

Abcesso submandibular causado por *Listeria monocytogenes*: relato de um caso raro

Caso Clínico

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Resumo

A infeção por *Listeria monocytogenes* manifesta-se tipicamente com sintomas gastrointestinais autolimitados, neurolisteriose ou bacteremia. O envolvimento focal é muito raro. Neste artigo, descrevemos um caso de abcesso submandibular, que se apresentou no serviço de urgência com uma tumefação submandibular e dor progressivas, desde há 3 semanas, tendo sido diagnosticado concomitantemente com diabetes *mellitus* tipo 2. Iniciou-se tratamento com antibioterapia empírica e drenou-se o abcesso. Após 13 dias, foi feito o câmbio para uma antibioterapia dirigida com Penicilina-G intramuscular 24MU/dia, observando-se uma resolução completa do processo infeccioso. Este caso clínico destaca a necessidade de controlar os fatores de risco, drenar e realizar a cultura do pús, por forma a tratar efetivamente a infeção. Embora muito raro, devemos considerar este agente na presença de um abcesso cervical. Palavras-chave: Doenças da glândula submandibular; *Listeria monocytogenes*; Infeção focal; Casos clínicos

Introduction

The majority of submandibular space infections have an odontogenic etiology¹. Gram-positive anaerobic cocci are the most frequently isolated microorganisms². The aerobic and gram-positive bacillus *Listeria monocytogenes* (Lm) is responsible for a food-borne illness named listeriosis³. In healthy individuals, it typically presents as a relatively non-severe self-limiting gastrointestinal infection. However, among patients with underlying risk factors such as extremes of age, pregnancy and immunosuppression it can be more severe, presenting as meningitis and bacteremia³. Focal invasive infections with abscess formation are rare, mainly as a consequence of hematogenous seeding or direct inoculation⁴. Typically, they are only

diagnosed after the culture results become available⁵. We aim to present a case report of a rare focal manifestation of this infection, in accordance with the CARE reporting checklist⁶.

Case report

A 75-year-old male, with a history of chronic alcohol abuse, abstinent for the past month, and no regular medication, presented to our institution with a 3-week history of progressing pain and submandibular swelling. On physical examination, the patient also presented with paralysis of the mandibular branch of the facial nerve and a decayed single tooth on the lower left quadrant. His body temperature was 37.5°C, and all the other vital signs were within normal range. A contrast-enhanced computed tomography (CT) revealed a 1,7cm submandibular abscess, with parapharyngeal extension (Figure 1). From blood analysis it was

possible to do the inaugural diagnosis of type 2 diabetes *mellitus* (T2DM). A transcervical incision and drainage of the abscess was performed, and empirical intravenous treatment with amoxicillin plus clavulanic acid 1,2g 8/8 hours (h) and clindamycin 600mg 8/8h was started.

During hospitalization, an extraction of the root remnant in the third quadrant was performed. Blood cultures were negative. From the drained pus culture, Lm was isolated, and a mandatory notification has been made. At this time, a new anamnesis was carried out, with further deepening of social history, but there was no consumption of suspicious food. According to the antibiotic sensitivity test results, a targeted antibiotic therapy with intramuscular G-penicillin 24MU/day was initiated 13 days after admission. At this point, there was a clinical and imagiological significant improvement revealed by a new CT

Figure 1

Emergency department CT scan: Axial section contrast-enhanced CT scan performed in the emergency department, demonstrating a 1,7cm submandibular abscess



Figure 2

Hospital stay CT scan: Axial section contrast-enhanced CT scan performed on the 13th day of hospitalization, demonstrating improvement of submandibular abscess and an abscessed ganglion (arrow).



scan (Figure 2). After 16 days, the patient was discharged home on oral amoxicillin 1g 8/8h, over a 10-day period, with good adherence and tolerability. In follow-up appointments, a complete resolution of the infection was observed, however, the patient remained with sequelae of mandibular branch paralysis. Furthermore, the patient started treatment with metformin 850mg/day plus a basal-bolus insulin therapy and was referred to the general practitioner (GP) for further follow-up.

Discussion

Although the annual rate of sporadic listeriosis in Europe and North America is less than 1 per 100,000 population per year, this infection remains underdiagnosed³. As listeriosis has an unusual clinical presentation and morphological resemblance with other bacteria, the true prevalence remains unknown^{7,8}. The bacteria *Lm* is ubiquitous, leading to common contamination of the surface of meat and vegetables, at a rate of up to 15%³. It is most frequently found in dairy products and processed meat⁷. In this case, the source of infection was not found.

As mentioned before, focal invasive infection is rare. There are clinical cases describing endocarditis, myocarditis, pericarditis, arteritis, pneumonia, pleuritis, cholecystitis, peritonitis, liver/spleen abscess, brain abscess, breast abscess, arthritis, osteomyelitis, sinusitis, otitis, lymphadenitis, necrotizing fasciitis, conjunctivitis, and endophthalmitis^{7,9}. However, as far as our literature search could determine, only 3 articles mention unequivocally the presence of a cervical abscess⁴.

A recent systematic review described 19 culture-proven cases of *Lm* lymphadenitis, of which 44% had evidence of suppuration⁴. Taking into account the image results of our patient, we cannot exclude lymph node involvement. Given the negativity of blood cultures, the most probable route of infection is seeding from a locoregional portal of entry⁴. Therefore, in this clinical case, the submandibular abscess most probably results

from an extension of the periodontal infection. The predominant cervical involvement may be explained by a translocation in the mucosa-associated lymphoid tissue of the pharyngo-oral region⁴.

The immune response to *Lm* is cell-mediated, which means that conditions leading to T-cell suppression predispose to listeriosis. They include malignancies, HIV infection, chronic alcohol abuse, T2DM, extremes of age, and pregnancy⁹. Our patient had three risk factors, age over 65 years, chronic alcohol abuse and T2DM. According to Blot et al.⁴, the reported prevalence of this last disease among patients with lymphadenitis by *Lm* is 67%, being T2DM the most common associated risk factor. The same authors⁴ also highlighted the importance of evaluating for the presence of neoplasia, since it had a prevalence of 22%.

The first-line therapy against *Lm* is penicillin or ampicillin, alone or combined with an aminoglycoside. Alternatively, it can be administered trimethoprim-sulfamethoxazole¹⁰. In persons allergic to penicillin, vancomycin combined with an aminoglycoside may be used¹⁰. The administration of cephalosporins, quinolones, chloramphenicol, erythromycin and tetracycline should be avoided⁹. By consensus, treatment should last for 3 weeks or more¹⁰. In this case, the patient was initially treated empirically for 13 days, and then the antibiotics were switched to a targeted therapy for more 13 days.

Infections within the submandibular space can spread into the neck and mediastinum, making the involvement of this region very dangerous². Also, the mortality rate associated with listeriosis ranges from 20% to 30%¹⁰. These high values can be explained by a combination of immunocompromised patients and a delayed diagnosis³.

In conclusion, given its impact in public health, rarity, and high mortality rate, listeriosis is a notifiable disease. This clinical case highlights the need to drain abscesses, culture pus samples, and test antimicrobial susceptible patterns to treat the disease correctly and effectively. Although very rare, we must think

of Lm in the presence of a cervical abscess. Moreover, it is extremely important to control the identified risk factors.

Conflicts of interest

The authors declare that there is no conflict of interests regarding the publication of this paper.

Data confidentiality

The authors declare having followed the protocols in use at their working center regarding patients' data publication.

Privacy Policy, Informed Consent, and Ethics Committee Authorization

The authors declare that they have obtained written consent for the use of patient information in this article.

Protection of humans and animals

The authors declare that the procedures were followed according to the regulations established by the Clinical Research and Ethics Committee and to the 2013 Helsinki Declaration of the World Medical Association.

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Availability of scientific data

There are no datasets available publicly related to this work.

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