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Case illustrated

Thrombophlebitis of the external jugular vein: A variant of Lemierre's syndrome

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A previously healthy 41-year-old woman presented with a 4-day history of right mandibular pain, toothache, and trismus, followed by fever and chills. Her fever was recorded at 41.0°C. Physical examination revealed swelling and redness that extended from the patient's right cheek to the lower area of her jaw, with inability to open her mouth by one cm. Contrast-enhanced computed tomography showed abscesses in the root of the right mandibular second molar and right parapharyngeal and submandibular space (Figs. 1 and 2), right external jugular vein (EJV) thrombosis (Fig. 1), and septic pulmonary emboli (Fig. 3). She underwent extraction of the right mandibular second molar, aspiration of thrombus in the right EJV, and surgical drainage of the abscesses. Cultures of blood and the EJV thrombus were positive for alpha-hemolytic *Streptococcus*, thereby leading to a diagnosis of right EJV thrombophlebitis. Ampicillin sulbactam was administered for four weeks, and the patient was discharged without complications.

A combination of thrombophlebitis of the internal jugular vein (IJV) and septic emboli is classically termed Lemierre's syndrome (LS), which is rare and caused by an anaerobic organism following an oropharyngeal infection [1]. Thrombophlebitis may be caused by bloodstream infection, while direct invasion from soft tissue infection or spread via the lymphatic pathway is also anticipated [2]. Although rare, several variants of LS with thrombophlebitis of the facial vein, subclavian vein, cavernous sinus, sigmoid sinus, and EJV have been reported. The involvement of the EJV is anatomically

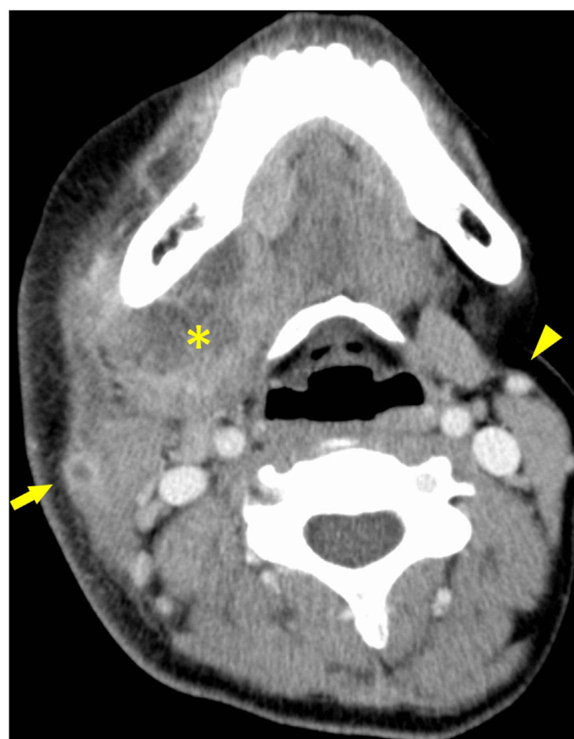

Fig. 1. Contrast-enhanced computed tomography showing the thrombosis in the right external jugular vein (arrow), an abscess in the right parapharyngeal to the submandibular space (asterisk), and left, external jugular vein which was intact (arrowhead).

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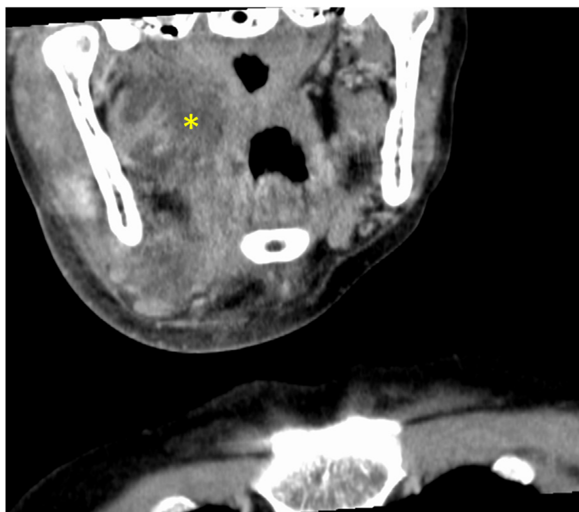

Fig. 2. Contrast-enhanced computed tomography showing the abscesses extending from the right parapharyngeal to the submandibular space (asterisk).

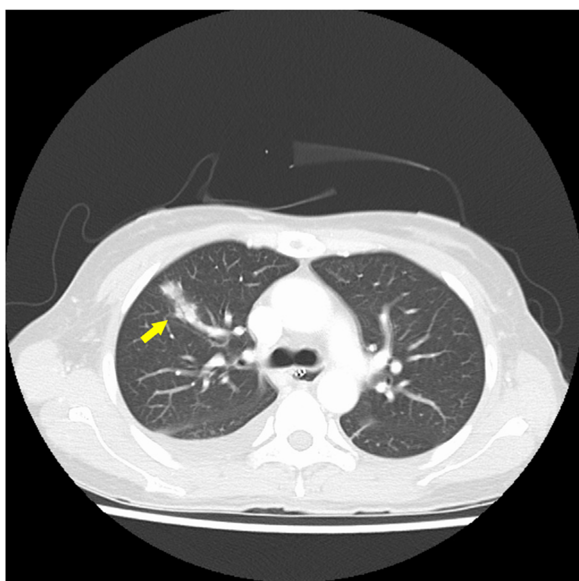

Fig. 3. Contrast-enhanced computed tomography showed consolidation in the S3 region of the upper lobe in the right lung (arrow).

unexpected and has been reported less frequently [3]. As the EJV is not typically supplied from the veins of the oral cavity and tonsils, thrombophlebitis caused by bloodstream infection is less likely to develop in the EJV than in the IJV. However, a variant connection of the veins of the oral cavity or tonsils to the EJV has been reported [4], which could be the cause of isolated EJV involvement. Our case thus represented a rare variant of LS that involved the EJV alone. Further, no causative organism was identified in the EJV thrombus in previous cases involving the EJV, whereas we identified alpha-hemolytic streptococcus in the EJV thrombus.

Surgical intervention is considered when septic emboli develop despite prolonged treatment with antibiotics, the recommended duration of which is 3–6 weeks [1]. Anticoagulation for thrombi may be beneficial in patients who are refractory to antibiotic treatment or have intracranial thrombi or hemophilia [5]. In our case, we did not perform anticoagulation for the cervical thrombus.

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Consent

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CRediT authorship contribution statement

Yasuhiro Suzuki: Conceptualization, Data curation, Writing - original draft. **Akira Kuriyama:** Conceptualization, Data curation, Supervision, Writing - original draft, Writing - review & editing. **Satoshi Tsuruta:** Writing - review & editing.

Declaration of Competing Interest

The authors report no declarations of interest.

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