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Case Report **General Surgery**

Rare Localization of Hodgkin's Lymphoma: A Case Report

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Abstract

Hodgkin's disease or Hodgkin's lymphoma is a rare malignant hemopathy characterized by the presence of Reed-Sternberg cells. It is a lymphoid disease that the cause remains unknown. Subdiaphragmatic localized forms are rare and represent 6 to 13% [4-6]. The revelation of these forms by acute intestinal intussusception is exceptional, and attested by very few data published in the literature. We report the case of a 70-year-old patient with a rare case of subdiaphragmatic Hodgkin's lymphoma revealed by acute intestinal intussusception.

Keywords: Hodgkin's Lymphoma, Small Intestine, Intestinal Intussusception.

BACKGROUND

The history of Hodgkin's lymphoma dates back to 1832 with Thomas Hodgkin's macroscopic anatomical description of a painless increase in the volume of the lymph nodes and the spleen in six patients [1]. Two of the six cases described by him are what we now call Hodgkin's lymphoma (HL). The characteristic cells were described in detail by Carl Sternberg in 1898 and Dorothy Reed in 1902, hence their current name, Sternberg or Reed-Sternberg (SRS) cells. It is a cancer of the lymph nodes characterized by the disappearance of their normal architecture and the presence of very few Sternberg or Reed-Sternberg (SRS) cells (1 to 3%) in the middle of inflammatory cells. This Reed-Sternberg (SRS) cell is the basis of the diagnosis. The anatomopathological examination makes the diagnosis.

Clinical and biological studies have demonstrated that it is a lymphoid disease grouping two very different entities according to the 2008 WHO classification of malignant hemopathies: classical Hodgkin lymphoma (CHL), by far the most frequent, and nodular lymphocyte-predominant Hodgkin lymphoma (NPLH), rare which represents less than 5% of cases [2, 3].

The Ann Arbor classification modified by Cotswolds defined four (4) stages I to IV according to the extension of the disease. It was used for several decades to define the therapeutic strategy. It distinguished between localized stages (I and II) and extensive stages (III and IV) [4]. The most frequent localizations are the mediastinum, left or right cervical observed in approximately 60% of patients. Other sites include the splenic, axillary, abdominal, hilar or inguinofemoral sites [5]. Localized stages are categorized into supradiaphragmatic and subdiaphragmatic. Subdiaphragmatic represents a rare form with 6 to 13% of localized forms and very little published data [6, 7]. However, it is recognized that this subdiaphragmatic form is a risk factor just like the infection by the Epstein-Barr virus (EBV) in the genesis of Hodgkin's lymphoma [8, 9]. Indeed, the EBV-LH link is more frequent in underdeveloped countries than in France and in developed countries [10]. However, the risk of developing Hodgkin's lymphoma after an EBV infection is relatively low, around 1/10,007 patients [11].

In terms of treatment, Hodgkin's disease was the first curable malignant disease, although it had been almost always fatal until the 1960s. The goal of treating affected patients is cure without sequels and maintaining optimal quality of life. The radiotherapy in localized forms allows to obtain healing. However, during successive therapeutic trials, the role of radiotherapy has been restricted in favor of an association of chemo-radiotherapy. The modalities of this association are still debated. However, chemotherapy modalities are now adapted to the early response assessed by PET. Efforts

are still needed to reduce treatment toxicity and improve the prognosis of forms that relapse or are resistant to standard treatment. Currently, provided an adequate initial classification and an appropriate therapeutic strategy, Hodgkin's lymphoma can be cured in the vast majority of cases, with a rate of 90% [12-15].

Intussusception is defined as the telescoping of a segment of the gastrointestinal tract into an adjacent one. Although intussusception is a common cause of severe intestinal obstruction in pediatrics, it is rare in adults and represents approximately 1 to 5% of all cases of intestinal obstruction in adults [16]. While this condition is very rarely observed in developed countries, it is relatively common in Africa, particularly in intertropical areas. The reasons for these geographical differences are unknown, and factors such as diet and parasites have been suggested [17]. In adults, an organic cause is found in 70 to 90% of cases of intussusception, while approximately 8 to 20% are idiopathic. Conversely, in children, intestinal intussusception is predominantly primary in 90% of cases [18]. The main causes of intussusception in adults include neoplasms, adhesions, surgical anastomoses, and stranger things. Rarely, metastases in the small intestine, such as those from melanoma, may be the starting point [19].

The clinical presentation of intussusception is variable and can be either chronic or acute. It realizes many kinds of mechanical intestinal obstruction. The symptoms and the context of occurrence help for the diagnosis and indicate surgical exploration, sometimes urgently. However, due to the high localization of the lesions, the clinical presentation can be subtle and confusing. Hydro-electrolytic disorders make the emergency of the treatment. Early diagnosis requires abdominal CT scan and high clinical suspicion, because the symptoms of intussusception are often nonspecific [20]. The mechanism of the intussusception can be enteric, ileo-cecal or colic. If there is doubt about the etiology of intussusception, surgical resection is recommended [21].

CASE PRESENTATION

A 70-year-old male patient with no known pathological history was admitted to our hospital for occlusive syndrome evolving since seven (07) days. The symptomatology includes permanent epigastric pain accompanied by bilious vomiting that became secondarily fecaloid and the impossibility of emitting faeces and gas. The abdominal CT scan revels a small intestine intussusception, with several deep aorto-mesenteric and porto-cave lymphadenopathies. Laparotomy revealed a 10 cm long intussusception of the last ileal intestine [Figure 1]. Exploration of the rest of the abdomen showed the presence of multiple adenopathies, and absence of ischemia signs. Small intestine tumor was suspected. We realized an ileocecal resection within some nodules and an ileocolic anastomosis. The tumor on the open specimen measured 3 cm [Figure 2]. The postoperative was no complications. Histological examination of the surgical specimen showed a malignant Hodgkin lymphoma.

After his discharge, the patient was referred to the clinical hematology department for continued treatment.

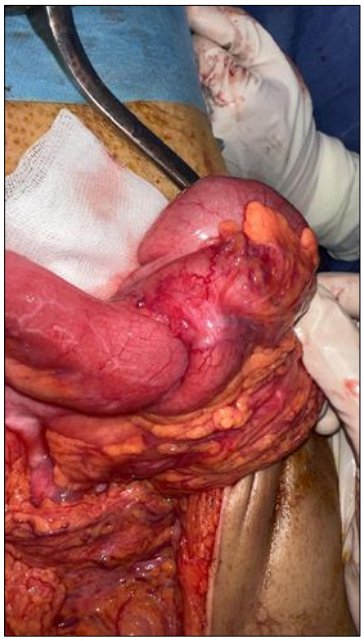

Figure 1: per operative picture of the intussusception

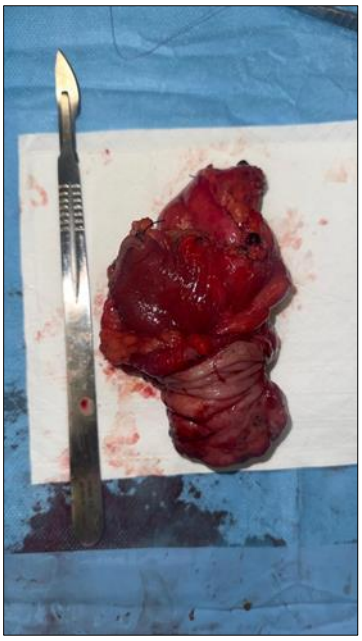

Figure 2: picture of the specimen

DISCUSSION

We report the case of subdiaphragmatic Hodgkin lymphoma revealed by acute intestinal intussusception in a 70-year-old patient. Two rare pathologies in adults.

Indeed, intussusception of the small intestine, common in infants, is rare in adults where it is estimated to 1% of all cases of small intestine occlusion, and 5% of all intussusceptions' cases [22]. It can occur at any age due to its diverse etiologies and in both sexes [23].

As for Hodgkin's lymphoma, Colonna in France found that it mainly affects adults, with two peaks of frequency observed in industrialized countries, but not elsewhere, one around 30 years and the other after 60 years, a slight female predominance (sex ratio: 0.92) and the incidence in children is low in developed countries, higher in developing countries with a strong male predominance, of the order of 4 boys for one girl [15]. In the United States, in a sample of 80,500 patients, Shanbhag found that Hodgkin's lymphoma represented 10% of lymphomas (90% non-Hodgkin's lymphomas) or 0.2% of all cancers, a male predominance of 56% and a median age of 39 years [11]. Kahn, for his part, found an incidence of approximately 85,000 patients per year and a predilection for adolescents and young adults aged between 15 and 39 years [14].

The association of the two pathologies, although rare, has been reported by other authors such as Amal Bennami in Morocco [24], and KAS in Senegal [25], who both presented cases of Hodgkin's lymphoma revealed by acute intestinal intussusception in elderly men as in our case. If this association of Hodgkin's lymphoma-intestinal intussusception is rare and very little documented, this is not the case for the association of non-Hodgkin's lymphoma-intestinal intussusception which has been the subject of several publications in the literature as attested by AKBULUT who presented the case of a 62-year-old woman and found in the literature 34 cases of association of non-Hodgkin's lymphoma-intestinal intussusception, and two cases of association of Hodgkin's lymphoma-intestinal intussusception [26]. Similarly, several other cases of non-Hodgkin's lymphoma-intestinal intussusception have been described in the literature [27, 28].

Hodgkin's disease is revealed in most cases by the discovery of painless, firm, non-fixed superficial lymphadenopathy, preferentially affecting the lower cervical or supraclavicular areas [29]. In our case, it was a subdiaphragmatic form, which is rare.

Regarding intestinal intussusception, the clinical examination is not often suggestive of the diagnosis, hence the interest in using imaging, of which CT Scan is the most sensitive examination for the small bowel site [22]. The radiological image is very suggestive. The site is most often ileo-ileal, as in our case. There are also ileo-colic and multi-stage cases [30]. In the literature, the rare cases described were of inflammatory origin [31], although tumor causes are dominant [32].

The surgical approach is performed either by laparotomy or by laparoscopy. A resection of the intussusception is performed if the tumor cause is immediately recognized for oncological reasons and due to the frequency of necrosis [33]. In all cases, analysis of the surgical specimen should be carried out even if no occult tumor is found [34]. Depending on the conditions, a temporary ileostomy or an immediate anastomosis may be opted for. The postoperative course is generally

simple but can be burdened by a high mortality rate in poor conditions [35].

CONCLUSION

Acute intussusception due to a small intestine tumor in adults is a rare disease. Its severity is due to acute upper intestinal obstruction and its hydro electrolytic consequences. When properly managed, primarily by surgery, the results are positive. The revelation of digestive lymphoma by intussusception helps for its early diagnosis. Emergency chemotherapy offers hope for a complete cure.

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