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Lipoma of the Corpus Callosum Associated with Distal Anterior Cerebral Artery Aneurysm A Case Report

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ABSTRACT

We report what may be the first case of lipoma of the corpus callosum associated with a distal anterior cerebral artery saccular aneurysm. The pathogenetic correlation between the callosal lipoma and the distal anterior cerebral artery saccular aneurysm is discussed.

Key words: *Callosal lipoma, Distal anterior cerebral artery aneurysm, Pathogenesis*

Intracranial lipomas are rare. The corpus callosum is the most frequent site of their location, accounting for more than one-half of the cases[26]. Approximately 50% of lipomas of the corpus callosum were associated with dysgenesis such as agenesis of the corpus callosum, median cleft nose, cleft lip, spina bifida, myelomeningocele, funnel chest, cranial bone defect, agenesis of the cerebellar vermis, pituitary tumor and webbed toes[16,22,26]. Anomalies of the anterior cerebral artery and fusiform dilatation of the pericallosal artery have been established to be angiographic findings of callosal lipoma[4,5,9,22,23]. In our review of the literature, however, no case of lipoma associated with a distal anterior cerebral artery (DACA) saccular aneurysm could be found. In this paper a case of lipoma of the corpus callosum associated with a DACA saccular aneurysm is presented and the pathogenetic correlation between the callosal lipoma and the DACA aneurysm is discussed.

CASE REPORT

A 52-year-old woman was admitted to Hiroshima General Hospital after generalized convulsive seizure. She had a fainting attack two years before. Neurological examination revealed no abnormalities. Mental faculties were normal. There were no signs of intracranial hypertension. Plain skull radiographs showed a mulberry shaped calcification in the deep mid-frontal region (Fig. 1). Computerized tomogra-

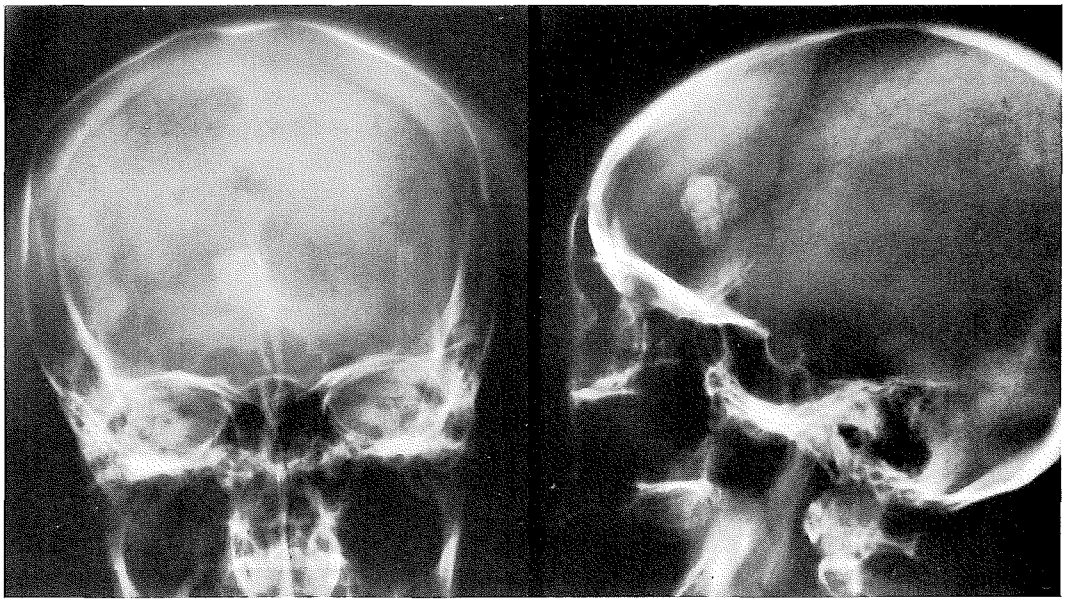

Fig. 1. Plain skull radiographs showing mulberry shaped calcification in the deep mid-frontal region.

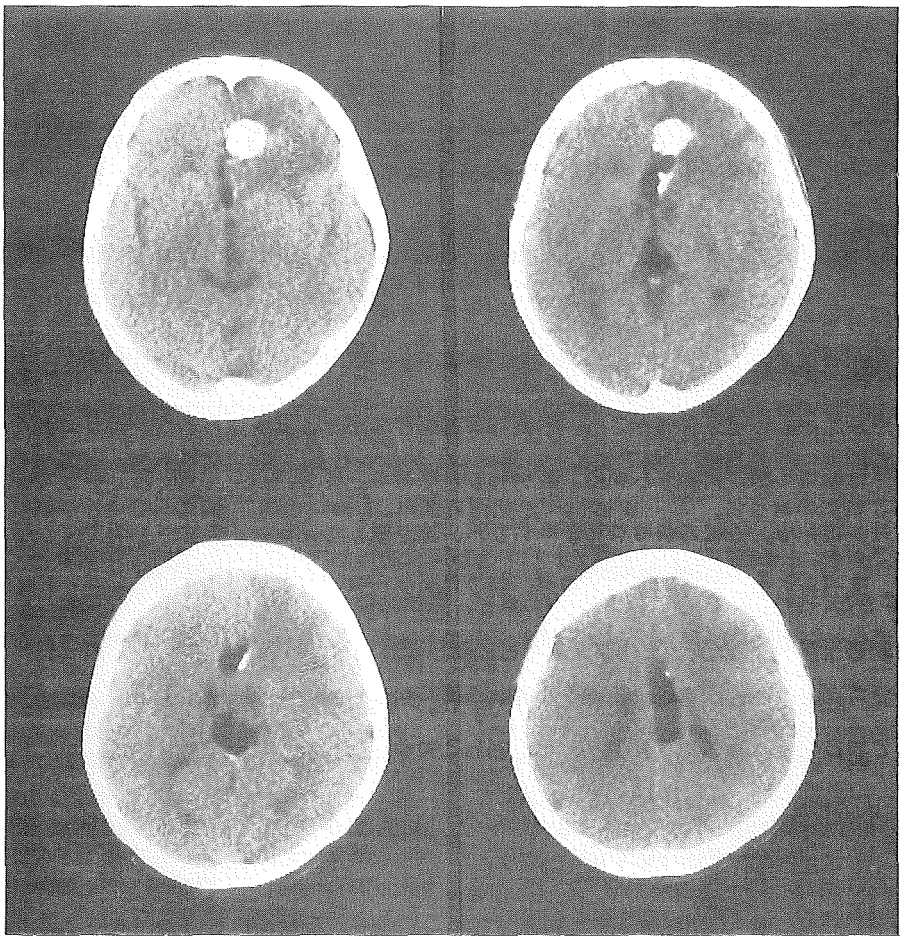

Fig. 2. Computerized tomography revealing a very low density area with a mean Hounsfield number of −71.3 in the area of corpus callosum located close to the calcification and a slightly low density area in the right frontal lobe.

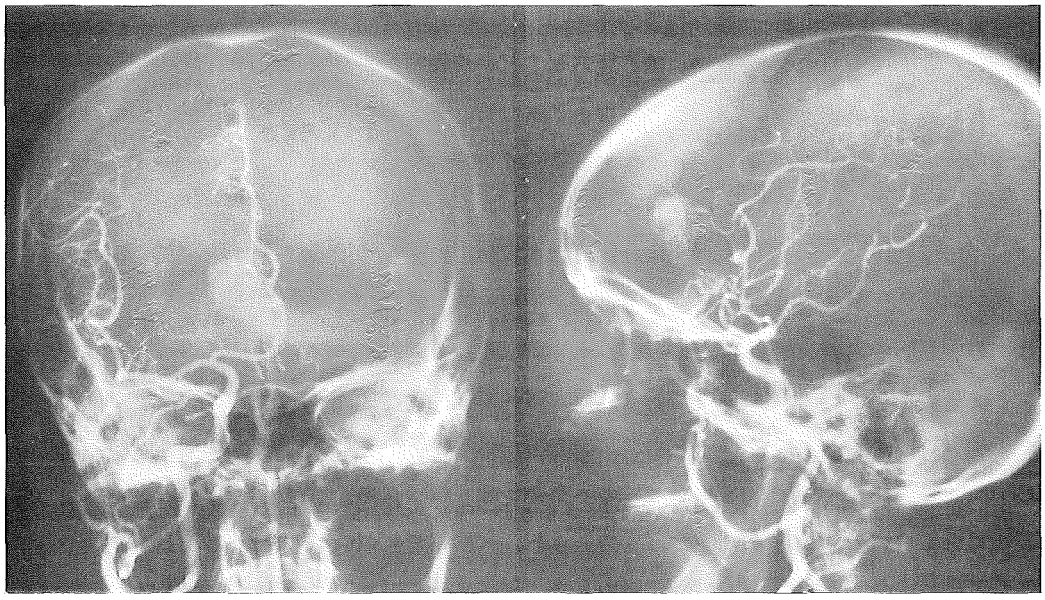

Fig. 3. Right carotid angiogram, anteroposterior (left) and lateral (right) views, revealing a distal anterior cerebral artery aneurysm and shift of A2 portion of anterior cerebral artery to the left and posteriorly.

phy revealed a very low density area with a mean Hounsfield number of −71.3 in the area of corpus callosum close to the calcification and a slightly low density area in the right frontal lobe (Fig. 2). Right carotid angiography revealed an aneurysm arising from the right distal anterior cerebral artery (Fig.

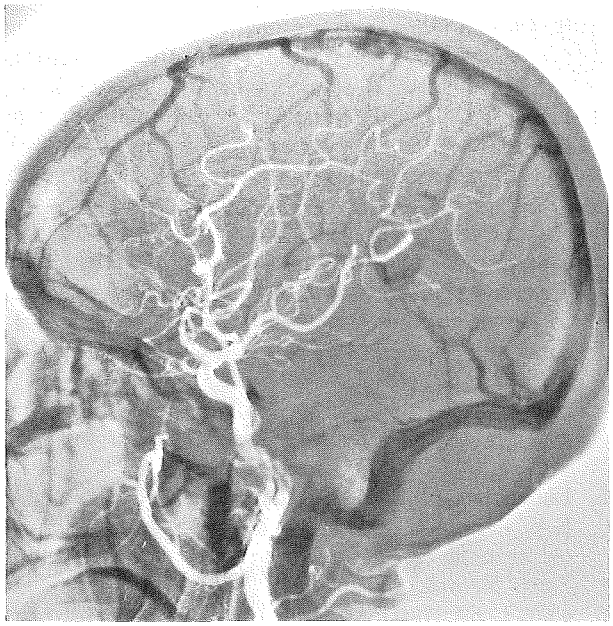

Fig. 4. Venous phase of the right carotid angiogram, lateral view, subtracted by arterial phase showing a normal distance between the internal cerebral vein and inferior sagittal sinus or pericallosal artery, indicating the structure of corpus callosum[14].

3). The A2 portion of the anterior cerebral artery shifted to the left and posteriorly (Fig. 3). Anomalies of the anterior cerebral artery or fusiform dilatation of the pericallosal artery were not seen. No abnormal vessels or tumor staining were presented. The distance between the internal cerebral vein and inferior sagittal sinus or pericallosal artery was not reduced indicating the structure of the corpus callosum (Fig. 4)[14]. EEG was within normal limits. Aneurysmal neck clipping was not performed because operation was refused.

DISCUSSION

Lipoma of the corpus callosum is a rare congenital condition. Only 100 cases have been reported[8] since Rokitansky[20] discovered it accidentally at autopsy in 1956. The patients were often asymptomatic[16,22]. The clinical features are not specific, but convulsions, headaches, mental retardation and hemiplegia have been observed[26]. Among these, convulsive seizures have been the most common manifestation[6,16]. CT scan gives the most characteristic and diagnostic findings of very low density area in the range of −50 to 100 Hounsfield units[12,18].

In our case, an episode of convulsive seizure was the only clinical symptom and the very low density area in the corpus callosum had a mean value of −71.3 Hounsfield units. The most probable diagnosis suggests lipoma of the corpus callosum. It is not known what is the low density area in the right frontal lobe not showing any increase in density with contrast medium in CT scan nor hypervascularity in angiography (Fig. 2 and 3). Possibilities suggest ischemia of the brain caused by compression of the brain with the calcification, fibrosis as described by Zettner[26] or a benign glioma.

Lipomas of the corpus callosum are often associated with various anomalies. The most frequent anomaly is agenesis of the corpus callosum[26]. Other associated anomalies include median cleft nose, cleft lip, spina bifida, myelomeningocele, funnel chest, cranial bone defect, cerebral hemiatrophy, agenesis of the septum pellucidum, agenesis of the cerebellar vermis, pituitary tumor, webbed toes and acoustic neurofibroma[16,22,26]. Although agenesis of the corpus callosum or other anomalies mentioned earlier were not observed in our case, aneurysm arising from the distal anterior cerebral artery (DACA) was accidentally found. Fusiform dilatation of the pericallosal artery[4,5] and anomalies of anterior cerebral arteries as classified into three types by Baptista[1] have been well documented[9,22,23] as angiographic findings of callosal lipoma, but a DACA saccular aneurysm as in this case has not been reported. The authors consider it to be of interest from the point of view of the pathogenesis of both callosal lipoma and DACA saccular aneurysm.

Lipoma of the corpus callosum is generally considered to be a congenital hamartomatous condition, because of the accumulation of cases in younger age[26] and a high coincidence with congenital anomaly[16,22,26]. It is speculated to develop earlier than the third or fourth fetal month when corpus callosum begins to appear[16,17]. On the other hand, anterior cerebral artery is formed at the second fetal month[19] which is close to the presumed time of appearance of the callosal lipoma.

Congenital etiology of DACA aneurysm is still poorly defined. Becker et al[2] are against a purely congenital etiology for DACA aneurysm because of the failure to detect it in the angiography of over 15,000 infants. However 15,000 infants is considered to be too small a number to deny congenital etiology at least for the DACA aneurysm. This is because of the very low incidence of DACA aneurysm which may be calculated to be 1-2% × 2-6% (general incidence of intracranial aneurysm × incidence of DACA aneurysm). About 300 cases of DACA aneurysms have been reported up to the present, the incidence being from 2 to 6% of intracranial aneurysms[2,13,25]. A higher incidence of DACA aneurysms is reported in patients with the azygos anterior cerebral artery. Huber et al have observed it in 7 out of 17 patients (41.1%)[10]. Some papers have emphasized hemodynamic factors for the pathogenesis of DACA aneurysm with the azygos anterior cerebral artery[11,13]. This incidence of DACA aneurysm is too high in patients with the azygos anterior cerebral artery in comparing only hemodynamic factors with those of basilar artery bifurcation which is under the same hemodynamic

condition. The multiplicity of DACA aneurysms is higher than that of other sites of intracranial aneurysms[2,13,24,25]. The incidence of DACA aneurysms in the saccular aneurysms of infancy and childhood is about 10%[3,15] which is higher than that of adult (2-6%). Verdura et al reported on familial DACA aneurysms[21]. Garcia-Chavez and Moossy reported on DACA aneurysm associated with agenesis of the corpus callosum[7]. These data and our case suggest that the congenital factor is more important in the pathogenesis of DACA aneurysm than in intracranial aneurysms of other sites. Callosal lipoma and DACA aneurysm are different in pathological origin but may be related genetically in this case.

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