CASE REPORT

Trousseau’s Syndrome due to Anaplastic Thyroid Carcinoma Presenting with Multiple Ischemic Strokes

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Abstract

Trousseau’s syndrome is characterized by a cerebral or systemic thromboembolism caused by coagulation abnormalities in malignancy. We herein report a case of multiple ischemic strokes as the initial manifestation of anaplastic thyroid carcinoma (ATC). An 86-year-old man was admitted to our hospital due to a sudden-onset weakness of the left limbs. Brain magnetic resonance imaging revealed multiple ischemic lesions in the right middle cerebral artery territory and a mass in the left frontal lobe. Computed tomography revealed a thyroid mass and multiple lung tumors. A diagnosis of ATC was confirmed by a thyroid biopsy. Our case indicates that ATC should be considered as a cause of Trousseau’s syndrome.

Key words: Trousseau’s syndrome, anaplastic thyroid carcinoma, coagulation abnormality, ischemic stroke

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Introduction

Trousseau’s syndrome is characterized by a cerebral or systemic thromboembolism caused by coagulation abnormalities in malignancy (1) and it often occurs in patients with adenocarcinoma of the lung, pancreas or ovary, but rarely in those with thyroid carcinoma (2). We herein report a case of Trousseau’s syndrome due to anaplastic thyroid carcinoma (ATC) in a patient presenting with multiple ischemic strokes.

Case Report

An 86-year-old man, who had experienced trouble concentrating and calculating for several weeks, was admitted to our hospital for a sudden-onset weakness of the left limbs with a consciousness disturbance. He had no risk factors for a stroke except for hypertension.

On admission, he exhibited a conjugate deviation of the eyes to the right, mild dysarthria and left hemiparesis. He was disoriented in terms of the time and place. His Glasgow coma scale score was E4V4M6, and his Japan coma scale score was 2. Neurological examinations showed left lateral gaze palsy, left facial palsy, paralysis of the left limbs and left hemispatial neglect. The NIH stroke scale score was 17. Brain magnetic resonance imaging revealed ischemic lesions in the right middle cerebral artery territory and a mass in the left frontal lobe (Fig. 1). Neck and chest computed tomography revealed a thyroid mass and multiple lung tumors (Fig. 2). Vessels of the neck and chest were not apparently compressed by the thyroid mass. Abdominal computed tomography showed no remarkable abnormalities. Atrial fibrillation was not demonstrated by Holter electrocardiography, and transthoracic echocardiography revealed no apparent causes of a cardiac embolic stroke. Carotid artery ultrasoundography did not show stenosis or atherosclerosis. Blood examinations showed elevated levels of D-dimer (11.63 μg/mL) and fibrin degradation products (50.4 μg/mL), whereas the levels of other coagulation factors were within the normal range. With regard to thyroid tumor markers, the thyroglobulin level was slightly elevated (171 ng/mL), whereas the calcitonin level was within the normal range (46 pg/mL). The results of thyroid function tests were normal. We were unable to characterize the thyroid tumor by a fine-needle aspiration biopsy because the sample did not contain a sufficient number of tumor cells. Therefore, we performed a thyroid biopsy on day 15 of hospitalization. The biopsy...
specimen exhibited a sarcomatoid appearance. The thyroid-specific markers thyroid transcription factor-1 and thyroglobulin were undetectable. We confirmed a diagnosis of ATC after detecting cell adhesion molecule 5.2 (CAM5.2), an endothelial marker, in the tumor cells (Fig. 3).

We intended to start anticoagulation therapy after the bi-
Table. Cases of Trousseau’s Syndrome due to Thyroid Carcinoma.

<table>
<thead>
<tr>
<th>Reference</th>
<th>Age (year)</th>
<th>Histology</th>
<th>Initial manifestation</th>
<th>Treatment</th>
<th>Prognosis</th>
</tr>
</thead>
<tbody>
<tr>
<td>3</td>
<td>69</td>
<td>MTC</td>
<td>Cerebral venous sinus thrombosis, deep vein thrombosis</td>
<td>Surgery, anticoagulation</td>
<td>No recurrence</td>
</tr>
<tr>
<td>4</td>
<td>55</td>
<td>PTC</td>
<td>Internal jugular vein thrombosis</td>
<td>Surgery, radioactive iodine, anticoagulation</td>
<td>No recurrence</td>
</tr>
<tr>
<td>5</td>
<td>59</td>
<td>MTC</td>
<td>Nonbacterial endocarditis</td>
<td>Surgery, anticoagulation</td>
<td>Deceased</td>
</tr>
<tr>
<td>6</td>
<td>37</td>
<td>PTC</td>
<td>Multiple Ischemic strokes</td>
<td>Surgery, anticoagulation</td>
<td>No recurrence</td>
</tr>
<tr>
<td>Present case</td>
<td>86</td>
<td>ATC</td>
<td>Multiple Ischemic strokes</td>
<td>None</td>
<td>Deceased</td>
</tr>
</tbody>
</table>

MTC: medullary thyroid carcinoma, PTC: papillary thyroid carcinoma, ATC: anaplastic thyroid carcinoma

opsy, but were unable to do so because the patient developed pseudomembranous colitis on day 16 of hospitalization, which led to disseminated intravascular coagulation, such as hypofibrinogenemia and a prolonged prothrombin time. He died of an intracranial hemorrhage from the metastatic brain tumor on day 34 of hospitalization. An autopsy was not performed because of the family’s refusal.

Discussion

This case highlights that thyroid carcinoma can cause Trousseau’s syndrome, which was initially reported to occur in mucin-producing adenocarcinoma, although not all cases are associated with this type of malignancy (1). Four previously published case reports (3-6) have described Trousseau’s syndrome due to thyroid carcinoma (Table); Trousseau’s syndrome was associated with medullary thyroid carcinoma (MTC) in two cases and with papillary thyroid carcinoma (PTC) in the other two. Each of these patients received anticoagulation therapy and underwent surgery. Three patients experienced no recurrence or additional thromboembolic events after the surgery, whereas the fourth with MTC developed lung metastases and died eight months after the initial presentation. To the best of our knowledge, the present case is the first case of Trousseau’s syndrome due to ATC to be reported.

There are two major causes of an ischemic stroke in patients with malignancy: nonbacterial thrombotic endocarditis and intravascular coagulation (7). Additionally, the compression or occlusion of cerebral vessels by a tumor can cause ischemic stroke (8). In the present case, the patient had no obvious risk factors for ischemic stroke, and the thyroid tumor did not compress the adjacent vessels; therefore, we considered that the coagulation abnormalities seen in malignancy caused the multiple ischemic strokes. However, the mechanism underlying the ischemic stroke in this patient was not clearly identified because we were unable to perform invasive testing, such as transesophageal echocardiography.

In conclusion, thyroid carcinoma can cause Trousseau’s syndrome. When Trousseau’s syndrome is clinically suspected, then thyroid carcinoma should be considered as a potential cause.

The authors state that they have no Conflict of Interest (COI).

Acknowledgement

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References