A case report of management of gastric perforation in situs inversus totalis in a 45-year-old adult. A case report

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ABSTRACT

INTRODUCTION: Situs inversus is a rare congenital malformation often discovered during childhood. It can cause diagnosis errors in adulthood. Its association with gastric perforation is an extremely rare event in the literature. Its diagnosis is made by an adequate morphological assessment.

PRESENTATION OF CASE: A 45-years-old man, was admitted to the surgical emergency department for generalized acute abdominal pain initially sitting in the right hypochondrium, accompanied by bilious vomiting and a stop in intestinal transit, progressing for 48 h. He had no known surgical history. The clinical examination noted an altered general state (WHO III) and a peritoneal syndrome. A diagnosis of generalized acute peritonitis has been made. An x-ray of the abdomen without preparation revealed a bilateral pneumoperitoneum with a cardiac point on the right. The thoraco-abdomino-pelvic CT scan confirmed the diagnosis. After resuscitation, the patient underwent a laparotomy with gastrostomy and appendectomy. The postoperative follow-ups were without an uneventful, over a 15-month follow-up.

DISCUSSION: Situs inversus totalis is an uncommon event. Its exact etiology is still unknown. Some authors incriminate an autosomal recessive gene. In our context, the diagnosis is a surprise. Its revelation by gastric perforation is an extremely rare event. Surgical treatment must be performed early. Prognosis is generally better.

CONCLUSION: In developing countries the diagnosis of situs inversus is a surprise during a pathology which led the patient to a medical consultation. CT-scan is one of the key paraclinical exams for its diagnosis.

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1. Introduction

Described for the first time in human beings by Fabricius in 1600, situs inversus is defined as a congenital malformation characterized by a reversal of the position of the intra-thoracic-abdominal organs. It can be totals or incomplete. It is most often revealed during childhood [1]. When situs inversus is totalis, it is defined as a mirror position inversion of the intra-thoracic-abdominal organs from their normal position. In situs inversus totalis (SIT), there are generally no major health problems [2]. Its incidence is around 1 in 10,000 to 1 in 50,000 births per year. Until today, the etiology of situs inversus remains unclear. However, some authors incriminate an autosomal recessive gene [2, 3]. Although some cases of association of situs inversus with other pathologies have been described in the literature, the revelation of SIT by gastric perforation remains an extremely rare event which can be a source of diagnostic and therapeutic errors in adulthood [4]. Therefore, making a diagnosis before the onset of a surgical pathology would be of great help to avoid these errors. The morphological assessment is therefore of paramount importance for diagnosing situs inversus. Acute peritonitis generalized by gastric perforation is certainly frequent, but to our knowledge, no case on SIT has yet been described in our context. We, therefore, report the very first case of situs inversus totalis revealed by gastric perforation treated in our context. Authors declare that this work has been reported in line with the SCARE [5].

2. Case presentation

A 45-year-old, a civil engineer, was admitted to the surgical emergency for generalized acute abdominal pain but initially sitting in the right hypochondrium. The pain was accompanied by
biliary vomiting and cessation of intestinal transit. This symptomatology had evolved for 48 h. The patient had no known surgical history but sometimes complained of burns in the right hypochondrium, radiating to the epigastrium and the back. These burns were treated by self-medication with an intermittent amendment. He was also not known to be a carrier of a situs inversus. On examination, he was rated a WHO III stage with a good conscience. The body mass index was 25.5 kg/m² and the temperature at 38.6 °C. Moderate anemia, polypnea (26 cycles/min), a pinched blood pressure (100/70 mmHg), a pulse rate of 110/min were observed. The physical examination noted a peritoneal syndrome. The diagnosis of generalized acute peritonitis was made and after conditioning, an x-ray of the abdomen without preparation was performed. It revealed a bilateral pneumoperitoneum with a cardiac point on the right (Fig. 1). The thoraco-abdomino-pelvic scanner was performed and confirmed the diagnosis of gastric perforation on situs inversus totalis: the heart and the arch of the aorta are located on the right, (Fig. 2b), the liver, the gallbladder, and the caeco-appendix were located on the left; the stomach and spleen were on the right (Fig. 2a). The position of the kidneys was also reversed (Fig. 2b). The biological assessment noted a hemoglobin level of 10.2 g/dl, the white blood cells at 12,500/mm³, the CRP at 50 mg/L. Also noted were electrolyte disturbances in the blood ionogram. After resuscitation, the indication for an emergency laparotomy was performed. In the operating room, one liter of biliary peritoneal fluid was aspirated and exploration revealed a perforation of about 0.5 cm in diameter, located on the anterior side of the gastric antrum, (which was in the right) with regular and flexible banks (Fig. 3a). The complete exploration of the abdominal cavity confirmed the reversal of the position of the viscera (The complete exploration of the abdominal cavity found the reversal of the position of the viscera: the stomach (the great curvature), the spleen and the sigmoid were on the right; the duodenum, the right lob of liver, the gallbladder and the caeco-appendix were located on the left relative to the midline (the caeco-appendix being in the left iliac fossa). Their appearance was normal. The procedures carried out consisted of a biopsy of the banks of the perforation for histology examination, gastrorraphy, appendectomy, peritoneal toilet and the parietal closure. In postoperative period, resuscitation was continued, associate with...
omeprazole 40 mg twice-a-day. The resumption of intestinal transit was effective on the third day. The patient was released on the fifth day postoperatively. The conclusion of histology results was: chronic inflammatory cells, no signs of metaplasia and no identified Helicobacter Pylori. The postoperative follow-up was simple after 15 months of follow-up.

3. Discussion

The exact cause of situs inversus totalis, has still not been identified by scientific research today, although an autosomal recessive gene has been implicated [6]. Some authors have mentioned the immobility of the nodal cilia during embryogenesis, leading to an abnormality in the rotation of the intra thoraco-abdominal organs, which would be responsible for the occurrence of this anomaly [7]. It is recognized in the literature that SIT would generally not affect the health of the patient [2,3,6]. But some authors have reported that around 60% of patients with SIT may have other birth anomalies of the digestive tract such as atresia of gallbladder or bowel, splenic agenesis or colonic duplication [7]. These anomalies are manifested during childhood by digestive signs which lead to early diagnosis, failing prenatal diagnosis [8]. However, no associated congenital anomaly was identified in our patient. Our patient was asymptomatic apart from his history of abdominal pain treated with self-medication. It should be noted that until the onset of peritonitis by gastric perforation, he did not know he was a carrier of situs inversus and, no history of dextrocardia in his family had been mentioned. The absence of a history of serious pathologies in our patient would partly justify the delayed diagnosis. In addition, in our context, the population rarely consults a physician at the first sign of a pathology. Generally, a physician is consulted after failure of traditional treatment [8]. The discovery of situs inversus was a surprise for our patient and his family. A case similar to ours was reported by Biswajit et al. [9]. It was a 62-year-old person who had no family history of dextrocardia whose diagnosis of visceral position abnormality was made during the investigation of acute abdominal pain syndrome. The revelation of the situs inversus by a peritoneal syndrome would be an extremely rare event. Thus, we have identified three cases of duodenal perforation [10–12] and two cases of perforation of the anterior aspect of the gastric antrum [13,14] on SIT in the literature. In the majority of these cases, the diagnosis was made before the onset of abdominal pain syndrome. In our patient, it is the morphological assessment requested to support the diagnosis of perforation of a hollow organ, which has made it possible to discover the dextrocardia hitherto ignored. Indeed, the emergency x-ray of the abdomen without preparation revealed a cardiac point on the right as well as a bilateral pneumoperitoneum (Fig. 1). The thoraco-abdominal CT scan had confirmed the diagnosis of SIT by showing a reversal of the position of the viscera (Fig. 2): the point of the heart was on the right, the liver on the left, stomach and spleen on the right. These two paraclinical examinations are part of the key imaging allowing the diagnosis of this anomaly to be made [7,8,10,13]. The classic treatment for acute sthenic peritonitis is a laparotomy. This attitude was adopted for the management of our patient. Exploration in the operating room confirmed the diagnosis of reversal of viscera position (Fig. 3). The treatment consisted of gastrotomy, appendectomy in principle, abdominal toilet and the classic closure of the wall. Several authors have adopted the same attitude [12,13,15]. The postoperative follow-up was simple after 15 months of follow-up.

4. Conclusion

In developed countries, the diagnosis of SIT is often made in antenatal or childhood. However, in developing countries its diagnosis is often a surprise; so, a systematic examination of any patient, would make it possible to suspect and confirm it by medical imaging.

Declaration of Competing Interest

The authors declare that they have no competing interests regarding the publication of this manuscript.

Funding

Authors declare that there is no sponsors for the publication of this case report.

Ethical approval

This study of clinical case report is exempt from ethical approval in our Institution.

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

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Data collection: OUEDRAOGO NLM.
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Registration of research studies

In the Research Registry GuideBook, it is mentioned in page 4 «The only study types not included in the Research Registry are case reports that are not first-in-man and animal studies». So, this work doesn’t need to be registered.

Guarantor

Guarantor is Prof. Si Simon TRAORE.

Provenance and peer review

Not commissioned, externally peer-reviewed.

Acknowledgements

We thank the whole team of Saint Camille Hospital and of General and Digestive Surgery Department of Yalgado Ouedraogo University Hospital Center, for administrative and technical support. We would also like to thank the patient who gave us his permission for the publication.

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