abdominal pain), being an uncommon cause of intestinal obstruction. They are categorized according to their composition, with phytobezoars being the most common, formed from the ingestion of insoluble plant fiber,[2] as in the case described.

The occurrence of intestinal obstruction due to bezoar is rare and poorly reported in AN. Symptom overlap between bezoar and the ED *per se* can obscure and delay diagnosis. Literature describes 4 cases of bezoar in patients with AN (2 cases presented with intestinal obstruction, 1 resolved with emergency surgery and 1 with conservative therapy).[3] Case #3 involved a patient with purging AN who, during hospitalization, experienced vomiting with bezoar-like contents confirmed solved by endoscopy without further treatment.[4] Case #4 involved a patient with trichotillomania admitted for suspected restrictive AN (BMI = 15 kg/m^2) who, during dietary progression, developed intestinal obstruction requiring emergenct surgery, revealing a trichobezoar.[5]

The best imaging modality for initial bezoar diagnosis is CT. Although upper endoscopy can be used for definitive diagnosis, it should not be performed in cases of obstruction.[2] Phytobezoar treatment may be conservative using chemical dissolution or endoscopic/surgical approach based on severity. In the described case, conservative therapy used Coca-Cola® as a solvent with gastric lavage through a nasogastric tube. A systematic review of 24 observational studies with 46 patients showed Coca-Cola® administration resolved phytobezoars in 50% of cases.[6]

In the evaluation of GI symptoms in AN patients, the potential emergence of rare but severe complications requiring specific treatment must be considered.

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References

1. American Psychiatric Association. Diagnostic and statistical manual of mental disorders. Arlington: American Psychiatric Publishing; 2013.
2. Gelrud D, Talley NJ, Robson KM [Accessed 20 April 2024]. Available from: Gastric bezoars. UpTo-Date; 2023 https://ffr.uptodate.com/contents/gastric-bezoars/print
3. Birmingham CL, Cardew S, Gritzner S. Gastric bezoar in anorexia nervosa. Eating Weight Disord. 2007;12:e28–9.
4. Scobie BA. Acute gastric dilatation and duodenal ileus in anorexia nervosa. Med J Aust. 1973;2:932–4.
5. Saldanha NE, Meisel JA, Prince JM. Delayed diagnosis of trichobezoar in a patient with presumed anorexia nervosa. Int J Adolesc Med Health. 2015;27(3):349–52.
6. Ladas SD, Kamberoglou D, Karamanolis G, Vlachogiannakos J, Zouboulis-Vafiadis I. Systematic review: Coca-Cola can effectively dissolve gastric phytobezoars as a first-line treatment. Aliment Pharmacol Ther. 2013;37(2):169–73.

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Rhinocerebral mucormycosis: an uncommon complication in patients with diabetes mellitus

Mucormicosis rinocerebral: una complicación infrecuente en pacientes con diabetes mellitus

Introduction

Mucormycosis is a rare complication in patients with diabetes mellitus. It is an infection caused by saprophytic fungi. The most common clinical form is rhinocerebral. The main risk factor is diabetes mellitus, especially when patients present acidosis, as it alters iron metabolism and favors fungal growth. It is an infection with a high mortality rate, and clinical suspicion is essential for early diagnosis and treatment. We present the case of a mucoral infection in a patient with poorly controlled diabetes mellitus.

Case report

A 59-year-old woman with poorly controlled type 1 diabetes mellitus was admitted to the Intensive Care Unit due to severe mixed hyperglycemic decompensation (severe ketoacidosis and hyperosmolar hyperglycemic state). She had good glycemic control with fluid therapy and IV insulin, requiring high doses. Initially, the possible triggering factor was attributed to pneumonia based on positive antigenuria for *Streptococcus pneumoniae*.

She was examined by Neurology for bradypsychia, suspected to have a metabolic etiology. Chest and abdominal CT scans were performed to rule out triggering factors,

revealing a left renal abscess, which was drained under radiological guidance uneventfully.

After several days of hospitalization, hyperemia, chemosis, and tearing were detected in the left eye. She was evaluated by Ophthalmology, who recommended topical antibiotics. Since bradypsychia remained despite improved glycemic control, an MRI was performed, showing acute sinusitis with postseptal orbital cellulitis. She was subsequently examined by Otolaryngology, endoscopic nasosinusal surgery was performed and antibiotic treatment initiated.

The initial suspicion was mucormycosis; however, since the patient showed clinical improvement with antibiotic therapy, the initiation of antifungal therapy was postponed until 5 days later, when the results for *Rhizopus oryzae* tested positive. Therefore, the diagnosis of rhinocerebral mucormycosis was confirmed, and treatment with liposomal amphotericin B at 10 mg/kg was started.

From a glycemic perspective, there was good initial control, followed by subsequent decompensation and difficult management. A follow-up CT scan showed deterioration. A new surgical procedure was considered, but the patient declined. At this point, a ferric chelator was started. She experienced progressive deterioration of her general condition and ultimately passed away.

Discussion

Mucormycosis (zygomycosis) is a rare complication in patients with diabetes mellitus. It is an infection caused by mucorales and entomophthorales (*Rhizopus*, *Mucor*, and *Rhizomucor*).[1,2] Fungi causing mucormycosis are found in soil, decaying organic matter, or contaminated food.[1,4]

In Spain, the incidence is low, with 0.43 cases per million inhabitants per year being reported.[2]

The infection is favored by certain underlying diseases (i.e., diabetes mellitus, renal failure) or risk factors (neutropenia, immunosuppression, iron overload). The most important risk factor is diabetes mellitus, especially in states of acidosis.[2] However, studies after 2003 have shown a greater association with hematologic diseases (50%), followed by diabetes (23%).[1]

Acidosis alters iron transport, leading to increased free iron, which promotes fungal growth.[5]

There are different clinical forms: orbitocerebral, pulmonary, cutaneous, digestive, or disseminated. The most common form is orbitocerebral, which is also the most common in diabetics.[2] The orbitocerebral form may present with acute sinusitis, fever, headache, nasal congestion, and even invade the nervous system, causing vision loss, cranial nerve involvement, or altered consciousness.[1] Cutaneous involvement may occur in immunocompetent patients with traumatic past medical histories. Other forms include GI and renal, though they are less common.[1]

Diagnosis must be quick to start treatment and improve survival outcomes. Demonstration of the fungus by visualizing hyphae and growth in culture (growth at 30 °C) is required to guide antifungal therapy. The hyphae of mucorales are characteristic: non-septate with right-angle branching. However, even if not visualized, if the clinical picture is highly indicative, diagnosis should not be ruled out.[3,5] Imaging modalities (CT and MRI) are not specific for diagnosis. In CT, it is highly suggestive to observe multiple pulmonary nodules (>10) and the reverse halo sign.[1]

Treatment is based on initiating antifungal therapy, eliminating the predisposing factor, and performing surgical debridement.[1] The antifungal agent of choice is liposomal amphotericin B. Dosage is 5 mg/kg/day, although higher doses of 7—10 mg/kg/day are used in severe cases.[1] Posaconazole and isavuconazole are used as maintenance or alternative therapies in cases of refractoriness.[1] The former can be administered orally or IV. Dose is 300 mg every 12 h, and then every 24 h. It should be used in cases of renal failure. Isavuconazole, however, may be a useful treatment in patients with renal failure and has the advantage of fewer drug interactions vs posaconazole. Dose is 200 mg every 12 h and then every 24 h.[2] Treatment should continue until clinical symptoms and radiological signs resolve, and until the underlying cause of immunosuppression has been addressed.[2] Other options include deferasirox—an iron chelator—or hyperbaric oxygen therapy. However, neither of these 2 therapies can be routinely recommended.[1]

Mortality depends on the clinical presentation, risk factors, and patient condition, and on the timing of surgery. The overall mortality rate is 40% up to 50%.[2,4]

Mucormycosis is a rare and extremely serious complication in patients with diabetes mellitus. Clinical suspicion is vital to initiate antifungal and surgical treatment. In this case, clinical suspicion was high since the patient exhibited both ocular (left eye chemosis) and neurological symptoms (bradypsychia), had poorly controlled diabetes and acidosis, and intraoperative lesions were compatible. A more timely initiation of antifungal therapy, along with obtaining samples for histology might have improved the patient's prognosis.

References

1. Steinbrink MJ, Miceli HM. Clinical review of mucormycosis. Infect Dis Clin North Am. 2021;35(2):435–52.
2. Ruiz-Camps I, Salavert-Lleití M. El tratamiento de la mucormicosis (cigomicosis) en el siglo XXI. Rev Iberoam Micol. 2018;35(4):217–21.
3. Alonso G, Fernández-García D, Reyes-García R, Muñoz-Torres M. Diabetes mellitus y mucormicosis rinocerebral: una asociación de mal pronóstico. Endocrinol Nutr. 2006;53(6):387–9.
4. Caballero-Aldama A, García-Duarte J, Aldama-Negrete F, Insaurralde S, Mendoza de Sánchez G, Celías L. Mucormicosis rinocerebral - reporte de 4 casos. An Fac Cienc Méd (Asunción). 2017;50(3):75–82.
5. Ruiz H, Oddo D, Valls G, González D, Prado A. Mucormicosis en una paciente diabética. Caso clínico y revisión de la literatura. Rev Chil Dermatol. 2010;26(2):148–53.

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