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Intestinal malrotation in an adult patient with other congenital malformations: A case report

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ABSTRACT

INTRODUCTION: Intestinal malrotation refers to a variety of abnormalities which occur between weeks 5–12 of embryological development. Most presentations occur before the first year of life. However, patients persisting beyond this period report chronic abdominal symptoms making it difficult to diagnose. Although uncommon, it is important that emerging surgeons and radiologists are made aware of the diagnosis and management of adult intestinal malrotation cases.

PRESENTATION OF CASE: We present the case of a 40 year old patient admitted with subacute abdominal pain on a background of chronic abdominal pain, alternating constipation and diarrhoea requiring several previous hospitalisations and other congenital malformations. Outpatient computed tomography (CT) of her abdomen demonstrated intestinal malrotation and emergency laparotomy revealed Ladd's bands compressing the duodenum. Ladd's procedure was performed and she had an uncomplicated recovery in hospital.

DISCUSSION: Intestinal malrotation can present acutely as volvulus mimicking an obstruction or more commonly, as chronic symptoms such as intermittent cramping, alternating constipation and malnourishment. Gold standard diagnosis in adults is by computed tomography imaging with oral contrast demonstrating inappropriate bowel position and/or inversion of superior mesenteric vessels. It is accepted that the definitive management is via the Ladd's procedure although there is controversy regarding when laparoscopy or laparotomy should be considered.

CONCLUSION: Intestinal malrotation is uncommon amongst adults but its complications can be devastating if not recognised early. Ladd's procedure either laparoscopically or via laparotomy can provide good resolution of symptoms if performed astutely.

1. Introduction

Cases of adult intestinal malrotation are rare but can have devastating consequences if not recognised early. Usually, adults with intestinal malrotation report chronic abdominal symptoms such as intermittent abdominal pain and vomiting, bloating, malabsorption and alternating constipation and diarrhoea [1–3]. Gold standard diagnosis in adults is usually with cross-sectional imaging with oral and intravenous contrast (computed tomography (CT)), although it is often encountered intra-operatively as well [4]. There is still no means of predicting which patients will proceed to midgut volvulus or bowel ischaemia [5]. Due to the low incidence and nonspecific presenting symptoms, it is important that emerging general surgeons and radiologists are made aware of this diagnosis and subsequent management.

The standard for definitive management is with Ladd's procedure which can be performed via laparotomy or laparoscopically. Adult patients undergoing either surgical approach have good resolution of both acute and chronic symptoms.

We present a unique case of a 40-year old lady with a background of congenital abnormalities including Goldenhar and Klippel-Feil syndrome and contrast-media allergy who presented with acute on chronic abdominal pain. An approach for diagnosis, surgical management and when to consider laparotomy or laparoscopic Ladd's procedure for the modern-era general surgeon is elucidated. This case has been reported in line with the SCARE criteria [6].

2. Presentation of case

A 40-year old lady presented to our emergency department with generalised abdominal pain. She was noted to be opening her bow-

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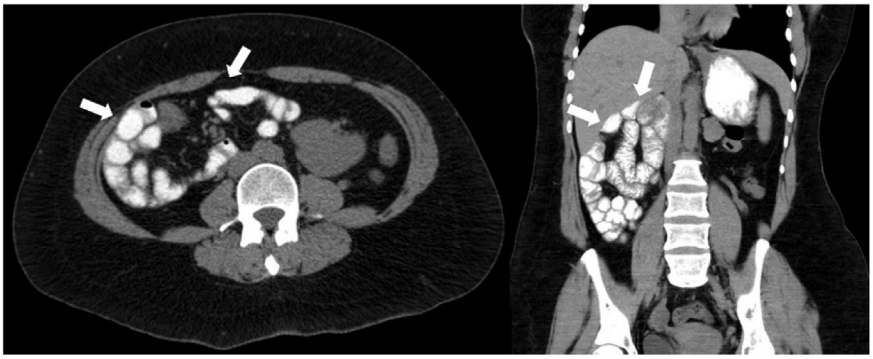

Fig. 1. Axial and coronal CT images with barium oral contrast, of our patient with arrows demonstrating all small bowel positioned on the right side; this is diagnostic of malrotation.

els and was not vomiting on initial presentation. Five days previous to this, she had presented and was admitted to another peripheral hospital with similar symptoms. During the admission, she was noted to have a normal abdominal ultrasound, her pain improved and she was discharged home.

On presentation to our hospital, her respiratory rate was 18 breaths per minute, oxygen saturation was 98% on room air, blood pressure was 121/75 mmHg, heart rate was 55 beats per minute and she was afebrile at 36.5 °C. Physical examination revealed a tender abdomen especially in the epigastric region with voluntary guarding.

She was known to have other congenital abnormalities including Goldenhar Syndrome and Klippel-Feil Syndrome. She described a background of chronic abdominal pain with alternating diarrhoea and constipation requiring many previous hospital admissions. However, she explained that her abdominal pain on this presentation was different (sharper and cramping). She had multiple previous gastroscopies and colonoscopies which had revealed no obvious abnormalities, a cholecystectomy and an open duplex appendicectomy over 25 years ago in a peripheral hospital. The patient also described that she had diagnostic laparoscopies performed by a gynaecologist for her chronic 'abdomino-pelvic pain' for possible endometriosis; she was never formally diagnosed with any gynaecological abnormality.

Despite several years of chronic abdominal pain, the patient did not have any contrast studies or CT of her abdomen because she had a severe allergy to iodine contrast media. However, she presented on this occasion with an outpatient CT (with oral barium contrast, no intravenous contrast) which demonstrated all of the small bowel on the right side of her abdomen (Fig. 1).

The general surgeon to whom she was known to was on call for acute surgery the day she had presented and immediately reviewed the patient. She was kept nil by mouth, commenced on IV antibiotics and booked for emergency surgery.

A midline laparotomy was performed under general anaesthesia by the consultant general surgeon. Intra-operatively, the patient was noted to have a mobile caecum and the small bowel was noted to all be on the right side of the abdomen (Fig. 2). Ladd's bands from the caecum to the right abdominal wall compressing the duodenum were divided and the Ladd's procedure was completed by fixing the caecum to the left upper quadrant (Figs. 3 & 4). None of the small bowel was noted to be ischaemic or necrotic and bowel resection was not necessary. Due to previous appendicectomy, this part of the Ladd's procedure was not necessary.

She had an uneventful post-operative recovery and was discharged eight days after laparotomy. She was followed up by the surgeon at three and seven weeks post-operatively and there were no major post-operative complications. The chronic abdominal pain she had pre-operatively had completely resolved and the patient was content with the outcome.

3. Discussion

Midgut malrotation is a congenital anomaly of intestinal rotation. It is estimated that more than 90% of patients with intestinal malrotation will present in the first 12 months of life [7,8]. However, some children may escape this period if they were asymptomatic or only had vague abdominal symptoms which were misinterpreted for another cause [5]. Diagnosis is rare and unexpected in adults due to presentation with non-specific symptoms such as colicky abdominal pain, bloating, chronic vomiting, malabsorption/inability to gain weight and alternating diarrhoea and constipation [1–3].

Midgut malrotation occurs due to the failure of the normal 270° anti-clockwise rotation of the midgut along its vascular pedicle as

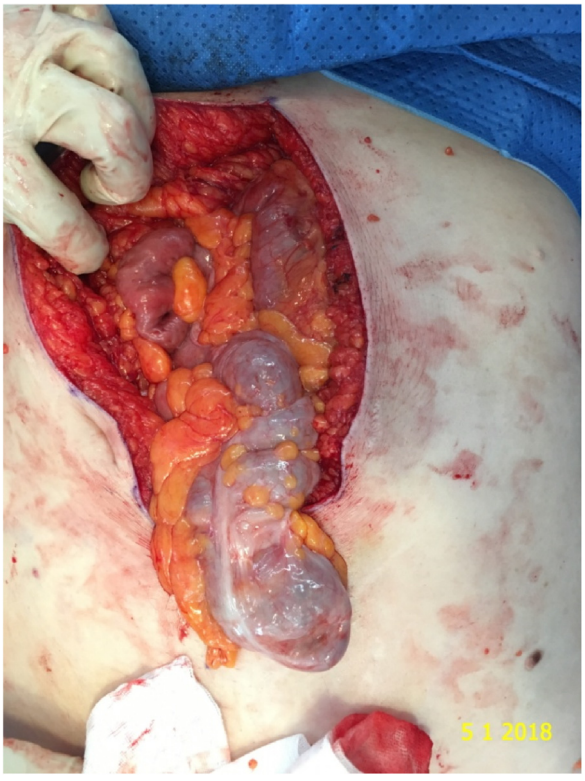

Fig. 2. Small bowel noted completely on right side of abdomen.