness in the right iliac fossa. The rest of the examination was unremarkable; the viral serology performed (HIV, HBV, HCV, and syphilis) was negative; EBV serology came back positive. An abdominal CT was ordered and showed regular pseudo-stenotic parietal thickening of the last ileal loop measuring 36 mm with discrete infiltration of the mesenteric fat opposite (Figs. 1 and 2A) associated with distension of the ileal bowel and ascending colon (Fig. 2).

Histopathological examination with immunohistochemical analysis after endoscopy was in favor of Burkitt's lymphoma (Fig. 3).

Discussion

Primary malignant tumors of the small intestine are very rare, accounting for less than 2% of all gastrointestinal malignancies. Burkitt's lymphoma is a fairly aggressive, fast-growing but potentially curable disease that belongs to the group of non-Hodgkin's lymphomas (NHL) and accounts for less than

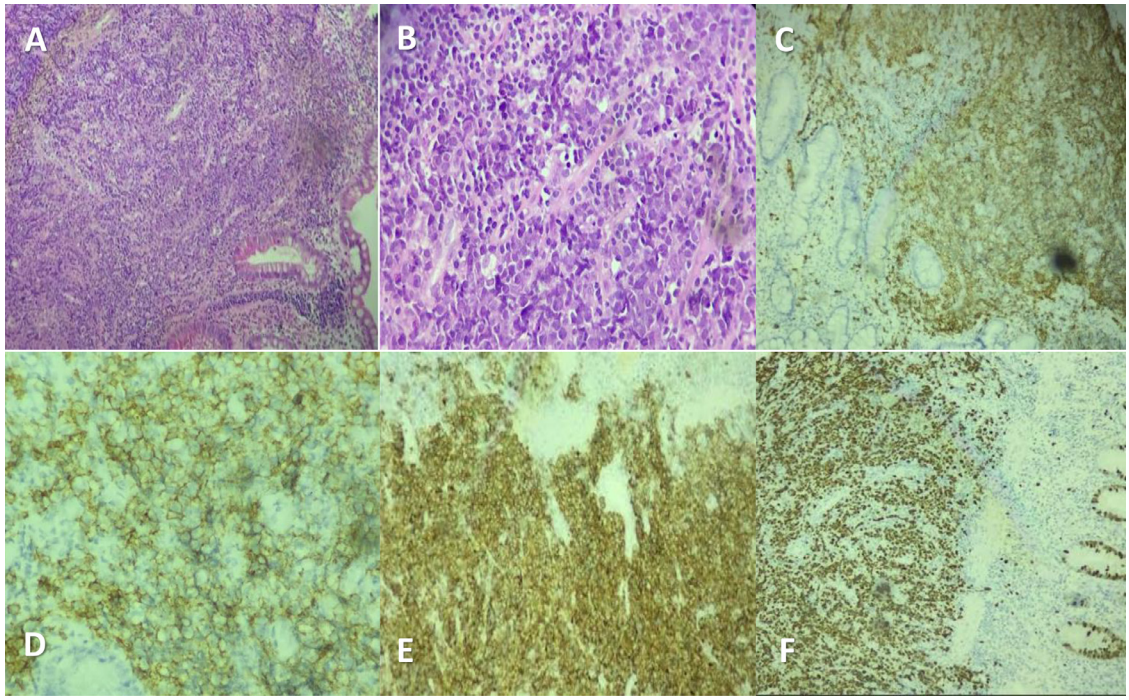

Fig. 3 – Pathological and immunohistochemical images of an ileal biopsy: HE Gx10 (A) and HE Gx40 (B): ileal mucosa infiltrated by a lymphomatous-like process arranged in a diffuse sheet made up of medium-sized cells with sparse basophilic cytoplasm and ovoid nuclei with fine chromatin and one or more nucleoli.
- Diffuse and intense positive membrane immunostaining with anti CD20 (C) and CD10 (D) antibodies confirming the diagnosis of BL.
- Positive nuclear staining with anti-BCL 6 antibodies (E).
- Positive Ki proliferation index estimated at 98% (F).

1% of NHL [6,7]. Three forms of BL have been recognized by the World Health Organization: endemic, immunodeficiency-associated and sporadic [8].

The endemic form accounts for 30%-50% of childhood cancers in equatorial Africa, with an incidence rate 50 times higher. The significant increase in this incidence is due to the frequency of falciparum plasmodium (malaria) in Africa and its link with Epstein-Barr virus (EBV). However, the etiological role of the EBV virus has yet to be determined. Jaw involvement remains the most common [9,10].

The immunodeficiency variant is mainly observed in HIV-infected individuals [11], contrasting with a higher CD4 count [12], and generally in the absence of opportunistic infections. It may also occur in patients with hereditary immune deficiencies, or in those taking immunosuppressive drugs to prevent rejection after organ transplantation [13]. Its localization is similar to that of the sporadic form.

The sporadic form is common in the United States and Europe, accounting for 30% of pediatric lymphomas, with peak incidence at age 11. In adults, BL is more common in Caucasians, with a median age of 30 [10]; it often presents voluminous tumors in extra-nodal sites, with a preferential abdominal location in 70-90%, generally in the region of the ileocaecal valve or mesentery [14]. Tumor cells express Ig M membrane immunoglobulin, Ig light chain, B-cell antigen, B-cell lymphoma (BCL) protein 6, differentiation group (CD) 10, 19, 20, and 22, while expressing negative results for CD 5, 23, and BCL 2 [9]. It is characterized by a specific translocation of the C-myc gene in band q24 of chromosome 8 [15,16]. BL has a very high proliferation index (Ki67), generally above 95%. So it's not surprising that BL is the fastest-growing human cancer [9].

Clinical symptoms of BL in the gastrointestinal tract may be non-specific, usually presenting as an abdominal mass, and are often accompanied by abdominal pain, nausea, vomiting, weight loss and rarely acute obstructive symptoms [16,17]. They may also manifest as acute appendicitis or intussusception in the pediatric population. Direct compression of the intestinal lumen is responsible for obstruction. Persistent symptoms of obstruction and weight loss can be significant clinical clues to the presence of gastrointestinal lymphoma [18]. This presentation was present in our case.

Radiologically, intestinal BL may present as symmetrical or slightly asymmetrical dedifferentiated thickening of the circumferential wall of the coves up to 7 cm, or as intestinal pseudoaneurysmal dilatation as described by Balthazar et al [19] resulting from tumor replacement of the muscularis and destruction of the autonomic nerve plexus. This can result in intestinal occlusion due to extrinsic compression, with or without infiltration of adjacent loops. However, this remains a very rare consequence of lymphoma due to the absence of a desmoplastic reaction [20]. The CT scan should also look for signs of severe occlusion.

The main differential diagnoses are adenocarcinoma of the small intestine and thickening of the intestinal wall of infectious or inflammatory origin, notably ileocaecal tuberculosis. However, certain criteria can guide the diagnosis; in the case of lymphoma, parietal thickening is circumferential segmental or multi-segmental, moderately enhanced, and the junction with the normal intestinal segment is progressive [20,21]. The slight enhancement of the wall and the discreet infiltration of fat led us to this diagnosis.

BL is an aggressive cancer that requires rapid recognition and treatment to improve outcomes. Il n'y a pas de consensus quant au traitement de première intention du BL, mais une chimiothérapie combinée à une chimioprophylaxie est conseillée. There is no consensus on the first-line treatment of BL, but chemotherapy combined with chemoprophylaxis is recommended.

The Ann Arbor classification with Musshoff modification is commonly used to classify gastrointestinal lymphoma, and the International Prognostic Index has been used to define prognostic subgroups; increasingly, the Paris classification has gained prominence [18].

Other forms of non-Hodgkin's lymphoma, such as lymphoblastic lymphoma, blastic mantle cell lymphoma and diffuse large B-cell lymphoma, may have identical imaging features and thus mimic Burkitt's lymphoma. Differentiation of course requires histopathological analysis [10], which provides a definitive diagnosis by digestive endoscopy with biopsy of the anantomopathological specimens.

Histopathologically, almost 90% of primary gastrointestinal lymphomas are of the B-cell lineage, with very few T-cell and Hodgkin lymphomas [17]. The characteristic histopathological feature is that of small, monomorphic, densely clustered, uncleaved cells, producing the classic "starry sky" appearance [3].

The characteristic histopathological feature is that of small, monomorphic, densely clustered, uncleaved cells, producing the classic "starry sky" appearance [3].

The role of surgery in gastrointestinal Burkitt lymphoma remains controversial and is limited to complications such as abdominal syndromes, intussusception or perforation [20]. Nevertheless, it responds dramatically to combination chemotherapy, which induces rapid initial tumor resolution and often long-term remission [18]. Our patient benefited from medical treatment with a good clinical course.

Conclusion

Small bowel obstruction is rarely due to Burkitt's lymphoma in the adult population and is more frequently observed in children and young people. Burkitt's lymphoma is very aggressive, with rapid cell turnover and significant morbidity. It may present clinical and scanographic aspects that are not specific to the disease. In the face of deteriorating general condition or rapid recurrence of an acute abdominal process, a more sinister cause such as malignancy must be suspected, investigated and confirmed by anatomopathological examination ; recognition of the various radiological aspects of Burkitt's Lymphoma, even if they are not specific, may enable the clinician to accelerate appropriate chemotherapeutic management of these patients.

Patient consent

Informed consent was obtained from the patient included in this study to be published in this article.

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