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Fatal outcome of gastric perforation due to infection with Sarcina spp. A case report

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ABSTRACT

Sarcina ventriculi is an extremely rare pathogen. These gram-positive cocci bacteria are rarely identified in gastric biopsies and usually described in the scientific literature as an incidental finding, particularly in patients with delayed gastric emptying, gastroparesis, emphysematous gastritis or gastric perforation. It occurs most commonly in adult women and can be identified easily by its distinctive morphologic features, such as basophilic staining, cuboidal shape, tetrad arrangement, red blood cell-sized packets, flattened cell walls, and refractile nature in bright field microscopy. Although the pathogenesis of the microorganism is highly debated in humans, this bacterium is a well-known pathogen in livestock. Fewer than 30 cases of human infection have been described in the scientific literature so far, but none mentioned this micro-organism as a potential cause of death. We report the case of a 76-year-old patient with gastric perforation due to massive infection with *Sarcina ventriculi*. To date, this is the first report of human infection with *Sarcina ventriculi* in Romania.

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Introduction

In the context of a modern changing world, supporting advances in clinical practice is of tremendous importance. Clinical evaluation among with various means of biomaterials investigation such as failure analysis can make a contribution to the progress of medical science and education [1-6]. Case reports and randomized controlled trials occupy an important place in the improvement of patient outcomes, especially when referring to rare diseases [7-10]. Sarcina is a genus of Gram-positive, nonmotile, anaerobic bacteria with exclusive carbohydrate fermentative metabolism [11], which is able survive in a wide range of environmental conditions, even in the low pH of the stomach [12]. Although similar in appearance to Micrococcus species, specific morphological features such as: larger size, and non-cluster forming pattern, help differentiate Sarcina species from Micrococcus [13]. The type species is Sarcina ventriculi, a variety found on the surface of cereal seeds, in soil, mud, and in the stomach of humans,

* Corresponding author. E-mail address: daniela.mgm8@gmail.com (D. Gheorghiță). rabbits, and guinea pigs [14]. *Sarcina ventriculi* was classified as an anaerobic coccus in 1960s and first described in humans by Goodsir [15] and later by Ferrier [16]. Various reports in veterinary literature have implicated Sarcina in the development of gastric dilatation and death of livestock, cats, goats and horses [17].

Sarcina has also been reported to be found in feces of healthy humans consuming a predominantly vegetarian diet [18]. Even though it has been identified as a potential pathogen for humans since 1842 [19] and isolated in pure culture from gastric fluids since 1911 [20], more recent studies have shown an association between Sarcina in the stomach and chronic nausea, dyspepsia, abdominal pain, gastric ulcers, and rarely, emphysematous gastritis and gastric perforation [21-24]. However, Sarcina ventriculi has also been identified in gastric biopsies from asymptomatic patients and no pathologic changes associated to the gastric mucosa, suggesting that it may be an innocent bystander rather than a harmful bacterium [13]. To date, few cases of Sarcina ventriculi isolated from gastric biopsy specimens have been reported, usually in patients with gastroparesis, iron deficiency anemia or delayed stomach emptying, all of which are excellent environments for bacterial overgrowth [24,25]. Also, there are several case reports which linked Sarcina microorganism

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Case report







Fig. 1. Sarcina organisms arranged in distinctive basophile tetrad packets in the background of cellular debris mucin and inflammatory cells in a patient with gastric perforation. Note that a vast number of colonies are intravascular. Hematoxylin-eosin, original magnification x200 (a) and x400 (b).

coexistence with *Helicobacter pylori* [26] or on a background of malignancy [27], cystic fibrosis [28], coeliac disease [9], or upper gastric surgery [24,29]. We report a rare case of death-related infection with *Sarcina ventriculi* in a 76-year-old male patient with gastric ulcer perforation. To our knowledge, less than a dozen cases of *Sarcina ventriculi* isolated from gastric specimens have been reported, our case being the first one documented in Romania and one of the few reported deaths due to infection with this microorganism.

Case report

A 76-year-old male with severe abdominal pain was admitted to the Emergency University Hospital in Bucharest for evaluation of acute abdominal pain with generalized rebound tenderness and altered mental status. Arterial blood gases confirmed elevated lactate with severe metabolic acidosis. After initial fluid resuscitation a plain film of the abdomen (PFA) showed bilateral pneumoperitoneum and a large hiatus hernia. Although such clinical presentation and PFA findings should undoubtedly prompt emergency surgery the differential diagnosis with adenocarcinoma, lymphoma and perforated ulcer is mandatory in stable patients in order to tailor the surgical intervention appropriately. Hence the PFA was followed by a CT of the abdomen and pelvis for further characterization of the viscera demonstrating: large pneumoperitoneum with parietal tear along the lesser curvature of the stomach with no features suggestive of obstruction, lymphadenopathies or intra-abdominal masses. Intraoperative findings confirmed gastric dilatation and rupture of the gastric wall along the lesser curvature with no gastric outlet obstruction or other macroscopic findings to support a causal relation.

During the surgical procedure in which the perforation of the gastric wall was repaired, tissue samples were subsequently sent for evaluation to the Pathology Department of the same hospital. The specimen samples were fixated with 10% buffered formalin and were processed by conventional histopathological methods using paraffin embedding, 2 microns sectioning and Hematoxylin–Eosin (H.E.) staining. Unfortunately, the patient died shortly after surgery, due to the complications induced by sepsis and multiorgan failure.

Histopathological examination of standard-stained H.E. slides revealed marked acute inflammation, hemorrhagic areas and extensive necrosis of the fatty tissue. No gastric mucosa was identified. However, there was abundant bacterial overgrowth, including the presence of *Sarcina ventriculi* microorganisms with characteristic microscopic features: tetrad or packet-forming arrangement of large cuboidal basophilic bacteria with thin surface walls. Many such bacteria were present in the blood capillaries of the analyzed fragments (Fig. 1a, b), this confirming an active infection and excluding a possible contamination. Ancillary special stains such as Gram, Periodic Acid-Schiff (PAS) and Giemsa have been performed, in order to confirm the diagnosis. As expected, the



Fig. 2. Sarcina ventriculi overgrowth and necrotic debris. Hematoxylin-eosin, original magnification X400.



Fig. 3. Characteristic morphology S. ventriculi. Giemsa stain. original magnification x400.

bacteria were highlighted by strong positivity in Gram (Fig. 2) and Giemsa stains (Fig. 3), but were negative in PAS staining. The final diagnosis of infection with *Sarcina ventriculi* was established based on the characteristic morphology of this microorganism and strong Gram positivity, without the need for additional studies, such as polymerase chain reaction and sequencing of the 16S ribosomal RNA gene and pyruvate decarboxylase gene.

Discussion

Although the pathogenesis of Sarcina spp. is well studied in veterinary literature, its role in human disease is not entirely understood and very few scientific studies identify this microorganism as a potential cause of death. Even if the presence of this bacteria is rarely documented in humans, the infection in animals is well-known to cause abomasal bloating (the abomasum is the fourth component of the stomach in ruminants) in goats [21], gastric bloating in lambs and calves [22] and acute gastric dilatation in dogs and horses [23], all with severe outcome for the host.

In humans, it is unclear if the presence of microorganisms from Sarcina spp. signifies an indicator for another disease state or represents a causative agent itself, which would require medical intervention. Some authors [13] suggest that most cases could represent overgrowth of a commensal organism, and therefore do not require pharmacological intervention. If Sarcina spp. are identified in patients with dysphagia or substernal burning, a combination of proton pump inhibitors and prokinetic therapy is recommended. However, if the microorganism is associated with ulceration of the gastro-esophageal mucosa, eradication with antimicrobials is required, in order to avoid perforation [30]. Confirmation of eradication also warrants follow up endoscopy.

It is presumed that infections in both humans and animals occur due to ingestion of contaminated food [28]. One particular case reports a veterinary doctor who developed infection with Sarcina spp. probably due to close animal contact [24]. Any deferment – delay in gastric output which leads to accumulation of food debris associated with low gastric pH provides an ideal environment for Sarcina spp. overgrowth and can result in a characteristic frothy vomit known as "sarcinous vomit" and/or emphysematous gastritis [25].

Although this microorganism has been known for more than 170 years, less than 30 cases have been reported in the literature, most of them during the past 10 years. In a review published in 2016, Al Rasheed et al. found that 68 % cases of Sarcina spp. were reported in female patients and more than half of the documented cases had a history of gastrointestinal surgery, gastroparesis, or gastric outlet obstruction [25]. Also, there are reports which found Sarcina ventriculi in patients with gastric perforation [23], emphysematous gastritis [20] and in association with malignancies such as gastric and pancreatic cancer [13,27]. There are even cases of Sarcina ventriculi isolated from the blood of the patients with suffering from congenital chloride diarrhea [15] a rare inherited disease that belongs to the Finnish disease heritage caused by mutations in the SLC26A3 gene on the chromosome 7 [25] leading to the function disruption of the major anion exchanger on the surface of intestinal epithelial cells. Our patient did not have a known genetic disease and it is not known whether or not this micro-organism was present in his blood, however multiple intracapillary Sarcina colonies can be observed on the histopathological specimen.

In vitro culture of Sarcina spp. is challenging due to the intricate nutritional necessities of the bacterium, which explains our inability to culture the pathogen. Sarcina maxima, recently reclassified to *Clostridium* maximum may be differentiated from *Sarcina ventriculi* (also phylogenetical placed into Clostridiaceae family, Firmicutes phylum) [15] based on its fermentation characteristics, such as production of butyrate by the former organism and production of ethyl alcohol by the latter. However, morphological differentiation between these two bacteria is not readily achieved since they resemble each other in bright field microscopy.

Scientific literature suggests that favourable outcome may be achieved by early treatment with antimicrobials effective against anaerobes. Current reports indicate successful eradication of *Sarcina ventriculi*, with treatment consisting of metronidazole in combination with another antibacterial, but there is no consensus on the type and duration of treatment [13,20,24]. Since Sarcina involves carbohydrates for fermentation, another important aspect is to maintain nil per os (nothing at all by mouth).

Conclusion

Given its association with life threatening illness, we believe that patients identified with *Sarcina ventriculi* should initiate treatment with antibiotics and anti-ulcer therapy, until further understanding of this microorganism is achieved.

Ethical standards

We undersign, certificate that the procedures and the experiments we have done respect the ethical standards in the Helsinki Declaration of 1975, as revised in 2000 (5), as well as the national law.

CRediT authorship contribution statement

Adrian Dumitru: Conceptualization, Methodology, Validation, Investigation, Writing - original draft, Project administration. Cătălin Aliuș: Methodology, Software, Resources, Writing original draft, Visualization. Adriana Elena Nica: Conceptualization, Formal analysis, Data curation, Writing - original draft. Iulian Antoniac: Validation, Formal analysis, Investigation, Writing - original draft, Writing - review & editing, Visualization, Supervision. Daniela Gheorghiță: Software, Data curation, Writing - original draft, Visualization. Sebastian Grădinaru: Conceptualization, Methodology, Validation, Resources, Data curation, Writing - original draft, Writing - review & editing, Supervision.

Declaration of Competing Interest

The authors declare no conflict of interests.

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