Available online at www.sciencedirect.com

ScienceDirect

British Journal of Oral and Maxillofacial Surgery 57 (2019) 1153–1155

BRITISH Journal of Oral and Maxillofacial Surgery

www.bjoms.com

Short communication

Lemierre syndrome leading to ankylosis of the temporomandibular joint

O. Johnson King *, V. Sharma

Great Ormond Street Hospital

Accepted 16 October 2019
Available online 8 November 2019

Abstract

Lemierre syndrome, also known as Lemierre's disease is a rare condition that was first described by Andre Lemierre in 1936. We present a case of a 3-year-old boy who presented with ankylosis of the right temporomandibular joint (TMJ) secondary to a mastoid infection as part of Lemierre syndrome. His ankylosis resulted in restricted mouth opening, which had a considerable impact on his speech and ability to eat. Surgical treatment comprising right coronoidectomy, gap arthroplasty of the TMJ, and interpositional fat grafting, enabled him to move his jaw and function has returned. This case report highlights the consequences of Lemierre syndrome and the need for clinicians to be aware of its features and the impact it may have on patients.

Keywords: Lemierre's syndrome; Mastoiditis; Temporomandibular joint ankylosis; Coronoidectomy; Gap arthroplasty; Interpositional fat graft

Introduction

Lemierre syndrome is caused by *Fusobacterium necrophorum* and is characterised by thrombophlebitis of the internal jugular vein secondary to oropharyngeal infection.[1,2] Thrombophlebitis can cause unilateral swelling and tenderness along the sternocleidomastoid muscle and lead to septic emboli, persistent fever, and postanginal septicaemia with metastatic infections in the lungs and bones. Some authors think that the primary foci of Lemierre's are the ears, mastoids, and teeth.[3]

The syndrome is thought to affect 0.8 million people a year. It was once called the "forgotten disease" because of its rarity, but its incidence is now thought to be increasing as a result of antibiotic resistance. It tends to affect young adults, with both sexes affected equally, and is usually treated with anticoagulants, antimicrobials, and operation. The sequelae can be life threatening, and most patients require admission to hospital. Mortality is thought to be 5%.[4]

Case report

A 3-year-old boy presented to our department with ankylosis of the right temporomandibular joint (TMJ) secondary to mastoiditis and Lemierre syndrome. The ankylosis had been recognised when he was admitted for an arthrogram of his left hip under general anaesthesia at a different hospital and the anaesthetist discovered that they could not open his mouth. Before this the patient had had multiple episodes of choking. His medical history included Lemierre syndrome, thrombosis of the internal jugular vein, and mannose-binding lectin deficiency, which is known to increase the susceptibility to infection.[5] Lemierre syndrome had caused osteomyelitis and septic arthritis. On clinical examination he had marked facial

* Corresponding author.
E-mail addresses: olivia.johnson-king@nhs.net (O. Johnson King), valmiki.sharma@gosh.nhs.uk (V. Sharma).

https://doi.org/10.1016/j.bjoms.2019.10.306

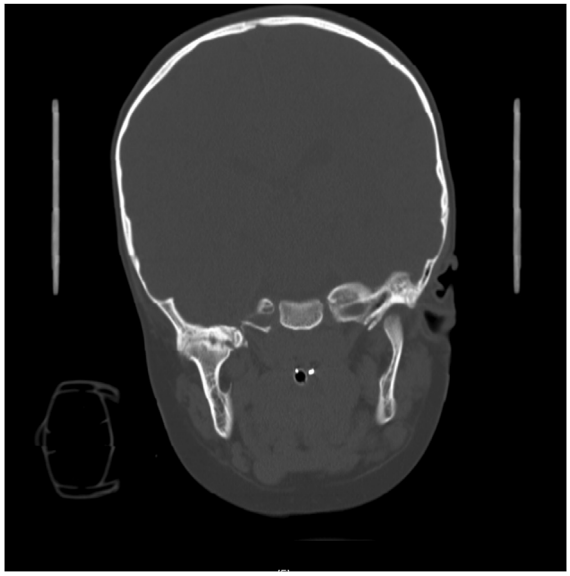

Fig. 1. Coronal computed tomogram of the head showing ankylosis of the right temporomandibular joint and glenoid fossa.

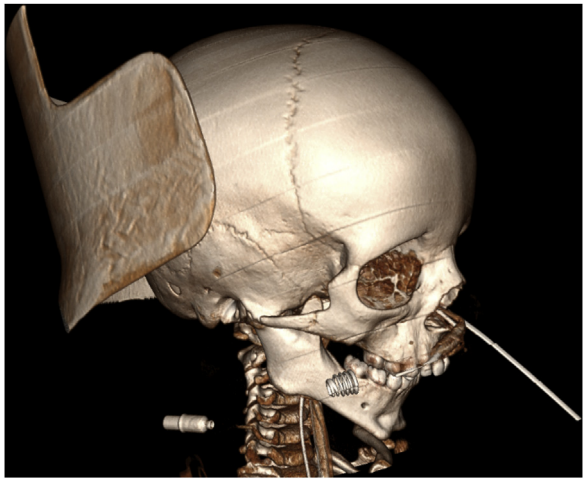

Fig. 2. Three-dimensional reconstruction of the computed tomogram of the head showing the prominent coronoid process that contributed to the limited opening.

asymmetry and his interincisal mouth opening was 1 mm secondary to ankylosis of the TMJ (Figs. 1 and 2).

He was treated with gap arthroplasty, right coronoidectomy, and interpositional fat grafting. At the end of the operation his mouth opening had increased to 33 mm (Fig. 3).

He was seen two weeks postoperatively and had recovered well. The postoperative swelling had subsided and he was able to tolerate soft foods. We gave him a children's TheraBite® Jaw Motion Rehabilitation System™ (Atos Medical) and he had speech and language therapy. As a result, his mouth opening was maintained and there were no clinical concerns. We will continue to monitor him at regular follow-up appointments.

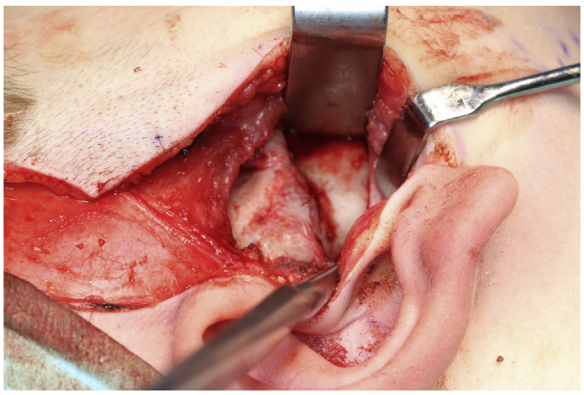

Fig. 3. Al-Kayat Bramley incision used to access the right temporomandibular joint, and ankylosis of the right condyle and glenoid fossa of the temporal bone.

Discussion

To our knowledge there are no published reports of ankylosis of the TMJ secondary to Lemierre syndrome. Ankylosis most commonly occurs as a result of trauma and infection and is a known complication after mastoiditis, particularly in children. Several management techniques have been suggested, but the results have not been uniformly successful.[6] The management protocol at our hospital involves access, usually by a preauricular incision with temporal extension, resection of the ankylotic mass, ipsilateral coronoidectomy (contralateral coronoidectomy if necessary), and interpositional tissue transfer if appropriate (fat/temporalis muscle).

Other authors have suggested the use of autogenous costochondral grafts for reconstruction of the TMJ.[7]

We are aware that the restoration of normal function can be challenging after ankylosis of the TMJ, and we acknowledge the importance of close follow up to ensure the early recognition of postoperative complications. It is likely that our patient will have long-term growth disturbance because of the history of damage and operation, and we appreciate that the incidence of recurrence is high and that he will probably require further operations, which may include distraction osteogenesis, costochondral grafting, and orthognathic surgery. In the long term he will probably need prosthetic replacement of the joint.

We hope that this case has highlighted the importance of an early diagnosis of Lemierre syndrome, and has raised awareness of the impact that it can have on patients' lives.

Conflict of interest

We have no conflicts of interest.

Ethics statement/confirmation of patient's permission

Ethics approval was not applicable. Written consent to publish these details was obtained from the patient's parents.

References

1. Lemierre A. On certain septicæmias due to anaerobic organism. *Lancet* 1936;**227**:701–3.
2. Moreno S, Garcia Altozano J, Pinilla B, et al. Lemierre's disease: postanginal bacteremia and pulmonary involvement caused by Fusobacterium necrophorum. *Rev Infect Dis* 1989;**11**:319–24.
3. Hagelskjaer LH, Prag J, Malczynski J, et al. Incidence and clinical epidemiology of necrobacillosis, including Lemierre's syndrome, in Denmark 1990–1995. *Eur J Clin Microbiol Infect Dis* 1998;**17**:561–5.
4. Karkos PD, Asrani S, Karkos CD, et al. Lemierre's syndrome: a systematic review. *Laryngoscope* 2009;**119**:1552–9.
5. Turner MW. The role of mannose-binding lectin in health and disease. *Mol Immunol* 2003;**40**:423–9.
6. Khadka A, Hu J. Autogenous grafts for condylar reconstruction in treatment of TMJ ankylosis: current concepts and considerations for the future. *Int J Oral Maxillofac Surg* 2012;**41**:94–102.
7. Awal DH, Charan G, Ball R, et al. Unilateral and bilateral temporomandibular joint reconstruction in children using autogenous costochondral grafting: An outcome study of 60 patients over a 28-year period. *Br J Oral Maxillofac Surg* 2016;**54**:e157.