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Rare and life-threatening complication after an attempted lower third molar extraction: Lemierre syndrome

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ABSTRACT

Lemierre's syndrome is also known as the forgotten disease, and is a rare but life-threatening complication that can arise after surgical extractions of infected mandibular third molars. Owing to its rarity, oral and maxillofacial surgeons might not immediately recognise or can underestimate the pathological signs, and consequently do not apply the appropriate therapy to treat the syndrome. Here, we report on the occurrence and management of a case of Lemierre's syndrome, where the complications affected the right sigmoid sinus. Since the condition appear to be underreported and not properly highlighted, eventual systematic review and meta-analysis of the occurrence of the Lemierre's syndrome are highly recommended.

KEYWORDS

Lemierre syndrome – Third molar extraction – Surgical complication – Internal jugular vein – Thrombophlebitis

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Introduction

One of the complications that can follow infection of the oral–pharyngeal space is known as Lemierre's syndrome, or human necrobacillosis. These 'postanginal septicaemias' are characterised by thrombosis of the internal jugular vein and septic emboli metastasising in other organs.[1] This condition was first described in a systematic way in 1936 by Lemierre,[2] with the clinical signs of the syndrome including anaerobic septicaemia, fever status, dysphagia, neck pain and bilateral or unilateral cervical lymphadenopathy. In addition, induration of the internal jugular vein can occur slightly inferior to the anterior border of the sternocleidomastoid muscle.

Here, we report on a case of Lemierre's syndrome that followed an attempted lower third molar extraction, and where the jugular thrombosis extended to the homolateral sigmoid sinus.

Case history

A 39-year-old woman was referred from the emergency department suffering from a very large swelling of the submandibular cervical region on the right side (Fig 1a) together with fever and pain. She also complained of a headache on the occipital–parietal and temporal right side. After a general clinical evaluation, the patient was hospitalised in the maxillofacial surgery unit. According to her medical history, one month earlier she had undergone an incomplete a tooth extraction (Fig 1b). During the following four weeks, two types of antibiotics had been prescribed by the dentist, but the infection had not lessened.

Contrast-enhanced computed tomography performed on hospitalisation revealed a wide area of abscess that showed a vascularised wall and fluid–gaseous content. The dimensions of the abscess were 3 cm × 3 cm × 7 cm. It was localised to the right subpharyngeal and parapharyngeal spaces, at the C3–C5 level. At the C5 level, the abscess formation appeared to communicate with the lumen of the jugular vein (Fig 2a). The jugular vein appeared thrombosed with gaseous emboli in the cervical direction. At the cranial level, the right sigmoid sinus was not opaque and it was possible to detect a thrombosis of the vessel (Fig 2b).

Therapy was by both pharmacological and surgical approaches. The pharmacological therapy included antibiotic

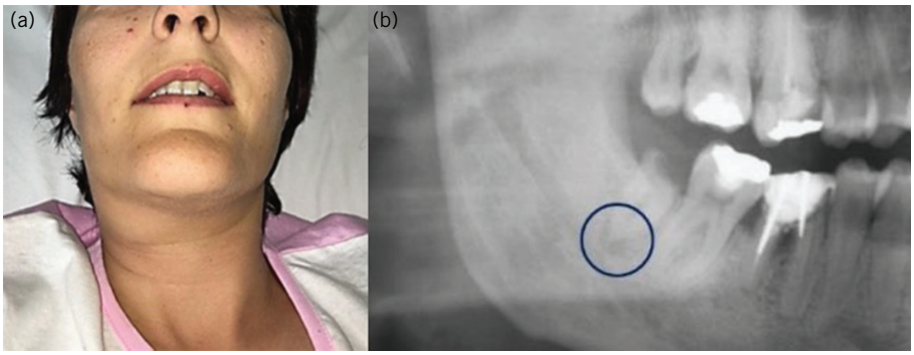

Figure 1 Extra oral (A) clinical presentation of the patient at admission. (B) Pretreatment panoral radiograph showing the causative tooth.

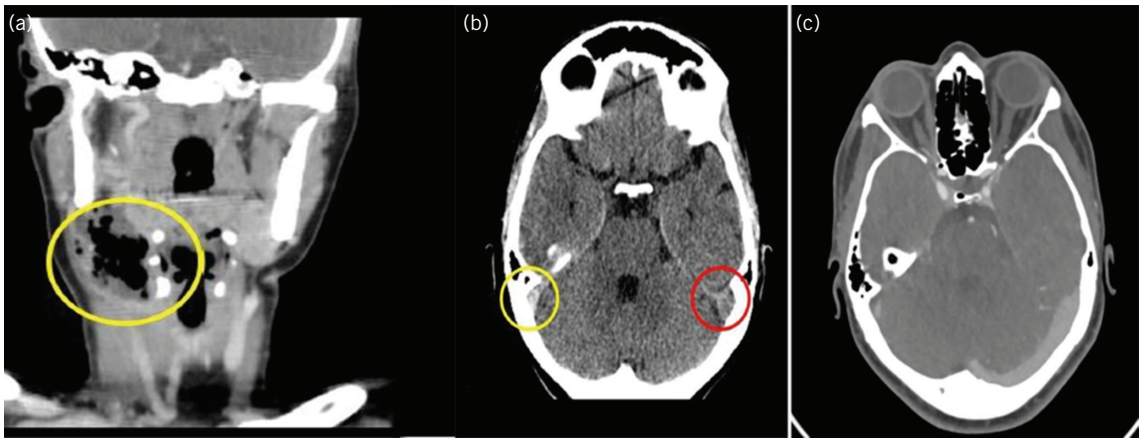

Figure 2 (A) Contrast-enhanced computed tomography of the abscess with jugular vein thrombosis. (B) The vessel thrombosis at the level of the sigmoid sinus. (C) One-year of follow-up, showing the full recovery of the patient.

administration: clindamycin 1 g intravenously every 8 hours, imipenem 500 mg intravenously every 8 hours and parnaparin 0.6 ml every 12 hours. The surgical approach included draining the abscess to release the purulent material. Culturing of the purulent material confirmed the susceptibility of the microorganisms to the antibiotic therapy, which was therefore continued. The patient was also monitored by the neurology unit due to the thrombosis that affected the right sigmoid sinus. The patient underwent anticoagulant therapy for six months, as warfarin 5 mg/day.

After two months, the maxillofacial team performed the surgical extraction of the tooth and full curettage of the surrounding bone. The one-year computed tomography follow-up showed complete recovery of the area (Fig 2c).

Discussion

Currently, the incidence of Lemierre's syndrome has been reported to range from 0.6 to 2.3 per million, with a two to one male to female predilection.[3] Lemierre's syndrome is characterised by thrombophlebitis of the internal jugular vein, owing to the anatomical course of the tributary trunk receiving the blood drainage from the pharyngeal and pterygoideal regions. Thus, an infection can migrate along this vascular tract to the internal jugular vein.

The clinical signs of thrombophlebitis of the internal jugular vein are cervical lymphadenopathy in the anterior triangle of the neck at the angle of the mandible anteriorly and parallel to the sternocleidomastoid muscle.[3,4] In addition, patients affected by Lemierre's syndrome can show hypotension, tachycardia, low oxygen saturation, leucocytosis, high levels of C-reactive protein and high blood levels of the liver enzymes.[4]

Contrast-enhanced computed tomography can reveal the thrombophlebitis of the internal jugular vein and the diagnosis of Lemierre's syndrome can be confirmed by the microbial analysis of the septic emboli.

The recommended antibiotic therapy, according to the literature, involves the administration of carbapenem and

piperacillin/tazobactam alone or in combination with metronidazole, for four weeks.[4] The use of anticoagulants, particularly as low-molecular-weight heparin, is still under debate.[4,5] To date, only one group is working on a systematic review and meta-analysis to clarify the treatment of the complication of Lemierre's syndrome.[5] Since the condition appears to be underreported and not properly highlighted, an eventual systematic review and meta-analysis of the occurrence of the Lemierre's syndrome is highly recommended.

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