Pulsatile Scalp Mass Induced by Head Injury: Angiolymphoid Hyperplasia with Eosinophilia – a Case Report –

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A 38-year-old woman had hit the right front of her head in a traffic accident six years earlier, suffered a scalp hematoma, and an induration had remained at the point of contact. Six years later, the induration suddenly started to swell and pulsate, so she came to the hospital. When she visited the hospital, the mass was 3 cm × 3.5 cm, pink on the surface, hard, and the pulsation disappeared when we pressed the superficial temporal artery (STA) anterior to the right external auditory meatus. Three-dimensional (3D) computed tomography (CT) angiography did not clearly show continuity between the mass and the STA, but we suspected a traumatic STA aneurysm and operated on her. Intraoperative findings showed that the mass was not an aneurysm and that, although it did not have direct continuity with the main trunk of the STA, the mass was accompanied by numerous blood vessels seemingly branching from STA branches. Pathological findings indicated the proliferation of small vessels with endothelial cells called epithelioid endothelial cells and the infiltration of eosinocytes and lymphocytes around the vessels, and led us to a diagnosis of angiolymphoid hyperplasia with eosinophilia (ALHE). Although the pathogenesis of this disease remains unknown, past studies suggested that one cause might be damage to blood vessels by trauma. We suggest that ALHE, rare as it is, should be considered in the differential diagnosis of a pulsatile scalp mass occurring after a head injury.

Key words: angiolymphoid hyperplasia with eosinophilia, head injury, pulsatile scalp mass, epithelioid endothelial cell, three-dimensional CT angiography

Introduction

Angiolymphoid hyperplasia with eosinophilia (ALHE) is a rarely seen vascular lesion that mainly occurs under the head or neck skin and whose pathogenesis remains unknown. We experienced a case of angiolymphoid hyperplasia with eosinophilia on the right forehead caused by head injury, which was difficult to distinguish preoperatively from a traumatic superficial temporal artery (STA) aneurysm.

Case report

A 38-year-old woman had bumped her head on the windshield in a traffic accident six years earlier and suffered a subcutaneous hematoma. A subcutaneous induration had remained where she had bumped her head. Six years later, the induration suddenly started to swell and pulsate, and gradually grew larger, so she came to the hospital.

Neurological examination revealed no deficit. The scalp mass was about 3 cm × 3.5 cm under the right forehead and the skin of the lesion was light pink (Figure–1). The mass was hard, pulsatile, and was not aching. The pulsation disappeared when we manually pressed the STA anterior to the right external auditory meatus. No superficial lymphadenopathy was evident. Routine laboratory studies showed no abnormality other than iron-deficiency anemia.

In computed tomography (CT) without contrast medium, the mass showed isodensity and, in CT with
contrast medium, showed homogeneous enhancement (Figure-2A). No abnormality was seen in the brain parenchyma or the skull. In three-dimensional (3D) CT angiography, the periphery of the STA was not well visualized and continuity with the mass was not clearly visualized (Figure-2B). In 3D-CT angiography, the relationship between the mass and STA was not clearly shown, but clinical findings suggested a traumatic STA aneurysm.

According to the wishes of the patient, a surgical excision was performed under general anesthesia. A semi-circular skin incision was made around the mass, which lay in the subcutaneous tissue. The boundary between the subcutaneous fatty tissue and the mass was relatively clear. There was no direct continuity with the main trunk of the STA, but there were numerous blood vessels around the mass seemingly branching from STA branches. We cut these vessels after coagulation hemostasis and removed the entire mass. There was little bleeding. The mass was hard and elastic, and the cut surface did not show any vessel structure that would have suggested an aneurysm.

Pathological findings indicated the proliferation of small vessels and the infiltration of lymphocytes and eosinocytes (Figure-3A). The endothelial cells of the small vessels had a large nucleus and abundant cytoplasm. The cells were arranged like cobblestones, protruded into the lumen and narrowed the space. Thrombi were observed, too. These endothelial cells seemed to be epithelioid endothelial cells (Figure-3B). Arteriovenous (AV) shunts (Figure-3C) and breakage of arterial walls with elastic lamina disruption were also observed (Figure-3D). From the above findings, the case was diagnosed as an angiolymphoid hyperplasia with eosinophilia. Immunohistochemically, a few IgG4-positive plasma cells were detected (4.8 cells/HPF). And the IgG4-positive/IgG-positive ratio of the plasma cells indicated 14%.

The patient has been without recurrence for about one year.

Discussion

Angiolymphoid hyperplasia with eosinophilia (ALHE) is a relatively rare disease that was first
reported by Wells and Whimster in 1969. In many cases, the lesion presents a nodular papule with a light pink or red brown surface and lies under the skin or in the subcutaneous tissue. Most frequently, it occurs among women in their 20s to 40s around the auricle, on the forehead or temples. The lesion can ache when pressed, or pulsate, beitchy, or bleed spontaneously. About half of the lesions are multicentric. Twenty per cent of cases are associated with peripheral blood eosinophilia, and some with regional lymphadenopathy.

Characteristically, pathological findings indicate the proliferation of small vessels with epithelioid endothelial cells and the infiltration of inflammatory cells such as eosinocytes and lymphocytes around the small vessels. Epithelioid endothelial cells are cuboidal or columnar cells having a large nucleus and abundant cytoplasm, arranged like cobblestones, and protrude into the lumen and narrowed the space. Arteriovenous (AV) shunts and thrombi are found in 40% and 20% of cases.

The pathogenesis of ALHE remains unknown, but opinions are broadly divided into two groups of causes: benign neoplasms, or a reactive vascular proliferation due to some reason. Recently, the latter theory has found more support after the publication of reports on frequent occurrence after a trauma like the present case, or associated with insect bites, infection, or hormonal imbalance. In the present case, the patient had suffered a head injury in a traffic accident six years earlier and the area suddenly and rapidly enlarged, and suffered from ALHE, so we think that the trauma was involved.

Vadlamudi and Schinella reported four cases of scalp mass accompanied by capillary vascular proliferations that occurred 1 to 10 months after the patient had a trauma. Since all of the cases showed intima and media proliferation accompanied by myxoid degeneration in small or medium-sized arteries and the lesion was seen also in traumatic pseudoaneurysm, they suspected that the trauma caused an inflammatory reactive proliferation of blood vessels accompanied by myxoid pseudoaneurysm or invasion of eosinocytes, which eventually induced ALHE. Further, as we have seen above, vascular malformation such as AV shunt was observed in 40% and thrombi in 20% of cases.
Busquest et al. suggested that the damage of local blood vessels due to trauma induced vascular malformation\(^2\). Fernandez supposed that, to improve the ischemic change of the lesion caused by the presence of AV shunts, renin is locally secreted and stimulates the proliferation of vascular endothelial cells\(^3\). These vascular changes may have secondarily caused the cell invasion of eosinocytes and lymphocytes. Considering that the present case also showed breakage of arterial walls with elastic lamina disruption, AV shunts, and thrombi, we judged that vascular damage due to the trauma was a cause of this disease.

Before the operation, we diagnosed the case as a traumatic STA aneurysm, because the mass was pulsatile and the pulsation disappeared when the STA anterior to the right external auditory meatus was pressed. We thought that the mass was pulsatile because of significant vascular proliferation. Most frequently, a traumatic STA aneurysm occurs 2 to 6 weeks after the head injury\(^{12}\) and rarely occurs after such a long period as six years, as in the present case. We should also have considered the fact that, in the preoperative 3D–CT angiography, continuity with STA was not clearly shown.

As to diagnostic imaging, few reports provide details. In external carotid angiography, the lesion is visualized as a hypervascular mass with clear boundaries that is deep-stained gradually from the arterial phase to the capillary phase and to the venous phase\(^4\). CT with contrast medium demonstrates homogeneously enhanced subcutaneous mass\(^5\). In the present case, 3D–CT angiography could not clearly visualize the relationship between the STA and the mass, but suggested the possibility of a mass receiving blood-flow from the STA. We find that 3D–CT angiography is a useful diagnostic imaging method that can be applied relatively noninvasively to the patient.

As the treatment method, surgical excision is generally used\(^5\)\(^2\)\(^3\)\(^11\)\(^13\). Other methods include steroid administration\(^7\), radiotherapy\(^6\), endovascular treatment\(^2\), and pulsed-dye laser treatment\(^14\). Each of these treatments is effective, but about one-third of the lesions recur\(^2\)\(^3\)\(^15\). It is also reasonable to observe the lesion for 3 to 6 months, because sometimes it regresses spontaneously\(^2\)\(^3\)\(^15\). In the present case, the lesion was entirely removed and has not recurred for about one year, but should be carefully followed up in the future for possible recurrence.

In this paper, we reported a case of frontal ALHE that occurred six years after the patient suffered a head injury. The case suggests that this disease should be considered in the differential diagnosis of a pulsatile scalp mass occurring after a head injury.

Conflict of interests

The authors declare no conflict of interest associated with this manuscript.

References