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Case Report

Emergency Angioembolization for Life-Threatening Hemorrhage in Wilms Tumor

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Abstract

Introduction: Renal artery embolization has been used in a palliative fashion for symptomatic relief of hematuria or flank pain in unresectable renal cell carcinoma in adults. There is limited data on the use of embolization for actively bleeding and unresectable tumors in the oncological pediatric population. **Case Description:** A previously healthy 5-year-old boy with no significant past medical or surgical history presented to the clinic with gradually worsening abdominal distension associated with occasional abdominal pain, gross hematuria, and lethargy for four months. Diagnostic investigations showed an 18-cm left-sided metastatic (pulmonary) renal tumor (Wilms), which was deemed unresectable on imaging. Treatment was planned according to the SIOP-RTSG protocol. However, he became hemodynamically and vitally unstable with acute, sudden distension of the abdomen on the left side after the first cycle of chemotherapy. Imaging showed active bleeding from an inferior branch of the left renal artery. Selective angioembolization was done, and chemotherapy was reinitiated with a patent left main renal artery. Following the fourth cycle of chemotherapy, he developed hemodynamic instability and abdominal pain; imaging revealed the resolution of pulmonary nodules and bleeding from the left renal artery (main); this was again embolized, and the patient was stabilized. The patient was operated on after optimization, and a complete resection of the mass was done with negative margins. On six months follow-up, he is well. **Practical Implications:** To the best of our knowledge, this is the first case where angioembolization has been done in conjunction with neoadjuvant chemotherapy to downsize a Wilms tumor to achieve favorable outcomes. Continued research efforts are necessary to optimize strategies and improve the prognosis for pediatric patients, and this case is one of the prime examples.

Keywords: Angioembolization, hemorrhagic tumor, intervention radiology, pediatric surgery, Wilms tumour

Introduction

There are several etiologies of internal solid visceral hemorrhage, with trauma and neoplasm being the most common. Embolization can be an effective treatment for active bleeding, which can be a life-threatening event. Ten percent of adult patients with advanced cancer experience tumor-related

hemorrhage.[1] Most case reports on embolization of bleeding tumors refer to those involving the head and neck, pelvis, lung, liver, or gastrointestinal tract. While embolization in trauma cases and as a preoperative procedure to minimize blood loss in surgery has been widely reported in the pediatric population, there is limited data on embolizing an actively bleeding tumor.

The role of angioembolization in treating trauma is widely studied. However, the data is limited for patients with tumors, with only a few cases reported over the years, all managed in variable ways.[1-4] This case study reports a case of a child with a Wilms tumor who presented with a life-threatening hemorrhage, undergoing management based on the International Society of Pediatric Oncology-Renal Tumor Study Group (SIOP-RTSG) Umbrella Protocol 2016 and the importance of hemodynamic stabilization before surgery to achieve a favorable outcome.

Case Description

A 5-year-old boy, previously healthy, with no significant past medical or surgical history, presented to the walk-in clinic with gradually worsening abdominal distension associated with occasional abdominal pain, gross hematuria, and lethargy for 4 months.

The parents reported that for the last 2–3 months, the child had been pale, lethargic, and constipated, with a decrease in appetite along with mild abdominal pain. Furthermore, they observed that while he remained lean, his clothes fit more tightly over the abdomen.

On examination, his height and weight were below the 50th percentile. Moreover, he had pallor, and his vitals showed tachypnoea (36 breaths/min, with oxygen saturation of 95% on room air), tachycardia (121 beats/min), normal blood pressure, and temperature.

Further examination showed bilateral equal air entry on chest auscultation and an asymmetrically distended abdomen with a bulge on the left side. It was a firm, fixed mass, approximately 18 × 14 cm, with vague borders. There were no other significant findings.

Diagnosis and management

After the initial examination, the child was admitted for workup and optimization, and his blood work and imaging were performed. He was found to have a hemoglobin level of 10.2 g/dL, 557 platelets, and a white count of 11.92 × 10^9/L. The renal function tests were within normal limits, and a computed tomography (CT) scan of the abdomen showed a 12 × 13.5 × 18 cm left renal mass partially encasing the aorta and pushing abdominal vasculature, and bowel loops medially, adherent to the tail of the pancreas and splenic vasculature [Figure 1]. Furthermore, there were bilateral multiple pleuropulmonary soft tissue density nodules (the largest measuring 4.7 mm).

It was planned to initiate neoadjuvant chemotherapy (combined vincristine, actinomycin D, and doxorubicin) based on the SIOP-RTSG UMBRELLA protocol in 2016.[5] Following the first cycle of chemotherapy, the child presented to the emergency room with Class III hemorrhagic shock and a 4-g drop in hemoglobin. After stabilization and resuscitation, he underwent a CT-scan of

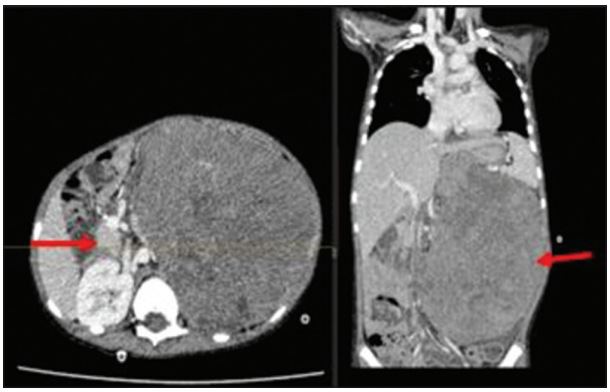

Figure 1: Contrast-enhanced computed tomography scan abdomen showed a 12 × 13.5 × 18 cm left renal mass, partially encasing the aorta and pushing abdominal vasculature and bowel loops medially, adherent to the tail of pancreas and splenic vasculature

the abdomen angiogram, which showed active bleeding and extravasation of contrast from a lower pole branch of the left renal artery. Since the tumor was adherent to the pancreas and spleen and the child was in a decompensated phase, a multidisciplinary decision was taken, and the child underwent selective angioembolization of the bleeding vessel.

The child responded well and stabilized. He then underwent chemotherapy as per the protocol [Figure 2]. The selective nature of angioembolization allowed continued chemotherapy via the left renal artery (main). At the end of 4 weeks of chemotherapy (combined vincristine, actinomycin D, and doxorubicin), on day 5, he developed an acute worsening of abdominal distension. He was brought to the emergency room, where he was found to have a Class II hemorrhagic shock. He was resuscitated and underwent another CT-scan of his chest and abdomen with an angiogram. This showed resolution of the pulmonary nodules, an interval decrease in size and necrosis in the left renal mass (now measuring 10.6 × 11.7 × 15.1 cm), and now visible planes visible between the aorta and splenic vessels, along with an active bleed from the left renal artery adjacent to the previously coiled branch. He underwent angioembolization of the left main renal artery with a good response to resuscitation efforts [Figure 3]. Once stabilized, he underwent an emergent left radical nephrectomy with aortocaval lymph node sampling (40 h after the angioembolization).

Intraoperatively, he was found to have a 15 × 12 cm left renal mass densely adherent to the diaphragm and the left mesocolon, overlying the aorta and inferior vena cava. The surgery team was able to resect it *en bloc*. Following surgery, he remained well, followed a routine postoperative course, and was discharged in stable condition on the 3rd postoperative day.

Histopathology showed a 15 × 10 cm mass (870 mg) Residual Wilms' tumor with 50% viable epithelial component, mixed type (45%, blastemal

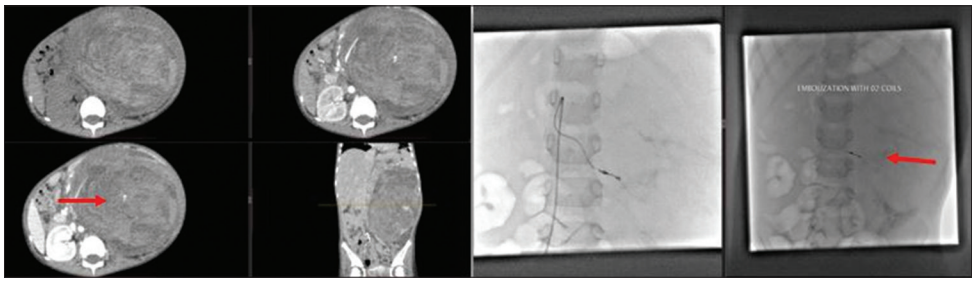

Figure 2: Computed tomography scan abdomen (angiogram) showing high-density haemorrhage within the tumour with active extravasation of contrast, followed by selective angioembolization of the causative vessel, the lower branch of the left renal artery

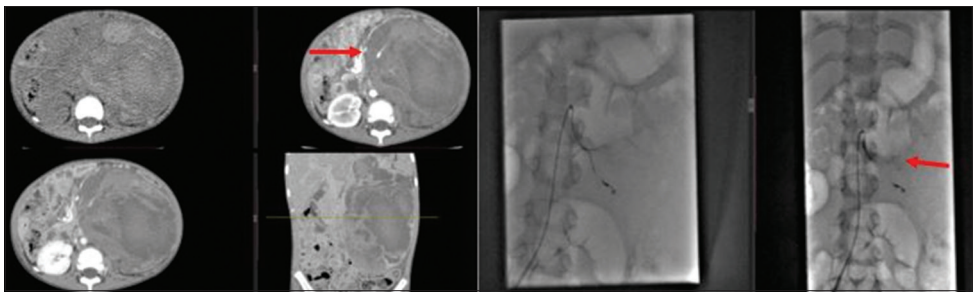

Figure 3: Computed tomography scan abdomen (angiogram) showing high-density hemorrhage within the tumor with active extravasation of contrast, followed by angioembolization left renal artery

40% and mesenchymal 15%), intermediate risk. The viable tumor involved renal sinus and perirenal fat. It was labeled SIOP Stage-II. The ureteric, vascular margins and left adrenal gland are free of tumor.

He then received the remaining two protocol-based preoperative chemotherapy doses in a postoperative setting (7th postoperative day), followed by the completion of postoperative chemotherapy based on staging and histopathology (AV 1 AVD 150 mg/m^2/day as per SIOP-RTSG Umbrella Protocol 2016; combined vincristine, actinomycin D, and doxorubicin). Six months following the completion of treatment, the child is stable, disease-free, and school-going.

Discussion

In adults, renal artery embolization is frequently employed to treat kidney disorders. In the case of renal cell carcinoma, it is applied before surgery to reduce intraoperative blood loss and the volume of tissue that needs to be excised.[6] Renal artery embolization has been employed palliatively to alleviate symptoms such as hematuria or flank pain in cases of unresectable renal cell carcinoma.

Further indications for utilizing renal artery embolization include conditions including renal angiomyolipoma, kidney damage due to abdominal trauma, iatrogenic injury, renal artery aneurysm, or arteriovenous malformation. Internal hemorrhage can stem from various causes, with trauma and neoplasms being particularly prevalent. Embolization proves to be a viable and effective intervention for managing active bleeding, a potentially life-threatening occurrence. Tumor-related bleeding occurs in approximately 10% of adults diagnosed with advanced cancer.[1]

Although the use of embolization in trauma situations and as a preoperative measure to reduce blood loss during surgery has been extensively documented in the pediatric population, there is limited information and published literature available regarding the embolization of an actively bleeding tumor.

A pediatric case report highlighted the successful application of renal artery embolization in addressing hematuria associated with a malignant rhabdoid tumor of the kidney.[2] Another study reported that six pediatric patients with malignancies (three with hemorrhagic cystitis, two with procedure-related internal iliac artery injuries, and one with a tumor rupture) underwent arterial embolization. Five of these patients were successfully treated without complications.[2]

Wilms tumor and embolization have also been previously reported four times. The first case used angioembolization to control severe hematuria following biopsy.[3] Similar to the present case, tumor-feeding arteries were selectively embolized in the second case. As opposed to the entire renal artery, with the intent to enable chemotherapy to downstage the tumor. However, the child developed colonic obstruction secondary to ischemia and underwent tumor resection in 48 h.[4] The third case reported emergency embolization of the main renal artery to control hemorrhage. They stabilized the patient and performed resection under the same general anesthesia.[7] A more recent case by Ruff *et al*. reported a similar case where the renal artery was embolised to control bleeding and stabilize the patient, followed by an upfront nephrectomy several days later on the elective list with minimal intraoperative blood loss.[8]

To our knowledge, this is the first time that angioembolization has been used in conjunction with neoadjuvant chemotherapy to downsize a pediatric patient with Wilms tumor to achieve favorable outcomes and resectability. Continued research efforts are necessary to optimize strategies and improve the prognosis for pediatric patients, and this case is one of the prime examples.

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Author Contributions

Conceived and designed the analysis: AS, TL; Collected the data: AS; Contributed data or analysis tools: AS, TL, ID; Performed the analysis: AS; Wrote the paper: AS, SA, MAS, TL, ID.