Intractable hiccup due to giant hydronephrosis: A rare case report and literature review

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Abstract

INTRODUCTION: Giant hydronephrosis (GH) is a rare disease that is found in adult patients. Although there are some common symptoms associated with hydronephrosis, such as surrounding organ compression, its rarer symptoms can render diagnosis very difficult, and treatment should also vary according to the cause.

PRESENTATION OF CASE: We here report an 82-year-old man who was admitted to the hospital for repeated intractable hiccups. After B-ultrasound and CT examination, the patient underwent laparoscopy surgery, which was converted to open nephrectomy, and the patient’s intractable hiccup symptoms disappeared.

DISCUSSION: GH is a rare disease, and its symptoms are diverse. The unusual symptoms of cystic hypertonic compression of surrounding organs, such as intractable hiccups, should be taken into account. GH is mainly diagnosed via ultrasound examination and CT scan. The choice of treatment for GH needs to be based on the etiology and renal function of hydronephrosis, and consider malignant lesions.

CONCLUSION: Giant hydronephrosis can present rare symptoms as “intractable hiccups”. The selection of treatment should be made depending on the cause.

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1. Introduction

Hydronephrosis is a common clinical condition that is often caused by obstruction of the ureteropelvic junction, but giant hydronephrosis (GH) is rare, especially in adults. In adults, GH is defined by Sterling firstly in 1939 as the presence of more than 1 L of fluid in the renal pelvis, or kidney occupying the hemiabdomen across the midline [1].

GH may present with vague symptoms, including increased abdominal girth, nausea, fatigue, indigestion, and loss of weight, but, to the best of our knowledge, there have been no reports of intractable hiccups due to GH. We here present a rare case of an 82-year-old male patient who suffered from intractable hiccups due to GH, along with a review of the past decade. This case has been reported in accordance with the surgical case report guidelines (SCARE) criteria [2].

2. Case report

An 82-year-old man complained of a gradual increase in his abdominal girth over the past two years and of abdominal distension and intermittent nausea for the past six months. He was admitted to the hospital for repeated intractable hiccups having lasted two months. The patient had a history of intestinal necrosis due to an intestinal obstruction 25 years earlier and of occlusion of the inferior vena cava filter and left common iliac vein stent placement due to left common venous thrombosis five years earlier. Physical examination showed the patient was of average build with a thin, anemic appearance, and his vital signs were stable. Cardiopulmonary examination produced normal results. An abdominal examination revealed a longitudinal surgical scar in the middle of the upper abdomen. The swelling of the mass was evident on the left side of the abdomen (Fig. 1). Palpation showed the abdomen to be soft but not tender, and bowel sounds were normal. Ultrasonography and computerized tomography (CT) showed the presence of a massive hypoechoic lesion occupying almost the entire abdomen. This lesion exerted pressure on the stomach, liver, pancreas, and spleen. There was a high-density shadow visible about 1.5 cm from the middle pole of the medial edge of the mass (Figs. 2 and 3). An isotope kidney scan showed poor kidney function and only 5% normal renal function in the left kidney.

After administration of two units of homologous red blood cells, the patient underwent what was initially laparoscopy and then open simple nephrectomy. Adhesion between the kidney and surrounding organs was evident during the procedure. About 7.5 liters of hydronephrosis fluid were drained. The left kidney and upper ureter were successfully removed (Fig. 4). The patient was diagnosed with GH caused by obstruction from a kidney

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stone at the junction of the ureter and renal pelvis, accompanied by renal cell transitional cell carcinoma (T2N0M0) (Fig. 5). The patient’s intractable hiccup symptoms disappeared, and his condition and appetite recovered after the operation. During the six-month follow-up, the patient did not experience hiccups.

3. Discussion

GH is a relatively rare urinary system disorder that can occur in patients of any age, but it is extraordinarily rare in adult patients [3]. It is generally defined as hydronephrosis of 1 L in an adult or 1.5% of total body weight in a child [1,3]. Quantities of fluid in the renal pelvic system exceeding 2 L have only rarely been reported [4]. We here report an 82-year-old case of massive hydronephrosis and review the clinical features of cases of large hydronephrosis involving more than 2 L of fluid in the last decade in Table 1. GH is generally thought to develop over time, so clinical symptoms are usually not noticeable. The hydronephrosis is gradually increased to a certain extent, and it contributes to surrounding organs having compressed symptoms. Causes of GH may include UPJ obstruction, renal pelvic-ureteric calculi, pelvic-ureteric tumors, trauma, obstruction of the outlet, and obstruction of the PUJ from cross-fusion kidney or ectopic kidney [5–8].

GH progresses slowly. As described in the literature, the associated massive abdominal mass or abdominal swelling may cause pain, hematuria, recurrent urinary tract infections, or other symptoms or complications described in the literature, including nausea, fatigue or indigestion, urinary tract infection, weight loss, renal insufficiency, and even kidney breakdown (Table 1). According to the literature, GH may also, though rarely, involve severe post-traumatic hematuria, intestinal compression symptoms, gastric obstruction, or respiratory distress [5,9,10]. A review of cases of GH involving over 2 L published during the past decade showed
that the dominant symptom had been massive hydrenephrosis of surrounding organs [5].

As far as we know, no previous reports have shown any case of hiccups caused by massive hydrenephrosis. Hiccups are an abnormal respiratory movement, a diaphragmatic spasm that occurs mainly because of vagus nerve reflex or direct stimulation of the phrenic nerve and diaphragm, which causes the diaphragm and intercostal muscle to involuntarily and synchronously contract. This instantly produces strong inspiratory movement. Intractable hiccups is defined if the attack lasts more than 1 month, and there are various factors that can cause intractable hiccups, including metabolic abnormalities, psychogenic diseases, malignant tumors, central nervous system pathology, medications, pulmonary disease, and gastrointestinal conditions [11]. In this case, the patient showed no central nervous system disease that could cause intractable hiccups. The most common symptoms were gastrointestinal and caused by massive hydrenephrosis. One possible cause of intractable hiccups is that huge hydrenephrosis oppresses the surrounding organ tissues. The compression of the neighboring gastrointestinal tract and diaphragm muscle simulates the vagus nerve and the phrenic nerve, causing hiccups. The patient’s intractable hiccups symptoms did not show significant relief after treatment with various drugs, and they disappeared immediately after nephrectomy.

GH is mainly diagnosed via ultrasound examination and CT scan [12,13]. In most cases, the differential diagnosis between GH and other posterior abdominal cystic structures is still difficult, especially in patients with large hydrenephrosis. The basic structure of the kidney disappeared, and, because of its large size, the hydrenephrosis exerted squeezing pressure all around the organ. A review of the literature on GH over the past decade showed that only 30% (4/13) of cases could be clearly diagnosed before surgery (Table 1). As shown in this case report, a coronal-view

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**Table 1**

List of cases of GH reviewed in the literature over the past decade.

| Case no. | Gender | Age | Cause | Pre-operative diagnosis | Symptoms | Diagnosis methods | Treatment and drainage volume | Author |
|---------|--------|-----|-------|-------------------------|----------|-------------------|-------------------------------|--------|
| 1       | M      | 17  | PUJ obstruction | Clear | Flank pain, abdominal swelling | Ultrasonography, contrast-enhanced CT | PCN, nephrectomy 4L | Ashish Sharma |
| 2       | M      | 49  | PUJ obstruction | Unclear | Malaise, suprapubic pain | Ultrasonography, CT, Radionuclide scan | Laparoscopic nephrectomy 3L | Pawel Obrocki |
| 3       | M      | 45  | PUJ obstruction, trauma | Unclear | Flank pain, fullness, gross hematuria | Ultrasonography, contrast-enhanced CT | Open pyeloplasty 7.5L | Ashok Kumar Sokhal |
| 4       | F      | 78  | Renal pelvic carcinoma, ureter stone | Unclear | Gross hematuria | Ultrasonography, contrast-enhanced CT | Open Nephroureterectomy | Tomohiro Wakamiya |
| 5       | F      | 18  | PUJ obstruction | Unclear | Mild abdominal pains | Ultrasonography CT | PCN 7.5L | Qi-FEI WANG |
| 6       | M      | 20  | PUJ obstruction | Unclear | Abdominal pain | Ultrasonography CT | Open nephrectomy 8L | Guanghi Hu |
| 7       | M      | 83  | Ureteral stone | Unclear | Abdominal flank pain | Ultrasonography CT | PCN 4L | Yalcin Golcuk |
| 8       | M      | 47  | PUJ obstruction | Unclear | Intestinal occlusion, abdominal distension | Abdominal X-ray CT | Nephrectomy 7.8L | Issam Yazough |
| 9       | F      | 31  | PUJ obstruction | Unclear | Pelvic cystic mass | Ultrasound, MRI | Nephrectomy 6L | Lin YJ |
| 10      | M      | 55  | Ureteral stone, tumor renal pelvis | Unclear | Gross hematuria, left shoulder pain | Enhance CT | Radical nephrectomy 7.8L | Kimura R |
| 11      | M      | 40  | Obstructing renal calculus | Clear | Abdominal pain, nausea, vomiting | CT | PCN 7L | Grover CA |
| 12      | M      | 45  | Ureteral stone | Clear | Abdominal distension, nausea | CT | Nephrectomy 5L | Chia-Chao Wu |
| 13      | M      | 62  | Renal pelvic tumor | Clear | Abdominal fullness, pain | Ultrasonography, CT, MRI | PCN 7L | Manuyama T |

M = Male; F = female; PUJ = Pelvic-ureteric junction; CT = Computerized tomography; MRI = magnetic resonance imaging; PCN = percutaneous nephrostomy. Abbreviations: M = Male; F = female; PUJ = Pelvic-ureteric junction; CT = Computerized tomography; MRI = magnetic resonance imaging; PCN = percutaneous nephrostomy; UPJ = ureteropelvic junction.

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**Fig. 5.** Pathological section from the GH showing an invasive papillary epithelial carcinoma (H&E staining, magnification, x200). H&E, hematoxylin and eosin.
CT examination of the patient revealed a high-density shadow of approximately 1.5 x 1.0 cm in the middle pole of the medial margin of the cyst (Figs. 2 and 3). The diagnosis of left pelvic ureteric junction stones was confirmed by gross pathology after the operation. The pathological findings concerning the papillary mass around the stone indicated renal pelvic transitional cell carcinoma (Fig. 5). Therefore, for the diagnosis of GH, considering the long-term stone obstruction, the local complications of malignant tumors should be taken into consideration. Similar studies have noted these types of pathological results [14]. Contrast-enhanced abdominal and pelvic CT can show the structure of the tumors around CY-19-9 pyeloplasty, account.

In conclusion, GH is a rare disease, and its symptoms are diverse. The rarer symptoms of cystic hypertonic compression of surrounding organs, such as intractable hiccup, should be taken into account. Treatment of GH includes percutaneous nephrostomy, pyeloplasty, renal cortical fold repair, and occasionally nephrectomy [17]. Patients with advanced age and anemia may not be good candidates for surgery. Because the patient’s intractable hiccups gradually increased, the patient and his family eventually selected right nephrectomy, and the symptoms had not recurred as of half a year of follow-up.

The treatment of GH involves nephrectomy if the kidneys are nonfunctional; for functional organs, the treatment includes percutaneous nephrostomy, pyeloplasty, renal cortical fold repair, and occasionally nephrectomy [17]. Patients with advanced age and anemia may not be good candidates for surgery. Because the patient’s intractable hiccups gradually increased, the patient and his family eventually selected right nephrectomy, and the symptoms had not recurred as of half a year of follow-up.

In conclusion, GH is a rare disease, and its symptoms are diverse. The rarer symptoms of cystic hypertonic compression of surrounding organs, such as intractable hiccup, should be taken into account. Treatment of GH includes percutaneous nephrostomy, pyeloplasty, renal cortical fold repair for functional kidneys, and nephrotoxicity for non-functional kidneys. This case and literature review showed that GH combined with malignant tumors of the renal pelvis is common, so unless it can be diagnosed as benign GH, malignant lesions should be taken into account when planning treatment, so as to prevent postoperative tumor recurrence and metastasis [18].

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Ethical approval
This study is exempt form ethical approval.

Consent
Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contribution
Xiaoxing Liao performed the surgical procedure, proposed the study and wrote the paper.
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