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Rapid progression of a pregnancy-associated intra-abdominal desmoid tumor in the post-partum period: A case report



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ABSTRACT

INTRODUCTION: The clinical behavior of desmoid tumors can be unpredictable, particularly when they arise in the ante-partum or post-partum period. We present a case of an intra-abdominal desmoid tumor that was identified in the ante-partum period, progressed rapidly in the post-partum period, and was subsequently resected.

PRESENTATION OF CASE: The patient is a 19 year-old female who was found to have a 12 cm intra-abdominal mass on a fetal assessment ultrasound. The decision was made to observe the patient and monitor the mass for growth. However, the mass rapidly grew in the post-partum period. The patient was transferred to our institution after an exploratory laparotomy revealed a large intra-abdominal mass emanating from the small bowel mesentery. The 30 cm × 24 cm × 16 cm mass was successfully resected with negative margins, and the pathologic diagnosis of desmoid tumor was confirmed. The patient had an uncomplicated post-operative course and was discharged on post-operative day 6.

DISCUSSION: The majority of pregnancy-associated desmoid tumors are in the abdominal wall, arising from the rectus abdominus muscle or from previous Cesarean section scars. These tumors may spontaneously regress in the post-partum period and therefore, patients with these tumors are often observed. Close follow-up is important so that rapid tumor progression, which may lead to unresectability, can be identified and managed appropriately.

CONCLUSION: A patient with a rare case of a giant pregnancy-associated, intra-abdominal desmoid tumor that rapidly progressed in the post-partum period and was successfully treated with surgical resection with negative margins.

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1. Introduction

Desmoid tumors are rare benign tumors exhibiting fibroblastic proliferation that typically arise from fascial or musculo-aponeurotic structures [1]. They account for 0.03% of all documented neoplasms and approximately 3% of all soft tissue tumors, with an incidence of 2–4 per million per year [2]. These tumors have a propensity to infiltrate surrounding structures, rather than metastasize, and have the potential to cause significant

morbidity and mortality due to pressure effects and obstruction of vital organs. According to consensus-based guidelines from the National Comprehensive Cancer Network (NCCN), surgical resection should be attempted if technically feasible in symptomatic patients or in patients with tumors that pose imminent risk to surrounding structures [3]. However, the clinical progression and behavior of these tumors can be unpredictable, particularly when they occur in the ante-partum or post-partum period in women.

We present a case of a giant intra-abdominal desmoid tumor that rapidly progressed in the post-partum period and was successfully resected.

2. Presentation of case

The patient is a 19 year old female who was found to have an intra-abdominal mass that measured 12 cm in greatest dimension during a fetal assessment ultrasound which was performed at 34

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weeks gestation. The patient was asymptomatic and the decision was made to proceed with observation. The patient had a successful spontaneous vaginal delivery without complications in December 2015. In the first post-partum week, the patient continued to report shortness of breath and abdominal pressure. A computed tomography (CT) scan was performed on December 31, 2015 which showed that the mass measured 14 cm × 13 cm × 13 cm and appeared to be attached to the uterus, suggestive of an exophytic fibroid. Again the decision was made to continue with observation.

Over the next 3 months, the patient's abdominal girth continued to increase. A follow-up MRI on April 1, 2016 showed that the mass had increased significantly in size, measuring 23 cm × 22 cm × 13 cm, and was potentially consistent with a desmoid tumor. The patient's only symptoms were increasing shortness of breath and abdominal pressure; she did not experience any change in bowel habits. The patient had no prior medical history or surgical history, and no family history of a similar condition, colorectal cancer, or familial adenomatous polyposis (FAP).

The patient was transferred to our institution with a negative pressure abdominal wound dressing on the evening of May 13, 2016 immediately after exploratory laparotomy at an outside hospital revealed a normal uterus and ovaries and a large intra-abdominal mass that was emanating from the small bowel mesentery. A CT angiogram of the abdomen and pelvis was performed to evaluate the vascular distribution to the mass and the small bowel. This showed a large mass filling the entire abdomen and displacing the bowel with a vascular supply from the superior mesenteric artery (SMA) (Fig. 1A–C). The proximal branches of the SMA supplying the small bowel and colon appeared to be separate from the mass.

The patient was taken to the operating room for surgical exploration. This demonstrated that the mass originated from the distal small bowel mesentery and a loop of small bowel was adherent to and inseparable from the mass (Fig. 2A). The small bowel was examined and it appeared that the patient would have sufficient small bowel remaining after resection. The mass was successfully resected en bloc with part of the ileum. A small portion of the terminal ileum, the ileocecal valve, and the middle colic artery and vascular supply to the proximal small bowel were preserved. The mass measured 30 cm × 24 cm × 16 cm (Fig. 2B). Pathology confirmed the diagnosis of desmoid tumor with negative surgical margins. (Fig. 3) The tumor was negative for estrogen and progesterone receptor. The patient had an uncomplicated post-operative course and was discharged on post-operative day 6.

On multidisciplinary review of the patient's case, adjuvant radiation therapy was not recommended due to the documented negative margins and tumor location [3]. The patient had no complaints at a 3-month follow-up clinic visit, and upper and lower endoscopic studies done at that time showed no evidence of polyps. Per NCCN guidelines, the patient will be followed with history, physical exam and imaging every 6 months for 2 years [3].

3. Discussion

Desmoid tumors are rare tumors that can occur anywhere in the body and have quite variable clinical behavior. The majority of desmoid tumors arise sporadically, while 5–15% are associated with familial adenomatous polyposis (FAP) [4,5]. Up to 20% of patients with FAP will develop a desmoid tumor, most which occur intra-abdominally or in the abdominal wall and arise at the site of prior surgical anastomoses or incisions. Sporadic desmoid tumors are more commonly located in the abdominal wall or at extra-abdominal locations, have a lower mortality rate than those associated with FAP, and have a higher rate of spontaneous regression [4,6]. Small retrospective studies have shown spontaneous regression rates of up to 29% in patients with sporadic desmoid



Fig. 1. (A–C): CT angiogram of abdomen and pelvis demonstrating a large intra-abdominal mass with vascular supply from the superior mesenteric artery.

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