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CLINICAL CASE

Wilkie syndrome. Case report*

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KEYWORDS

Wilkie syndrome; Superior mesenteric artery; Cast syndrome

Abstract

Background: Wilkie syndrome, also referred as superior mesenteric artery syndrome, is an unusual cause of a proximal small bowel obstruction. It is characterised by the compression of the duodenum in its third portion due to a narrowing of the space between the superior mesenteric artery and the aorta. Its presentation symptoms are consistent and include the obstruction of the proximal small bowel. However, the physical and laboratory findings are non-specific. Nevertheless, many imaging methods are useful for its diagnosis. The management of this condition varies between observation and surgery, depending on each particular case.

Clinical case: The case is presented of a 19-year-old male who began with acute, intense abdominal pain, nausea, vomiting, and diarrhoea. On examination, he had abdominal wall rigidity and hyperesthesia. Imaging studies were requested, revealing a decreased superior mesenteric artery angle, a shortening of the aortic mesenteric distance, and a decrease in the calibre of the third duodenal portion, all findings concomitant with Wilkie syndrome. Conservative treatment was applied and the patient was discharged without complications. Conclusions: Wilkie syndrome continues to be an unknown condition to the general practitioner, and the underdiagnosis of this condition may put a patient at risk of serious complications. A high index of suspicion is required to reach a diagnosis. Early treatment should give a good outcome most of the time.

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PALABRAS CLAVE

Síndrome de Wilkie; Arteria mesentérica superior; Síndrome de Cast

Síndrome de Wilkie. Reporte de un caso

Resumen

Antecedentes: El síndrome de Wilkie o síndrome de la arteria mesentérica superior es una causa inusual de oclusión intestinal alta, se caracteriza por la compresión del duodeno en su tercera porción, es secundaria a un estrechamiento del espacio entre la arteria mesentérica superior y la aorta. El cuadro clínico suele estar caracterizado por síntomas de oclusión intestinal alta, la exploración física y los hallazgos de laboratorio suelen ser poco específicos. Diversos estudios de imagen son útiles para el diagnóstico. El tratamiento puede ir desde conservador, hasta la cirugía, que depende de cada caso.

Caso clínico: Varón de 19 años, que cursó con dolor abdominal súbito, intenso, náuseas, vómitos y diarrea; motivo por el cual acudió al servicio de urgencias, donde a la exploración destacaron: rigidez abdominal con hiperestesia e hiperbaralgesia, y los estudios de imagen evidenciaron una disminución del ángulo de la arteria mesentérica superior y de la distancia aortomesentérica; además de disminución del calibre de la tercera porción del duodeno, hallazgos característicos del síndrome de Wilkie. Se dio tratamiento conservador, siendo dado de alta hospitalaria sin complicaciones.

Conclusiones: El síndrome de Wilkie continúa siendo una entidad poco conocida por el médico general, el que poco se diagnostique o se piense en este síndrome, puede ocasionar complicaciones serias por lo que se requiere que el médico guarde un alto índice de sospecha diagnóstica ante esta patología. El tratamiento oportuno confiere un buen pronóstico la mayoría de las veces.

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Background

Wilkie syndrome, which is also known as upper mesenteric artery syndrome, plaster corset syndrome or aortomesenteric compass syndrome, is characterised by compression of the third portion of the duodenum. It is secondary to narrowing of the space between the upper mesenteric artery and the aorta. Although this entity was first described by Rokitansky in 1861,¹ it was in 1921 when Wilkie² published a series of 75 cases resolved by surgery that it became known eponymously.

Several risk factors have been found in association with this pathology. One of the most widely accepted causal theories is the loss or reduction in the mesenteric fat which functions as a cushion between the aorta and upper mesenteric artery.³ However, other case reports associate Wilkie syndrome with: HIV, neoplasias, diabetes mellitus,⁴ orthopaedic surgery for scoliosis,⁵ bariatric surgery,^{6,7} vascular surgery, major catabolic states, sudden weight loss and eating disorders such as anorexia nerviosa.⁸

Patients may present with symptoms that commenced suddenly or ones with a tendency to become chronic. The most common symptoms are those of upper intestinal blockage. The reasons why these patients visit include: postprandial epigastric pain, early satiety, nauseas, vomiting bile, pyrosis, ingurgitation and weight loss peso; some postures may relieve the symptoms, as they free the duodenum from compression. Although findings in physical examination are not characteristic, some conditions which are usually found are: abdominal distension, poorly localised pain and alterations in peristaltic noises. Laboratory findings

may show hydroelectrolytic alterations after major vomiting or diarrhoea, given that late diagnosis may lead to fatal complications such as perforation and pylephlebitis.¹⁰

Several imaging studies are useful in the diagnostic approach when there is a suspicion that a patient has Wilkie syndrome, which in all cases requires that the doctor retain a high degree of suspicion to achieve a true and accurate diagnosis. At the present time computerised axial tomography is considered to be the gold standard diagnostic method. Nevertheless, findings using other imaging techniques may lead to the suspicion of a diagnosis when the clinical symptoms and risk factors are present. It is due to this that the approach usually commences with simpler and more economical methods. Simple abdominal X-ray may show major gastric dilation; a gastroduodenal series with oral contrast usually shows duodenal compression in the third part of the duodenum with active peristalsis, slow contrast transit and duodenal dilation close to the obstruction. 9 Abdominal ultrasound scan by an expert is a good diagnostic alternative, given that it is possible to measure the aortomesenteric angle with the advantages of not exposing the patient to ionising radiation, its accessibility and low. However, this technique has the major disadvantage of being operatordependent. 11 Arteriographs are currently not used for the diagnosis of this pathology, and this technique is only used in cases with a high level of diagnostic suspicion and inconclusive results using other imaging methods.

Computerised tomography has almost 100% specificity and sensitivity in diagnosis when the following criteria are fulfilled: (1) abrupt obstruction in the third portion of the duodenum, with active peristalsis, (2) an aortomesenteric

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