To the Editor: In the past decade, there were increasing reports with radiologic and ultrasonographic findings arguing that idiopathic carotidynia may well be a distinctive disease entity characterized by unilateral neck pain, contrast-enhanced signal surrounding carotid artery or thickening of carotid wall, and excellent response to non-steroidal anti-inflammatory drug or steroid therapy.[1-4] Here, we reported a case of imaging proved carotidynia with rare clinical manifestations as vocal hoarseness, swallowing difficulty, and syncope in addition to unilateral neck pain and headache.

A 71-year-old female patient was admitted with pain in the left neck and temporal areas for 10 days, accompanied by hoarse voice, swallowing difficulty, and episodes of transient loss of consciousness. The patient developed mild-moderate dull headache 10 days before admission, and the dull headache evolved to localized sharp pain in the left temporal area of head and the upper part of neck with worsening intensity. She also complained vocal hoarseness and transient aggravation of pain accompanied by choking and coughing upon swallowing. She experienced two episodes of transitory loss of consciousness and collapse of posture over last 2 days. The fainting attack lasted 2 to 3 min, with pale appearance and sweating but no limb jerks. The medical history was not remarkable.

Neurologic examination on admission revealed that she was clear with painful appearance and hoarse voice, and showed tenderness to palpation in the neck corresponding to the path of carotid artery on the left side. The gag reflex was retained but she reported tenderness in response to touching the left-side laryngeal wall using a tongue depressor. Palpitation in the carotid bulb area failed to induce syncope attack. General examination was otherwise normal. Cervical magnetic resonance imaging (MRI) showed high T2-weighted signals of soft tissue and enhancement of tissue between the carotid artery and internal jugular vein in the left carotid sheath [Figure 1A–1D]. The abnormal signals extended from the proximal to distal part of common carotid artery with maximal abnormal signal in the proximal segment surrounding about 1/2 of the vessel circumference. Cranial and cervical computed tomography angiography (CTA) was largely normal, with no sign of aneurysm, dissection, or stenosis in the left carotid artery except a small atherosclerotic-calcified plaque in the starting segment of internal carotid [Figure 1E]. Esophageal barium meal examination reported unsymmetrical piriform recess and slight dysfunction of the epiglottis movement without structural abnormality in esophagus. Cranial MRI and cervical spine MRI showed obsolete mild multiple lacunar infarction lesions, chronic sinusitis, and cervical degenerative alterations. Duplex ultrasound evaluation of the carotid vessels found a hyperchoic plaque (8.2 mm × 2.4 mm) in the initial segment of the left internal carotid artery without stenosis or dissection. Electroencephalogram was normal. Laboratory studies revealed that high-sensitive C-reactive protein, erythrocyte sedimentation rate, rheumatoid factor, complement C3, and complement C4 were within the normal range; rheumatism indicators including anti-nuclear antibody spectrum and anti-neutrophil cytoplasm auto-antibodies were negative; serum antibodies to syphilis, Lyme disease, hepatitis B, and hepatitis C were all negative; complete blood cell count, routine liver and kidney function tests, and thyroid function test were normal.

The patient was treated intravenously with corticosteroid (dexamethasone 10 mg daily). The pain in the neck and head resolved over next 3 days and the bulbar palsy-like symptoms improved gradually and complete recovery was observed upon discharge 11 days after the treatment. Follow-up MRI scan 1 month later showed the high-
intensity signal in the left carotid sheath partially resolved [Figure 1F], while the patient remained asymptomatic.

The clinical features of our patient included left neck pain radiating to ipsilateral temporal and laryngeal region, tenderness to palpation in the left neck overlying the carotid artery, high-intensity signal on T2-weighted imaging, and contrast enhancement surrounding the carotid and internal jugular vein in the carotid sheath and good response to steroid treatment, with no other accountable structural damages on MRI or CTA, fulfilling the diagnostic criteria of idiopathic carotidynia of International Headache Society in 1988 as well as the recently diagnostic criteria of idiopathic carotidynia (“idiopathic carotiditis”) proposed by Tardy et al. Conditions resembling idiopathic carotidynia, including vascular as well as non-vascular origins, should be excluded to establish the diagnosis. For the current case, there were no indications suggesting carotid dissection, aneurysm, sclerotic stenosis, and other structure damages in MRI, CTA, and ultrasound studies. The lack of relevant clinical features and negative results in the comprehensive screening laboratory investigations for rheumatoid disease and secondary infectious vasculitis made the impossibility of vasculitis affecting large arteries such as Takayasu arteritis, giant-cell arteritis and secondary vasculitis related to Syphilis, Lyme disease, and tuberculosis for this patient.

Apart from confirming the findings about idiopathic carotidynia in literature, our case had several additional features. First, clinical symptoms were more complex than previously reported. In addition to typical unilateral neck pain radiating to temporal and laryngeal region, our patient also had vocal hoarseness, choking, and coughing upon swallowing food and fainting attacks. We speculated that these symptoms might result from the involvement of vagus nerve and glossopharyngeal nerve in the carotid sheath inflammatory damage. The fainting attack might be caused by over-sensation of carotid sinus and increased baroreceptor stimulation. Sato et al. reported a case of carotidynia with fainting attacks, but no other vagus nerve and glossopharyngeal-nerve-related symptoms were presented in this patient. The second feature worthy of noting in the present case was the extensive pathologic damage along the route of carotid sheath as demonstrated in MRI. In previously reported cases, abnormal signals of imaging findings in idiopathic carotidynia mostly confined to the segment near bifurcation of carotid artery. For our patient, the high-intensity signals in MRI extended from proximal part to the distal part in the carotid sheath [Figure 1D], indicating the inflammatory damage in idiopathic carotidynia may involve other segments except carotid bifurcation in carotid sheath, more extensive than previously assumed.

The case provided evidence that the clinical features for idiopathic carotidynia might be more complicated than previously reported. In addition to typical unilateral neck pain radiating to temporal and laryngeal region, patient may also presented with vocal hoarseness, choking and...
coughing, and fainting attacks which may result from the involvement of vagus nerve and glossopharyngeal nerve in the inflammatory damage in carotid sheath.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has her consent for her images and other clinical information to be reported in the journal. The patient understands that her names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

None.

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