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Case report

Quain hernia - A rare cause of acute small bowel obstruction. A case report and an updated literature review

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ABSTRACT

Introduction and importance: Internal herniae are a rare cause of acute small bowel obstruction (SBO), accounting for <1% of all causes of SBO. Given their low incidence and often vague presenting symptoms there can be a delay in their diagnosis - which can lead to unnecessary morbidity for patients.

Case presentation: We present a case of a 34 year-old nulliparous female who presented with acute abdominal pain and transpired to have a closed loop obstruction of her ileum through a congenital defect in her broad ligament, or a Quain hernia.

Discussion: This paper will describe this case and provide an updated literature review of Quain herniae from recent research. With regards to surgical management of these hernia, both laparoscopic and open approaches are appropriate as long as appropriately trained surgical staff are involved. If a contraltateral defect in the Broad ligament is identified, this should be repaired prophylactically at the time in order to prevent future instances of internal herniation.

Conclusion: Increased awareness of the potential presenting symptoms and radiological features of Quain hernia, as outlined in this paper, is vital in order to reduce patient morbidity and mortality.

1. Case report

We present a case of a 34-year-old nulliparous female who presented to the acute surgical team at a district general hospital with acute abdominal pain, and was subsequently found to have a Quain hernia intra-operatively. We will discuss the patient's presentation and investigations prior to this point, before presenting an updated literature review of this rare type of internal hernia. There is some debate as to the gold-standard form of management of these herniae and we will discuss the evidence behind this, including whether open or laparoscopic repair should be considered and prophylactic management.

This patient presented to the acute surgical department with a 24 h history of right-sided, colicky upper abdominal pain. She had numerous episodes of non-bilious vomiting that morning and had last had a normal bowel movement 12 h previously. She had no dysuria, no per-vaginal bleeding and her last menstrual period was 3 weeks prior. She had a familial history of paternal inflammatory bowel disease (IBD) and was awaiting a colonoscopy for a six-month history of intermittent abdominal pain and fresh PR bleeding. She had no previous surgical history and no other relevant past medical history; was not taking any regular medications; was a non-smoker, a social drinker, and had no history of drug abuse.

On examination she was tender in the right upper quadrant, with mild tenderness over the right iliac fossa, but she was not peritonitic. She had no abdominal distention, her bowel sounds were initially normal and she had no guarding. Her hernial orifices were normal. On admission her bloods showed a mild inflammatory response with a CRP of 8 mg/L and WCC of 12.6×10^9/L. Liver function tests, amylase and renal function were normal, lactate was 0.2 and serum beta-HCG was negative. Chest x-ray on admission was normal with no free air under the diaphragm and urine dip was clear. She was admitted for observation with a working diagnosis of biliary colic or possibly a first presentation of IBD.

An abdominal and pelvic USS on day 2 demonstrated a trace of free fluid posterior to the uterus and into the right adenexa which may have been physiological. There was no evidence of biliary disease and no other identified pathology. On day 3 her bowels remained inactive, and occasional non-bilious vomiting continued. However, the pain had now moved to the epigastrium. CRP rose to 50 mg/L and remained static thereafter. WBC trended down to 10.0×10^9/L then to 7.6×10^9/L over

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these two days. The working diagnosis became gastritis and a plan to discharge the patient with proton pump inhibitor therapy and an outpatient OGD was made. However, the pain remained severe despite PPI, and she remained in hospital for analgesia and plan for an in-patient OGD the following day.

On day five of admission, the patient reported a single episode of bilious vomiting. She remained systemically well apart from a borderline tachycardia of 98. CRP remained static at 51 mg/L and WCC was only mildly elevated at 11.1 $\times$ 10^9/L. The patient was made nil by mouth, and a CT of her abdomen and pelvis with contrast was organised that morning (Figs. 1 and 2). This revealed what was likely to be a closed-loop SBO with small bowel matted down in the pelvis, and a small to moderate volume of free fluid within the pelvis, tracking down the right para colic gutter. There was postulated to be a transition point in the pelvis and given the failure to improve and possibility of a closed loop obstruction, an emergency laparotomy was recommended. The possibility of Crohn's disease and potential need for an ileostomy was discussed with the patient pre-operatively.

After the above discussion, she was taken to theatre by an experienced consultant and registrar in General Surgery. Intra-operatively, small bowel obstruction was confirmed, with some free fluid within the pelvis but no overt signs of ischaemia. There were no macroscopic features of IBD. A transition point and closed loop were identified, involving the small bowel 20 cm proximal to the ileo-caecal junction. The transition point was identified as an internal herniation of the small bowel through a right sided complete peritoneal defect between the round ligament, fallopian tube and lateral pelvic wall. The obstruction point was tight, with mild mesenteric oedema and a shrunken bowel wall. The involved bowel was rested in hot saline-soaked gauze for 10 min and a small serosal defect was over-sewn at the constriction site, but no formal bowel resection was performed. The right sided peritoneal defect was primarily repaired, as was a smaller, but similarly located defect, on the left-hand side.

This patient made a good recovery and was discharged home on day four post-procedure. On further discussion it transpired that she had suffered from 7 years of intermittent right sided abdominal pain. She had numerous normal scans and investigations during this seven-year period, perhaps indicating a chronic component preceding this episode of acute SBO secondary to an internal hernia.

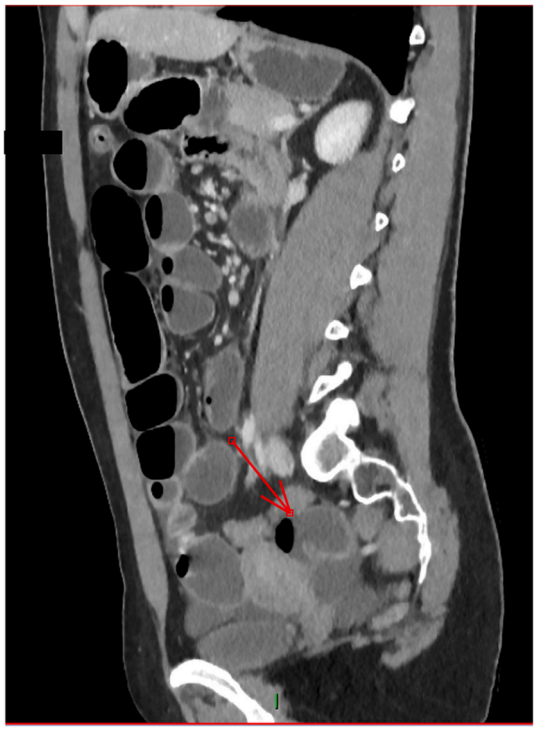

Fig. 1. Saggital plane CT image demonstrating dilated small bowel loops and pelvic matting of bowel as well as the potential transition point of small bowel within the pelvis.

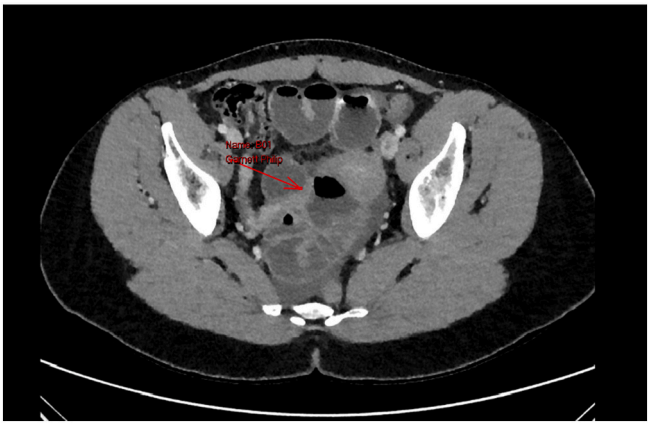

Fig. 2. Axial plane CT demonstrating dilated and fluid filled small bowel with a transition point in pelvis. Free fluid within the pelvis is visible also.

At 6 months' follow up this patient had the occasional pain at the scar site only but had no other issues with her bowel habit. She was considering starting a family and was advised there should be no contraindication to this in regard to her previous surgery or pathology, but the obstetric team will be informed of this in order to aid their decision making.

2. Discussion and literature review

Acute SBO accounts for between 10 and 20% of acute surgical admissions in the UK [2,3] and in the years 2018–2019 was the cause of 47.4% of Emergency Laparotomies performed in the UK [4]. Small bowel obstruction is most commonly caused by adhesions [2], followed by herniae, inflammatory bowel disease and malignancy amongst other aetiologies [3].

Internal herniation causing SBO is rare, with an reported incidence of between 0.2 and 0.9% [5] but can carry a reported average mortality rate of over 50% [6]. Internal herniae are defined as a viscus protruding through either an anatomically abnormal or normal peritoneal or mesenteric opening within the peritoneal cavity [7]. Para-duodenal herniae are the most common subtype of these, accounting for just over half of all internal herniae [5], whereas broad ligament defects, cause an estimated 4–7% of all internal herniae [8]. This type of hernia, often eponymously termed a 'Quain hernia' after his discovery of SBO secondary to a broad ligament defect during an autopsy in 1861 [1], have been described infrequently in the literature. Langan's review suggested that since the first documented report in 1861 up until 2010 there had been 75 published cases describing a Quain hernia [9].

A previous literature review published by Lim et al. [10] discussed the findings of 28 published case reports in the English literature up until 2012. Their findings were: the increasing use of laparoscopic techniques; the high prevalence of unilateral and secondary type of broad ligament herniae; and the generally poor detection rate of CT scanning in diagnosing Quain herniae.

This paper offers an updated and comprehensive literature review of the additional 17 cases of Quain herniae published in English literature since the previous review in 2012 as well as comparison with our own case outlined above, describing all significant aspects of investigation and management of these rare herniae [5,11,12–23] (Table 1).

The pathophysiology of Quain herniae have been previously discussed in the literature. As many as 85% of the cases described occur in parous women, it is therefore thought that the most common cause is

Table 1
Summary of findings of 17 case reports published since 2012.

		Subtotal of cases	Total cases
Modality of treatment	Laparotomy	9	17
	Laparoscopy	8	
Diagnosis	Pre- op CT	10	
	Intra-operatively	7	
Parity	Nulliparous	7	
	Parous	8	
	Unknown	2	
Bilateral	Unilateral	2	
	Bilateral	3	
	Unchecked	12	

some form of trauma occurring during pregnancy or delivery [24]. However, as shown by the above case, almost 20% of cases occur in nulliparous female patients [25]. Suspicion should remain high especially in patients with no history of abdominal surgery and there is a theory that congenital defects may account for a proportion of Quain herniae. This has been postulated to occur when para-mesonephric ducts fuse incompletely or when cystic remnants of the Mullerian ducts rupture [26]. As many as 90% of Quain herniae contain small bowel, particularly the ileum [7].

Clinical presentation has been described as a spectrum from mild, colicky, intermittent pain to complete intestinal obstruction [6]. CT diagnosis is advised frequently in the literature, with a review of CT diagnoses advocating the following diagnostic criteria; 1 - a pelvic transition point; 2 - small bowel loops dilated and herniated lateral to the uterus within the pelvic cavity; and 3 - displacement of uterus forwards from the opposite side of the hernia site [27]. In our case CT scanning successfully diagnosed a transition zone within the pelvis with associated dilated small bowel loops. However, the position of the uterus was not commented on and on retrospective review there was no obvious uterine displacement. In the recent published cases, 10 of the 17 cases correctly diagnosed a Quain hernia on the CT scan.

Two methods of classification of broad ligament herniae are used in current clinical practice. The first was proposed by Hunt in 1934 [28] and describes 3 types based on the degree of the defect, where-as the second classification was offered by Cilley et al. [29] in 1986 and is instead based on the anatomical location of the broad ligament defect. These classifications are summarised below in Table 2. In our case, this patient had a Type 2 (Ciley), Fenestral (Hunt) hernia.

Surgical management of these herniae has been a developing area in recent years. Whilst a midline laparotomy was previously the default surgery, laparoscopic repair has been increasingly utilised in recent years since the first published successful case in 2003 [10]. More recently, the apparent first single incision laparoscopic repair of a broad ligament hernia causing SBO was published by Takeyama et al. in 2017 [16]. As the clear diagnosis of an internal hernia was not identified on CT scanning and due to the high risk of ischaemia, our case underwent a laparotomy rather than a minimally invasive intervention, but this is something that should be considered and discussed with patients as part of the consent process. In the recent literature, this review found a 50/50 divide between laparoscopic and open management of Quain herniae, with one case converted to open due to intra-operative difficulties [19]. Of those successfully diagnosed on CT scan, only 60% were managed laparoscopically.

There has been a recorded case of recurrent internal Quain herniae [12] on the contralateral side to the original defect, demonstrating the importance of checking for and repairing any defects on the contralateral broad ligament as was done in our case. This was only recorded as having been done in 2 out of the 17 recent cases.

3. Conclusion

Quain herniae are a rare subtype of internal herniae and can cause acute small bowel obstruction resulting in a potentially high mortality rate. Non-specific presenting symptoms, along with lack of awareness of the potential diagnosis of internal herniae may lead to sub-optimal or delayed treatment. This case study and literature review demonstrates that early CT scanning should be considered in any patient with non-resolving acute abdominal pain, and suspicion of internal hernia, such as a Quain hernia in female patients, should be considered, although CT findings are not always diagnostic. Laparoscopic management of this pathology is appropriate as long as appropriately skilled surgeons are available in appropriately selected patients.

Written informed consent was obtained from the patient for publication of this case report and accompanying images. This work has been reported in keeping with SCARE 2020 criteria [30]. Provenance and peer review. Not commissioned, externally peer-reviewed.

Table 2
Summary of classification of board ligament herniae.

Classification	Type/ denomination	Definition
Hunt	Fenestra	Presence of defect in two peritoneal layers
	Pouch	Defect in only one layer of peritoneum
	Hernia sac	Attenuated peritoneum lines the herniated bowel in a double layer
Cilley et al.	Type 1	Mesovarium defect
	Type 2	Defect in mesovarium and mesosalpinx above round ligament
	Type 3	Mesoligamentum teres defect

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Ethical approval was not required for this article.

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

Each author contributed equally to the study concept, design, paper writing and final review prior to submission.

Registration of research studies

Not applicable.

Guarantor

The Guarantor for this work is the corresponding author, Mr. Scott MacDonald, who had access to the data and accepts full responsibility for the conduct of the study.

Declaration of competing interest

There are no formal conflicts of interest to declare.

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