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Solitary fibrous tumor with aneurysmal bone cyst–like change in the middle cranial fossa: illustrative case

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BACKGROUND Solitary fibrous tumor (SFT) is a rare mesenchymal tumor known for its propensity for recurrence and metastasis. Furthermore, aneurysmal bone cyst (ABC) is a benign osteolytic lesion. ABC-like areas can be seen in bone tumors that have undergone hemorrhagic cystic change. They are formed by disruptions in the osseous circulation caused by the associated lesion. The most common associated lesions are giant cell tumor, chondroblastoma, osteoblastoma, osteosarcoma, chondromyxoid fibroma, and fibrous dysplasia. There has been no reported case of SFT being the associated lesion.

OBSERVATIONS A 42-year-old woman presented with a 6-month history of headache and impaired memory. Radiological examinations revealed a 50-mm cystic lesion with multiple fluid levels arising from the left temporal bone. Total resection of the tumor was conducted, and postoperative course was uneventful. Histopathological examination was consistent with SFT with ABC-like change.

LESSONS This is the first documented case of SFT with ABC-like change in the cranial fossa. This should be considered a differential diagnosis when treating a lesion in the cranial fossa, such as in this case, to achieve complete resection of the tumor and have close follow-up postoperatively.

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KEYWORDS aneurysmal bone cyst; solitary fibrous tumor; left middle cranial fossa

Solitary fibrous tumor (SFT) is a rare mesenchymal tumor known for its propensity for recurrence and metastasis.[1] It was first described in the visceral pleura and later described in other sites such as pericardium, peritoneum, lung, liver, mediastinum, and meninges.[2] Meningeal SFT comprises approximately 0.4% of all brain tumors. It is typically dural based and CD34-positive.

Aneurysmal bone cyst (ABC) is a benign osteolytic tumor–like lesion, originally described by Jaffe and Lichtenstein in 1942.[3] It is more common in females and in patients younger than 20 years.[4] Approximately 50% of ABCs occur in the metaphysis of long bones and 20% occur in the spine. Only 3%–6% of the lesions develop in the bones of the cranium.[5] Traumatic or anomalous venous disruption in the osseous diploe causes the lesion to expand and present as ABC.

ABC-like areas can be seen in benign and malignant bone tumors that have undergone hemorrhagic cystic change.[6] They were previously referred to as "secondary ABC." They are formed by disruptions in the osseous circulation caused by the associated lesion. The most common lesions are giant cell tumor, chondroblastoma, osteoblastoma, osteosarcoma, chondromyxoid fibroma, and fibrous dysplasia.

In this report, we discuss our experience with a rare case of SFT with ABC-like change in the left temporal bone.

Illustrative Case

History and Examination

A 42-year-old woman presented with a 6-month history of headache and impaired memory that her family perceived approximately a month ago. She had no history of trauma or surgery. Her past medical history was unremarkable and she was taking no medication. Her neurocognitive examinations showed low level of verbal memory. Her scores for standard verbal paired-associate (VPA) learning test were

ABBREVIATIONS ABC = aneurysmal bone cyst; CT = computed tomography; MRI = magnetic resonance imaging; SFT = solitary fibrous tumor; VPA = verbal paired-associate.

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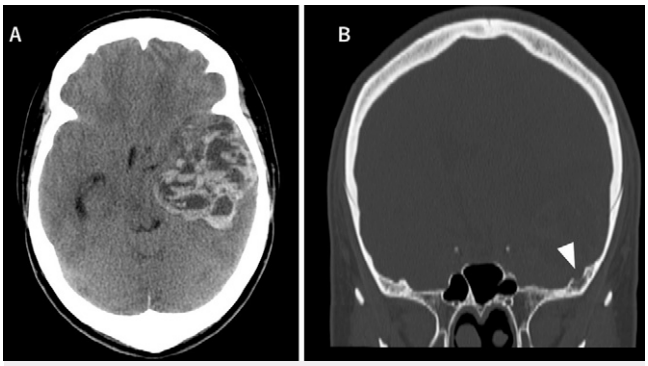

FIG. 1. A: Axial CT scan showed a 50-mm cystic lesion with multiple fluid levels arising from the left temporal bone. **B:** Coronal CT scan showed erosion of the inner table of the temporal bone (*arrowhead*).

0-1-1 for related words and 0-0-0 for nonrelated words. Her neurological examination was otherwise negative.

Computed tomography (CT) demonstrated a 57 × 50 × 50-mm cystic lesion with multiple fluid levels arising from the left temporal bone. The inner table of the left temporal bone was discontinued due to the erosion of the tumor. Midline shift was observed as well (Fig. 1). Magnetic resonance imaging (MRI) showed a well-circumscribed extraaxial tumor in the left temporal fossa. Multiple cystic lesions were formed, and a T1-weighted sequence showed strong peripheral and septal enhancement of the tumor, which was consistent with ABC (Fig. 2).

Angiography showed multiple feeders from the left middle meningeal artery; therefore, the patient received feeder and tumor embolization followed by total resection of the tumor 2 days later.

Surgical Procedure

Left temporal craniotomy and zygomectomy was conducted to fully expose the tumor in the middle fossa. The tumor originated from the temporal bone at the middle fossa, causing disconnection of the inner table. The tumor was red-purple in color and penetrated the dura mater. Almost the entire part of the tumor extended into the subdural space. Because the dura was tense, dural incision was made directly above the tumor, and the tumor was resected microscopically. The tumor was coagulated and detached at the dura of the middle fossa and then discreetly removed from the surface of the temporal lobe. The tumor was cystic, filled with blood, and the cyst walls were relatively tough. The tumor and the dura attached to the tumor were removed completely, and the temporal bone was drilled out to expose the normal bone tissue. Dural defect was covered using collagen-based dural graft matrix, and the cranial defect at the temporal base was covered with mesh plate.

The postoperative course was uneventful. Postoperative MRI showed no residual tumor, and follow-up CT scans showed improvement of the brain shift (Fig. 3). The patient's verbal memory returned

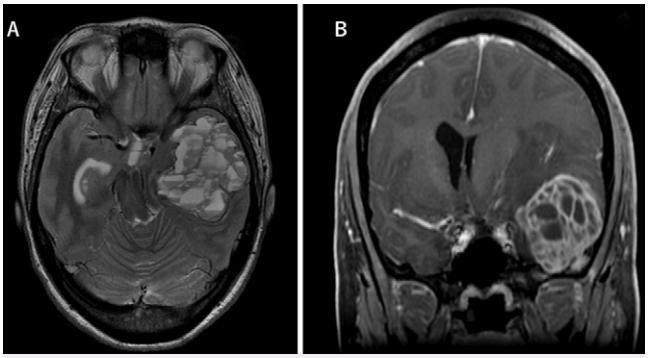

FIG. 2. A: Axial T2-weighted MRI sequence showed a well-circumscribed extraaxial tumor in the left temporal fossa. Multiple cystic lesions were formed. **B:** Coronal postcontrast T1-weighted sequence showed strong peripheral and septal enhancement.

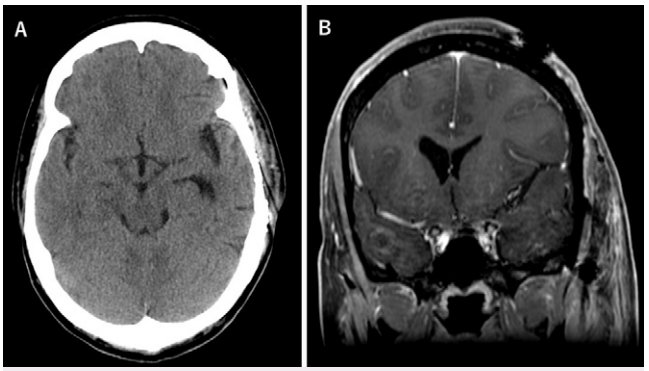

FIG. 3. A: Postoperative axial CT scan showed improvement of the brain shift. **B:** Coronal postcontrast T1-weighted MRI sequence showed no residual tumor.

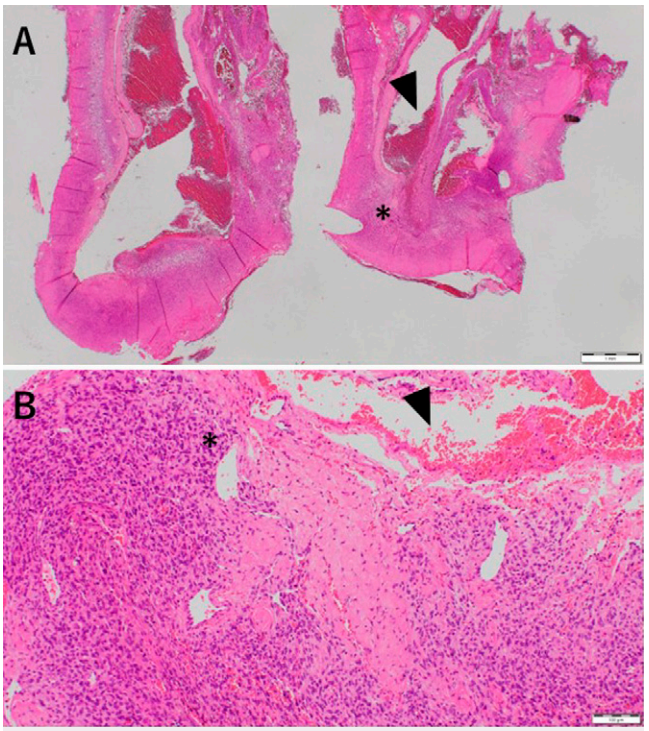

FIG. 4. Original magnification ×1.25 (**A**) and ×10 (**B**). Hematoxylin and eosin stain demonstrates hypercellular stroma, which consisted of spindle cells with thin-walled microvessels, staghorn vessels, and collagen fibers (*asterisks*), were shown. There were also dilated vessels with blood cells inside that formed cystic cavity (*arrowheads*).

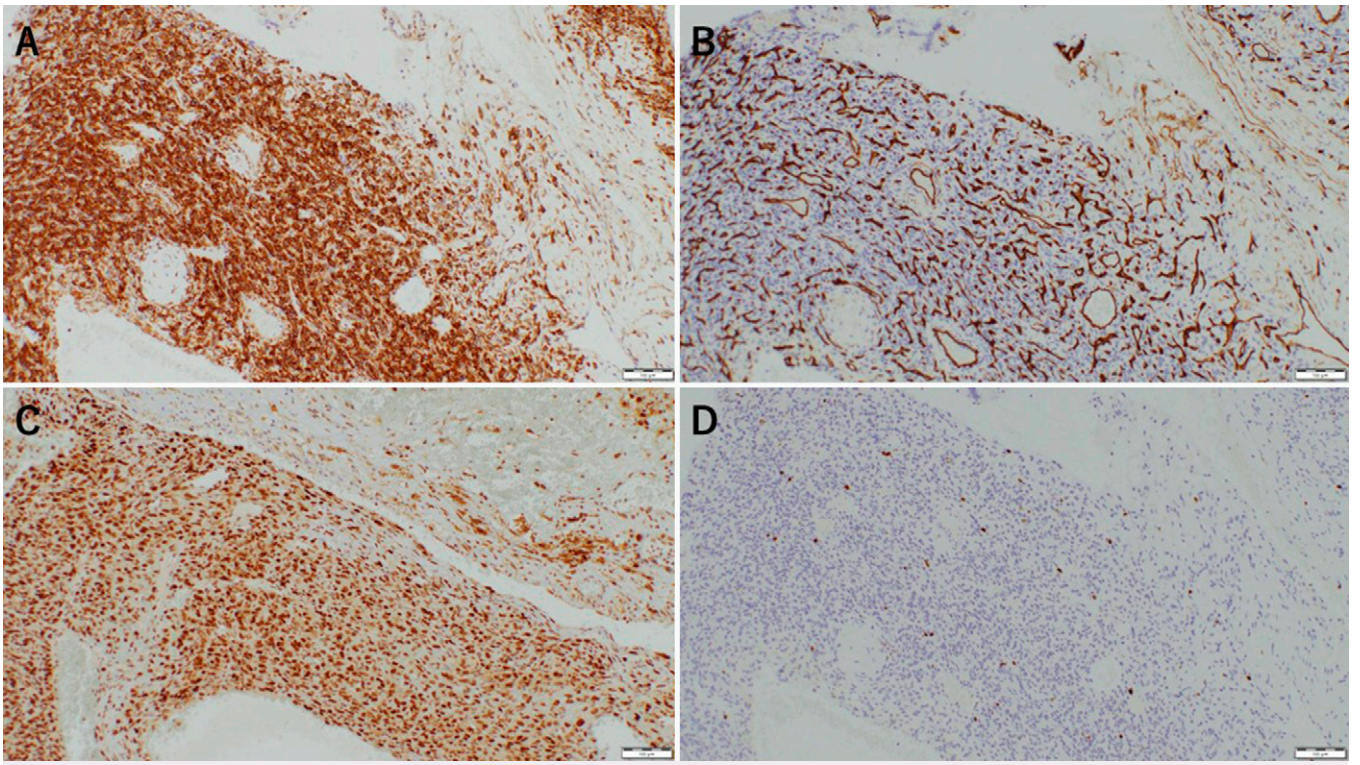

FIG. 5. Immunohistochemical analysis with bcl-2 (**A**), CD34 (**B**), STAT6 (**C**), and Ki-67 (**D**) (original magnification ×10, all panels). Spindle cells showed positive for bcl-2, partially positive for CD34, and positive for STAT6 (**C**). Ki-67 was positive in 2% of the cells.

to the standard level. Her postoperative VPA scores were 8-10-10 for related words and 1-4-6 for nonrelated words. She was discharged on postoperative day 13. Three months after surgery, she has experienced no complications.

Pathological Examination

Histopathological examination demonstrated hypercellular stroma, which consisted of spindle cells with thin-walled microvessels, staghorn vessels, and collagen fibers, suggesting SFT. There were also dilated vessels with blood cells inside that formed cystic cavity, which was consistent with ABC (Fig. 4). However, osteoblasts or osteoclasts, which are typical in ABCs, were not observed.

Immunohistochemistry of spindle cells showed positive for vimentin, STAT6, bcl-2, and CD99 and showed partially positive for CD34. EMA, AE1/AE3, desmin, and p53 were negative, and Ki-67 was positive in 2% of the cells (Fig. 5). This finding was consistent with SFT with ABC-like change.

Discussion

Observations

A few cases of ABCs in the cranial bone have been reported, but very few past papers report ABC-like changes in the cranial bone that developed in association with neoplasms.[7,8] These associated neoplasms are commonly giant cell tumor, chondroblastoma, osteoblastoma, osteosarcoma, chondromyxoid fibroma, and fibrous dysplasia. Capillary venous malformation with ABC-like change in a 5-year-old boy[7] and a few ABC-like lesions associated with calvarial osteoblastomas have been reported.[9–11] Gutierrez et al. reported 49 cases of ABC-like lesions associated with primary neoplasms, regardless of location.[12] Among them, 4 cases were located in the cranial bone, and their associated neoplasms were fibrous dysplasia, osteoblastoma, osteosarcoma, and chondromyxoid fibroma. Among all 49 cases, none were associated with SFTs. To date, no documents have reported SFT with ABC-like change.

SFT resembles other spindle cell tumors both radiologically and histopathologically; therefore, it can be misdiagnosed easily. As in our case, immunohistochemical studies are necessary for its differentiation.

Lessons

SFT may be the associated neoplasm of ABC-like lesion. Acknowledging this possibility enables surgeons to consider SFT with ABC-like change as a differential diagnosis when treating a lesion in the cranial fossa such as in this case and aim for total resection of the tumor. Preoperative radiological examination suggested ABC, and histopathological examination showed typical characteristics of ABC-like area and SFT. Immunohistochemical examinations lead to the definitive diagnosis of SFT with ABC-like change. This may be misdiagnosed as meningioma or ABC, but by diagnosing it as SFT, we ensure close follow-up for recurrence or metastasis in the future. Careful preoperative examination, complete resection of the tumor, and close postoperative observation are essential.

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Disclosures

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

Author Contributions

Conception and design: Yoshikawa, Matsuhashi. Acquisition of data: Yoshikawa, Matsuhashi. Analysis and interpretation of data: all authors. Drafting the article: Matsuhashi. Critically revising the article: Yoshikawa, Matsuhashi. Reviewed submitted version of manuscript: Yoshikawa, Matsuhashi. Approved the final version of the manuscript on behalf of all authors: Yoshikawa. Administrative/technical/material support: Yoshikawa. Study supervision: Yoshikawa.

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