Journal of Pediatric Surgery Case Reports 30 (2018) 68–73

Contents lists available at ScienceDirect

Journal of Pediatric Surgery Case Reports

journal homepage: www.elsevier.com/locate/epsc

The unique characteristics of intussusception after renal tumor surgery in children

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ARTICLE INFO

Keywords:
Postoperative intussusception
Wilms tumor
Nephroblastoma
Tumornephrectomy
Renal tumor surgery
Postoperative complication
Pediatric tumor

ABSTRACT

Introduction: To further optimize survival rates as well as quality of cure for pediatric kidney tumors, attention for treatment related morbidity and mortality has become increasingly important. Intussusception is a rare but important complication after tumornephrectomy in children, causing morbidity, mortality and prolonged hospitalization. In this study, we describe two recent cases in our institute and provide a comprehensive review of the literature.

Methods: For our narrative review, we searched for all reported cases of post tumor nephrectomy intussusception published until November 2016, using Pubmed and Embase libraries.

Results: A total of 52 pediatric renal tumor cases who developed intussusception after tumor nephrectomy were identified. Median age was 23 months (range 3–84). Median time of onset was postoperative day 6 (range 1–18). Of 41 patients described in detail, only 4/41 were ileocolic, the others suffered from a small bowel intussusception. Most frequent presenting symptom was bilious vomiting. Preceding treatment approach was documented in 47 cases; i.e. preoperative chemotherapy had been administered to 10/47 patients. In 29 of 30 well documented cases, successful manually reduction during re-laparotomy was described and only 1 patient needed resection. All patients survived without recurrence of intussusception.

Conclusion: In pediatric renal tumor patients, small bowel obstruction seems to reflect mostly post nephrectomy intussusception cases in contrast to the ileocolic idiopathic intussusceptions that are observed in healthy children. Symptoms of intussusception mimic chemotherapy related toxicity and general post-surgical symptoms, thereby initiating a significant delay in diagnosis. Awareness of intussusception after renal tumor surgery is warranted.

1. Introduction

Pediatric kidney tumors occur mainly in children under the age of 5 years thereby representing around 6% of all pediatric tumors [1]. The most common subtype is Wilms tumor (WT). Survival rates for children with Wilms tumor have increased up to 90–95% over the past decades. Consequently, to further optimize survival rates as well as quality of cure, attention for treatment related morbidity and mortality has become increasingly important. Radical tumor nephrectomy is an important component of pediatric renal tumor treatment. Complications following tumor nephrectomy include postoperative bowel obstruction that can be caused by adhesions or, (more rarely) intussusception [2,4–7]. Extensive research is pursued on postoperative adhesions after tumor nephrectomy in children and adults, however to date, only a few reports address intussusception after nephrectomy for pediatric renal tumors and a comprehensive review is lacking. Consequently, the frequency and the determinants of development of intussusception is unclear. In addition, intussusception symptoms can be disguised, as they mimic general post-surgical symptoms, and chronic chemotherapy related side effects such as nausea. This could potentially lead to delay in recognizing bowel obstruction in this clinical setting. Hence, it is important to recognize the rare event of intussusception after tumor nephrectomy and to be aware of its provoking determinants. We reviewed all available cases in order to identify possible determinants of such a condition, including a literature review of well documented cases as well as two recent patients in our institute.

2. Cases

Two patients in the Princess Máxima Center for pediatric oncology, diagnosed with a renal tumor, developed small bowel intussusception within 30 days after tumor nephrectomy. Both patients had received

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https://doi.org/10.1016/j.epsc.2017.12.016
Received 15 December 2017; Accepted 20 December 2017
Available online 21 December 2017

primary chemotherapy for localized stage renal tumor according to the SIOP 2001 protocol. Data on clinical presentation, patient characteristics, surgery, histopathology, treatment and outcome were collected from their medical records.

3. Case 1

A 5-years old male presented with a history of hematuria and abdominal pain. Ultrasound revealed a right sided kidney mass with a volume of 380 cm^3. Treatment with 4 weeks of preoperative chemotherapy according to SIOP 2001 (vincristine and actinomycin-D), preceded tumor nephrectomy. Three days after surgery, the patient suffered from bilious vomiting without passing stools and one day later, bowel sounds became impaired. Abdominal x-ray, performed on day 4 after surgery, showed a small bowel obstruction. Subsequent ultrasound of the abdomen confirmed the small bowel obstruction, however the cause of the obstruction was not seen. Relaparotomy showed an evident ileoileal intussusception without a leading point, which was remedied by manual reduction during relaparotomy. Recovery was slow but unremarkable, and the child was discharged on day 9 after tumor nephrectomy. Histology of the tumor revealed nephroblastoma. Follow up was unremarkable, without recurrence of intussusception.

4. Case 2

An 8 months old boy was diagnosed with a localized centrally located left kidney tumor with a volume of 29 cm^3 while being treated for urine tract infection. The patient was preoperatively treated according to the SIOP 2001 protocol. On the first postoperative day, the patient vomited and developed a distended abdomen and did not pass any stools. Abdominal X-ray showed a small bowel obstruction. Subsequent ultrasound of the abdomen confirmed a small bowel intussusception in the right upper quadrant. During relaparotomy an illeoileal intussusception of approximately 10 cm was found which was manually reduced. Further recovery was unremarkable, and the patient was discharged on day 5 after tumor nephrectomy. No recurrence episode occurred during the follow up time. Histology of the tumor later revealed a metanephric adenofibroma and no further therapy was administered.

5. Methods

We systematically searched for all reported cases of post tumor nephrectomy intussusception in literature in PubMed and Embase until November 2016. The search terms were: (child *OR* boy *OR* girl *OR* infant *OR* pediatr* *OR* paediatr*) AND (tumor nephrectomy *OR* nephrectomy *OR* surgery *OR* renal tumor surgery) AND (tumor, wilms'[MeSH Terms] *OR* bilateral wilms tumor[MeSH Terms] *OR* tumor, wilms[MeSH Terms] *OR* cancer, renal cell[MeSH Terms] *OR* renal cell carcinoma[MeSH Terms] *OR* renal cancer[MeSH Terms]) AND (complication, postoperative[MeSH Terms] *OR* complications, postoperative[MeSH Terms] *OR* intussusception[MeSH Terms] *OR* invagination, intestinal[MeSH Terms] *OR* intestinal obstruction[MeSH Terms] *OR* surgical complication *OR* complication).

Cross reference check was performed to identify additional publications of interest. All selected titles and abstracts were explored for relevance to the subject and full articles were analyzed. All studies written in English reporting patients undergoing tumor nephrectomy for renal tumors were included. In case two or more articles described the same patient cohort, the most recent or relevant article was included.

6. Literature review

Literature review identified 225 papers of which we selected 20 on the basis of full text assessments, using the following inclusion criteria: patients who underwent tumor nephrectomy (1) and developed intussusception within 30 days of surgery (2) (Fig. 1). Fifty postoperative intussusception cases after tumor nephrectomy in children in the literature, and 2 local cases, were summarized (Table 1). All cases revealed a nephroblastoma, with the exception of one of our patients. Median age (data available in 25/52 cases) was 23 months (range 3–84 months, Fig. 2a) and 10/25 patients were younger than 1 year. There was an equal distribution of gender. Data on preoperative chemotherapy were available in 47/52 patients (10/47 patients had preoperative chemotherapy). Site of tumor was available in 16/52 cases (left n = 8 and right n = 8), NSS surgery had been performed in 2/52 patients. No cases of tumor rupturation were described. Only 1 patient had received postoperative radiotherapy, before onset of symptoms of

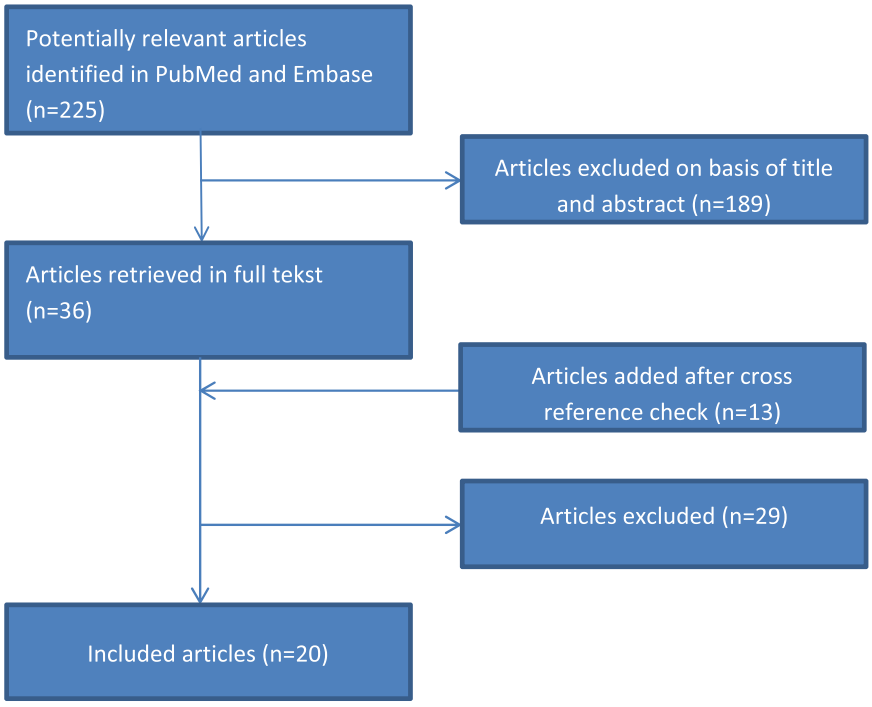

Fig. 1. Flow chart: selection of articles on intussusception after pediatric renal tumor nephrectomy for this narrative review.