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Case Report

A case of intracranial vasospasm in a patient with extensive retropharyngeal cellulitis ☆,☆☆,★

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ABSTRACT

Retropharyngeal cellulitis/abscesses are deep neck infections that may become life-threatening if airway compromise occurs. This condition is more common in children than in adults, and associated intracranial vessel narrowing has been reported. We report an adult patient with extensive retropharyngeal cellulitis and intracranial vasospasm. The patient was a 62-year-old woman who presented with fever, sore throat, and neck pain. She also had uncontrolled type 2 diabetes mellitus. Leukocytosis, prolonged erythrocyte sedimentation rate, elevated C-reactive protein, and hyperglycemia were present on admission. Computed tomography and contrast-enhanced magnetic resonance imaging revealed severe swelling in the nasopharyngeal, retropharyngeal, prevertebral, and bilateral carotid spaces. Gadolinium enhancement extended to the middle cranial fossa and visceral space. Multiple stenoses in several intracranial vessels was also identified. Intravenous antibiotic therapy was initiated, the patient's symptoms resolved, and repeat imaging confirmed improvement. Intracranial vasospasm should be considered in patients with retropharyngeal cellulitis.

Introduction

Retropharyngeal cellulitis/abscess is a relatively uncommon infection in adolescents, but much more common in children aged between 2 and 4 years [1,2], with an incidence of 0.10–0.22 per 10,000 children [3]. Retropharyngeal cellulitis/abscesses are deep neck space infections that can become an immediate life-threatening emergency with catastrophic complications [4]. Retropharyngeal lymphadenitis and abscess has been associated with a narrowed ipsilateral cervical part of the internal carotid artery (ICA) in children [5]; however, to our

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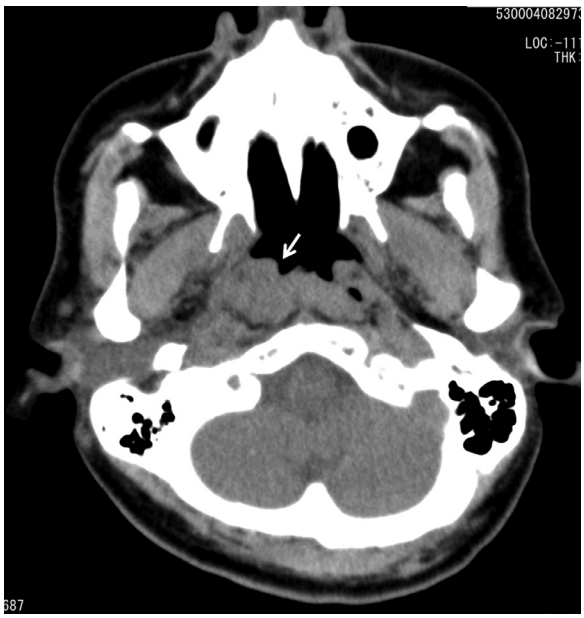

Fig. 1 – CT images of the neck obtained the day before the patient's admission. The images show swelling of the right posterior nasopharyngeal wall (white arrow) and no apparent abscess formation.

knowledge, no reports have described intracranial vasospasm caused by retropharyngeal cellulitis/abscess.

We report a rare case of a woman with intracranial vasospasm and extensive retropharyngeal cellulitis.

Case report

A 62-year-old Japanese woman presented with fever and progressively worsening sore throat and neck pain for 3 days. She also had type 2 diabetes mellitus, which was treated with oral medication, but was uncontrolled. Her medical and family history were otherwise unremarkable.

On admission to the otolaryngology department of our hospital, her temperature was 38.4°C, and her pulse was regular at 111 beats/min. Physical examination revealed swelling of the posterior pharyngeal wall suggestive of pharyngeal abscess and upper vocal cord swelling.

On laboratory examination, the patient's white blood cell count was 19.6 × 10^9/L (normal range: 3.3–9.0 × 10^9/L), the C-reactive protein concentration was 26.19 mg/dL (normal concentration: ≤ 0.1 mg/dL), and the erythrocyte sedimentation rate was > 140 mm/h (normal range: 3–15 mm/h). Her fasting blood sugar level was 287 mg/dl (normal range: 73–109 mg/dL), and her HbA1c level was 9.0% (normal range: 4.9%–6.0%).

Conservative treatment with appropriate antibiotics was performed instead of surgical drainage because her clinical condition was stable except for the mild fever, sore throat, and neck pain. Antibiotic therapy was instituted with piperacillin sodium (2 g once daily) and clindamycin (1200 mg once daily), and therapy was continued for 3 weeks. The patient was discharged after 3 weeks of intravenous antibiotic therapy and had no complaints after 2 months of follow-up.

Neck computed tomography (CT) was performed to evaluate the patient's sore throat and neck pain before the patient's admission and showed swelling of the right posterior nasopharyngeal wall and no apparent abscess formation (Fig. 1). We then performed contrast-enhanced neck and brain magnetic resonance imaging (MRI) to determine the site and extent of the infection 11 and 13 days, respectively, after the patient's admission. Gadolinium-enhanced neck MRI showed severe swelling and ill-defined enhancement of the bilateral nasopharynx. This enhancement extended into the retropharyngeal space, prevertebral space, and bilateral carotid space. The enhancement also extended to the dura matter of the left middle cranial fossa and visceral space. Moreover, edema and fluid retention were observed in the retropharyngeal space at the C1–C4 level (Fig. 2). These findings suggested extensive spread of infection. Brain MRI showed multiple stenoses in bilateral ICAs at the level of C1–C2, and in the middle cerebral

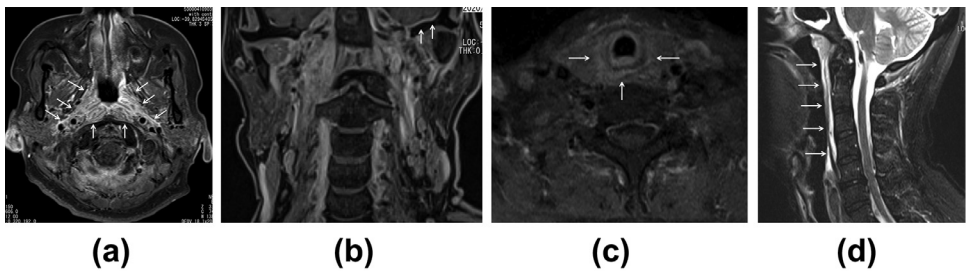

Fig. 2 – Neck contrast-enhanced MR image obtained 11 days after the patient's admission. (A) Gadolinium-enhanced MRI, T1 FATSAT sequence, axial section showing ill-defined enhancement of the bilateral nasopharynx, which extended into the retropharyngeal space, prevertebral space, and bilateral carotid space (white arrows). (B) Gadolinium-enhanced MRI, T1 FATSAT sequence, coronal section showing enhancement extending to the dura matter of the left middle cranial fossa (white arrows). (C) Gadolinium-enhanced MRI, T1 FATSAT sequence, axial section showing mild enhancement extending to the visceral space (white arrows). (D) MRI, short TI inversion recovery (STIR) sequence, sagittal section, showing edema and fluid retention in the retropharyngeal space at the C1–C4 level (white arrows).

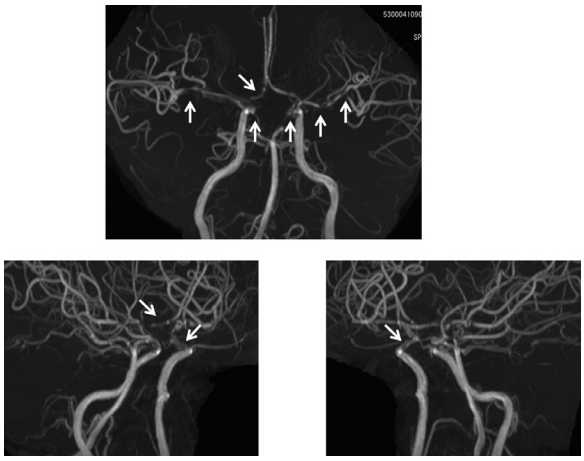

Fig. 3 – Brain contrast-enhanced MR image obtained 13 days after the patient's admission. The MR angiography (maximum intensity projection, MIP) image shows multiple stenosis of bilateral ICAs at C1–C2, MCAs at M1–M2, and right ACA at A1 (white arrows).

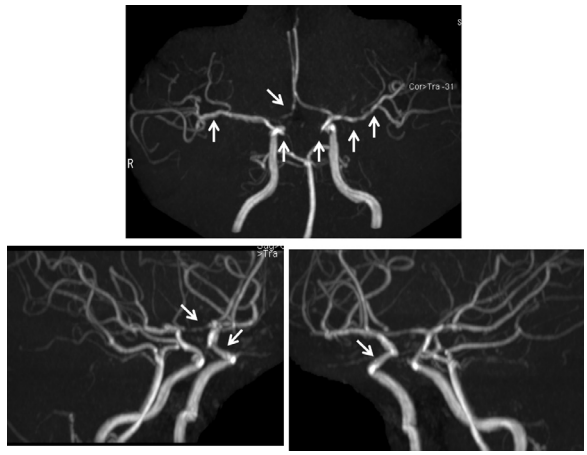

Fig. 5 – Follow-up brain contrast-enhanced MR image obtained 11 days after the first brain MRI. MR angiography (MIP) image showing reduction in the multiple stenoses in bilateral ICAs, MCAs, and right ACA (white arrows).

artery (MCA) at M1–M2 and the right anterior cerebral artery (ACA) at A1 (Fig. 3).

Her clinical symptoms and laboratory examination findings improved gradually, and we performed follow-up neck and brain MRI 13 and 11 days, respectively, from the first MRI examinations. Follow-up neck gadolinium-enhanced MRI showed improvement of the severe swelling and ill-defined enhancement of the bilateral nasopharynx. Enhancement of the dura matter of the left middle cranial fossa and visceral space had also disappeared (Fig. 4). Follow-up brain MRI showed a decrease in the multiple stenoses in bilateral ICAs, MCAs, and the right ACA (Fig. 5).

Written informed consent was obtained from the patient to publish this case report.

Discussion

Retropharyngeal abscesses are most commonly seen in children aged 2–4 years [2], but they have been reported at any age, including in infants and adults [6]. In adults, the common causes of retropharyngeal infections include penetrating trauma, odontogenic sepsis, and peritonsillar abscess. Our patient had no apparent history of penetrating trauma, but did have diabetes. The risk factors for odontogenic infections are immunosuppression, such as with HIV infection and chronic steroid use, and diabetes [7]. Therefore, the retropharyngeal cellulitis may have been caused by odontogenic sepsis. The common clinical presentation of retropharyngeal abscess is acute or subacute neck pain, fever, sore throat, neck mass, respiratory distress, dysphagia, odynophagia, limited neck range

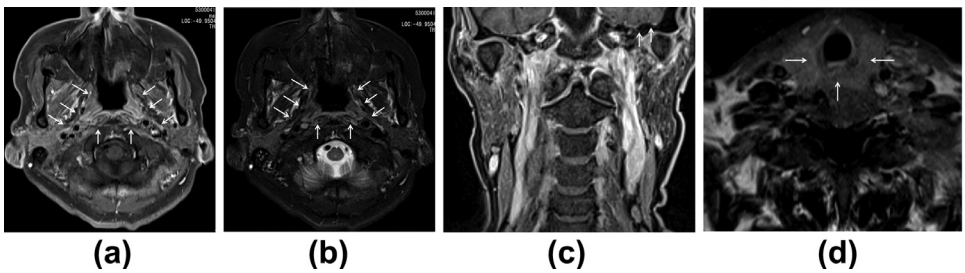

Fig. 4 – Follow-up neck contrast-enhanced MR image obtained 13 days from the first neck MRI. (A) Gadolinium-enhanced MRI, T1 FATSAT sequence, axial section, showing significant reduction in the previously-seen ill-defined enhancement of the bilateral nasopharynx that extended into the retropharyngeal space, prevertebral space, and bilateral carotid space (white arrows). (B) MRI, T2 FATSAT sequence, axial section, showing improved edema of the bilateral nasopharynx that previously extended into the retropharyngeal space, prevertebral space, and bilateral carotid space (white arrows). (C) Gadolinium-enhanced MRI, T1 FATSAT sequence, coronal section showing that the previous enhancement extending to the dura matter of the left middle cranial fossa has disappeared (white arrows). (C) Gadolinium-enhanced MRI, T1 FATSAT sequence, axial section, showing improvement of the previous mild enhancement that had spread to the visceral space (white arrows).

of motion, and torticollis [2,8]. Regarding diagnostic imaging, contrast-enhanced CT has high sensitivity and is usually essential to confirm the location and extent of deep neck space abscesses [9]. Gadolinium-enhanced MRI can depict the accurate progression of abscesses in soft tissues [10].

Retropharyngeal abscess can become an immediate life-threatening emergency, with the potential for airway compromise and other catastrophic complications [4]. Hudgins et al reported that retropharyngeal lymphadenitis and abscess were associated with a narrowed ipsilateral cervical part of the ICA in children [5]. Moreover, Lisan et al reported a case of infectious arteritis of the ICA complicating retropharyngeal abscess in an adult [11]. However, no reports have described retropharyngeal cellulitis/abscess caused by stenosis of remote intracranial arteries.

The cerebral vasculature, in particular the pial vessels, is densely supplied with noradrenergic sympathetic nerve fibers mainly originating in the superior cervical ganglion (SCG) that accompany the carotid artery and project into the ipsilateral hemisphere [12-14]. Intracerebral vessels narrow in response to cervical sympathetic stimulation and dilate when these fibers are interrupted. Miriam et al [15]. reported that patients with symptoms owing to cerebral vasospasm after aneurysmal subarachnoid hemorrhage benefited from transient cervical sympathetic block at the level of the SCG. In anatomic reports [16,17], the SCG is located ahead of the longus capitis and longus colli muscles and behind the ICA at approximately the C2 level. In our case, the retropharyngeal cellulitis extended extensively into the retropharyngeal space, prevertebral space, and bilateral carotid space. The cellulitis also spread vertically from the dura matter of the left middle cranial fossa to the visceral space. Therefore, the related inflammation may have spread to bilateral SCGs near the ICAs. As a result, the intracerebral arteries may have narrowed in response to cervical sympathetic stimulation. After antibacterial treatment, this intracranial vasospasm decreased with improvement of the retropharyngeal cellulitis. To the best of our knowledge, this is the first report of intracranial vasospasm in a patient with retropharyngeal cellulitis. Moreover, the most important point illustrated by our case is that it emphasizes the need to consider the possibility of an intracranial vasospasm in the work up of patients with retropharyngeal cellulitis. However, this case report has a limitation in that the findings are limited to a single case, and generalization may be difficult. Moreover, mechanistic insight on how retropharyngeal cellulitis may provoke intracranial vasospasm is required in further studies.

Supplementary materials

Supplementary material associated with this article can be found, in the online version, at doi:10.1016/j.radcr.2021.06.047.

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