

Distal Jejunal Intussusception from Melanoma Metastasis: Case Report

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Abstract: Introduction: The invagination of one intestinal loop within another rarely occurs in adults and it is usually caused by a polyp or another intraluminal lesion. Intussusceptions occur mostly in children; an organic lesion is not necessary, and colic pain syndrome, rectal bleeding, and a palpable mass (the intussuscepted segment) are characteristics. Justification: To compare data from a tertiary reference hospital with data described in the literature, considering the evolution of such patients is unknown in our field. Objective: To report the case of a patient with distal jejunal intussusception from melanoma metastasis. Method: Information was obtained through medical record reviews, patient interviews, photographic records of the diagnostic methods and the surgery to which patients were subjected, and literature review. Final considerations: The case reported and the publications surveyed bring to light the discussion on the therapeutics of the complex condition that is melanoma metastasis, showing that some patients may present satisfactory and lasting results regarding symptom relief and improvement of quality of life.

Key words: Intussusception, melanoma, metastases, jejunal-jejunum, malignancy.

1. Introduction

The telescoping of a segment of the intestinal colon (intussusceptum) to the adjacent segment (intussusciens) is the most common cause of intestinal obstruction in children aged between six months and two years. The intussusception process may result in gangrene of the intussusceptum. The most common form is the intussusception of the terminal ileum to the right colon (ileocolic intussusception), and in 95% of infants and children, it is idiopathic. The disease is more frequent midsummer and midwinter, and it is correlated to adenovirus infections. Most of the cases show hypertrophied Peyer plaques in the edge of the intestinal border. Mechanical factors such as Meckel's diverticulum (Henoch-Schönlein purpura) and intestinal lymphoma are increasingly present in patients older than two years.

Postoperative intussusception may occur in any age and it is usually ileal-ileum or jejunal-jejunum, and due to differential return of intestinal motility, oftentimes after retroperitoneal surgery. The man to woman ratio is 3:2. The peak age is infants aged between five and nine months, and 80% of the patients are younger than two years [1].

Adult intussusception is rare. It is estimated to occur in only 5% of intussusception cases, it represents only 1% of all small bowel obstructions, and it is responsible for 0.003-0.02% of all hospital admissions. Around 90% of intussusceptions in adults present a defined cause, such as neoplasia or postoperative conditions. Neoplasia is the most common cause and it is found in approximately 65% of cases. Malignant tumors are more frequent than benign colon tumors. In contrast, benign causes are the most common for the small bowel [2].

Regarding the symptoms, pain occurs in episodes of about 1 minute, alternating at intervals of apparent comfort. Reflex vomiting is an early sign, but vomiting

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due to intestinal obstruction occurs late. The blood from venous infarction and mucous produces is “gelatinous feces”. In small infants and postoperative patients, colic pain may not be apparent. Usually, a mass is palpated following the colon distribution, more often in the right upper quadrant of the abdomen. Occasionally, an intussusception is palpated to the rectal examination. The prolonged intussusception produces edema and hemorrhagic or ischemic infarction of the intussusceptum [1].

In adults, the clinical presentation may not be specific, rarely presenting itself as the classic triad of abdominal pain, palpable mass, and blood in feces; on the contrary, the symptoms are usually of intestinal obstruction. The most common symptom is abdominal pain associated with symptoms of partial obstruction, such as nausea, vomiting, stuffing, changes in the intestinal habit, constipation, and gastrointestinal bleeding. The symptoms are usually severe and last from days to weeks, and they are rarely chronic, lasting for years. The physical examination may reveal a distended and rigid abdomen, with defense and decrease or absence of bowel sounds. In the late appearance, the patient may present peritonism and intestinal ischemia [3].

The definite diagnosis is based on surgical findings; however, imaging examinations and minimally invasive procedures may be useful. Simple and contrast radiographs, ultrasound, computed tomography, endoscopy, and colonoscopy may reveal the segment affected by the disease. The simple abdominal radiograph may show signs of obstruction and estimate the abdominal topography. Barium studies may aid diagnosis, but are not indicated in cases of suspected ischemia or intestinal perforation. Colonoscopy may aid in cases of colonic obstruction [4]. The most used radiological method is the abdominal tomography, with up to 100% sensitivity and specificity [5].

The standard treatment for intussusception in adults has not been defined yet. Most of the authors agree that laparotomy is mandatory to identify an organic lesion,

which is oftentimes neoplastic. The controversies revolve around the need and the extension of the resection [4].

2. Case Report

Male patient, white, born in Passo Fundo-RS-Brazil, 60 years old, sought emergency care at the City Hospital of Passo Fundo on March 20, 2017 complaining of abdominal distension and pain, constipation, vomiting, and inappetence, which had started 60 days prior and had worsened over the last 15 days. He reported a 10 kg weight loss over the last 20 days.

In 2015, the patient had been diagnosed with nodular melanoma in the right lower limb, Breslow of 6.5 mm, ulceration, and 7 mitoses/field. At that time, he was subjected to the extension of surgical margins and mapping of patent blue sentinel lymph node. The sentinel lymph node was negative for neoplasia. It was staged as T4N0M0. So far, the patient had been under treatment with interferon.

The physical examination showed pain facies, tachycardia, hypotension, abdomen tympanic to percussion, defense on abdominal palpation, and rectal examination without feces.

The laboratory tests showed leukocytosis and increase of the C-reactive protein.

The abdominal tomography (Fig. 1) showed in the hypogastric region—involving distal jejunal loops—an image with “loop-within-loop” aspect, possibly related



Fig. 1 Abdominal computed tomography.

to the intestinal intussusception, which determined the upstream loop distension. The potential presence of a neoplastic implant as a causal factor should be considered.

Due to the case of obstructive acute abdomen, an evaluation from the hospital's Oncology Surgery Service was requested, and emergency surgery was indicated.

Laparotomy was performed. After opening the abdominal cavity an inventory was done, and multiple implants were identified in the visceral peritoneum of (Fig. 2) the small bowel with the invasion of light. During the palpation of the entire small bowel, multiple nodules were identified within the light, as well as a distal jejunal intussusception area (Fig. 3). The intussusception was undone (Fig. 4).

The partial enterectomy was performed with the help



Fig. 2 Implants in visceral peritoneum.



Fig. 3 Distal jejunal intussusception.



Fig. 4 After undoing intussusception in distal jejunal loop.



Fig. 5 Open surgical piece.

of linear staplers, resecting the entire area of intussusception and production of an ileostomy in the right abdominal wall. The surgical piece was opened (Fig. 5) and showed the presence of multiple vegetative lesions (melanoma metastasis).

The patient evolved satisfactorily in the postoperative period, recovering function and intestinal transit, with functioning ileostomy, adequate hydration, and recovery of appetite.

A cranial tomography was performed at hospital admission, revealing single brain metastasis, but the patient was asymptomatic from the neurological standpoint. An evaluation from the Neurosurgery Service was requested, and surgical treatment was indicated. The patient and his family did not agree with the performance of brain surgery, so radiotherapy was indicated.

The patient was released from the hospital on April 4, 2017 and referred to the Clinical Oncology Service to continue the treatment of his underlying disease. Treatment continued with interferon and with radiotherapy sessions for brain metastases, with the last session being at the end of the same month. The patient discontinued treatment and follow-up at the end of April, when he did the last radiation therapy session, not returning for the next appointments. He ended up dying 3 months later in his hometown.

The biopsy result showed melanoma metastasis, forming nodules in the small intestinal wall. The lesions measured between 0.2 cm and 4 cm on the largest axis and compromised the mucosa, submucosa, muscular and serous layers. There was presence of angiolymphatic and perineural infiltrations. The proximal and distal surgical limits were free of neoplasia.

3. Discussion

The incidence of melanoma is increasing faster than any other type of cancer. The reasons for such increase are unclear, but they are more likely related to the greater exposure to sunlight ultraviolet radiation. Individuals whose first sunburn occurred at an early age have a higher incidence of melanoma. Other risk factors include freckles, a light complexion, red or blonde hair, blue eyes, first-degree relatives with melanoma, and the presence of multiple or dysplastic nevi [6].

According to the National Cancer Institute in Brazil, 6,260 cases of melanoma are expected in the country in 2018. In Rio Grande do Sul (the state of our patient), 860 new cases are expected [7]. Intestinal melanoma metastasis is rather common, and the rate for small bowel involvement ranges from 35 to 70%. It is estimated that approximately 60% of patients with melanoma metastatic to the small bowel dies; however, from this rate, only 1.5 to 4.4% of metastases are diagnosed before death. This is because many patients carry the disease undetectably at the early stages, and

diagnosis is given when complications or even death occurs [8].

Although intussusception is common in the pediatric population, it is rare in adults, considering it is responsible for only 1% of the cases of intestinal obstruction. Unlike in children, intussusception is rarely idiopathic in adults and treatment mostly requires surgical intervention. The clinical presentation of intussusception in adults is variable, and so it may not be suspected on imaging examination. The preoperative diagnosis is difficult to determine, considering it is only given at laparotomy [9].

Although it is important to differentiate primary disease from metastatic disease, it may be difficult or even impossible to establish the exact origin (primary or secondary) of the small bowel melanoma. In some patients with mucosal melanoma metastasis, it is difficult to identify a precedent or a coexistent primary lesion [8]. The criteria for diagnosing primary melanoma include the absence of another site of primary melanoma and the absence of previous history of removing either melanoma or atypical melanocytic nevi in the skin, retina, anal canal, or occasionally in other locations such as esophagus, penis, and vagina [10].

The small bowel melanoma may be metastatic in cases of cutaneous, anal, or ocular melanoma. The most typical metastatic lesions in the intestine are melanomas, but the occurrence of intussusception from melanoma is rare [11]. There are only six cases being reported in the English medical literature to date [12].

The risk factors for the propagation of malignant melanoma to the gastrointestinal tract include superficially disseminated melanoma, axial primary tumor, Clark level III or IV, high degree of histological regression, presence of ulceration, and high mitotic rate [13].

A study performed in the Memorial Sloan Kettering, in the late 60 s, reported the following incidence of gastrointestinal melanoma metastases: liver 68%, small bowel 58%, colon 22%, stomach 20%, duodenum 12%,

rectum 5%, esophagus 4%, and anus 1%, based on the review of several autopsies [14].

Although gastrointestinal melanoma metastasis is more common in the liver, the favorite place for intussusception from metastatic melanoma is the small bowel, and the jejunum and ileum regions are equally prevalent. Metastases usually develop in 3 to 6 years after surgically removing the primary melanoma, but in some cases, they may be present at both the initial diagnosis and 6 months after the primary lesion [8].

Data from the archives of the Armed Forces Institute of Pathology in the United States of America allowed Bender and colleagues to define four different types of metastatic melanoma of the small bowel: cavitory, infiltrating, exoenteric, and polypoid. These four types are not always distinct. The metastatic melanoma of the small bowel typically forms multiple polypoid masses in the submucosal region. The presence of ulceration in these masses is common and it is rare for them to appear as a single mass in the intestine [8].

The investigation starts with anamnesis and a detailed clinical examination, but the preoperative diagnosis of small bowel metastasis from malignant melanoma is very difficult due to the late appearance and nonspecific symptomatology [15]. In order to complement the investigation, upper digestive endoscopy, colonoscopy, opaque enema, and computed tomography may be performed [16]. Endoscopically, the lesions may be nodules of ulcerated melanomas appearing in normal folds or as submucosal masses with ulcerated cores. The biopsies removed from the edges of malignant ulcerations are positive in 90% of cases [17].

The site of hematogenous deposition of neoplastic cells influences the radiological aspect of metastases. Depositions in the submucosal layer, for instance, result in intraluminal mass; depositions in the serous layer result in implants that, according to mass growth, may progressively compress the adjacent intestine; and depositions in the mesentery result in masses that, when extremely voluminous, cause significant

compression in the small bowel [18].

Traditionally, the potential for metastatic melanoma of the small bowel has been usually considered when a "target lesion" is shown in imaging studies of the small bowel [19]. However, recent studies suggest that melanoma should be included in the differential diagnosis when cavitory, infiltrating, or polypoid lesions are observed in the small bowel [20]. The small bowel intussusception secondary to the metastatic lesion from melanoma has already been described in the literature with variable presentation [21]. The typical sign of intussusception is one segment of intestinal loop "telescoping" within another [22].

Invariably, the surgical intervention on lesions metastatic to the gastrointestinal tract is reserved for symptomatic lesions that produce obstruction or bleeding. Patients with visceral melanoma metastases are usually treated with systemic chemotherapy, and the mean survival of such patients is only five to 11 months, depending on the site of metastasis [23].

Although patients with gastrointestinal melanoma metastases have a poor prognosis, most of them can have a good palliation of symptoms through surgical resection, with minimum morbidity and mortality. Survival rates of 28% have been reported at five years after surgical resection of gastrointestinal melanoma metastases in patients with successful excision of the primary lesion [24].

4. Conclusions

Although small bowel metastasis from melanoma is common, the intussusception from melanoma is rare. The preoperative diagnosis is difficult due to the nonspecific symptomatology and it should be reminded whenever there is a previous melanoma diagnosis, especially when the biopsy result show the superficially disseminated primary melanoma, axial primary tumor, Clark level III or IV, high degree of histological regression, presence of ulceration, or high mitotic rate.

When intussusception occurs, the metastasis should

be resected to prevent complications such as perforation or obstruction and to provide, as soon as diagnosed, an adequate treatment to the patient, improving quality of life, symptomatology, and prognosis.

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