

Thalamic syndrome caused by unruptured cerebral aneurysm

Case report

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✓ A case is reported of a 63-year-old woman with thalamic syndrome as the presenting feature of an unruptured cerebral aneurysm. Unruptured aneurysm is a rare cause of thalamic syndrome; the possible mechanisms of production of the sensory disturbance are discussed.

KEY WORDS • unruptured intracranial aneurysm • thalamic pain • hemifacial pain

SUBARACHNOID hemorrhage is the most common presentation of cerebral aneurysms, but unruptured cerebral aneurysms may present with cranial nerve palsy, visual failure, embolic phenomena, or they may be incidental. Giant aneurysms may cause neurological deficits due to mass effect, but this is uncommon with small lesions. Thalamic syndrome as a presentation of an unruptured aneurysm is a rare occurrence^{3,10} and has been attributed to ischemia from the compression of perforating vessels. We present a case of thalamic syndrome as the presenting feature of an unruptured aneurysm in which the evidence appears to favor local irritation of thalamic nuclei as the cause of the dysesthesia.

Case Report

This 63-year-old woman complained of mild pain and paresthesias in the right side of her face for many years. During the previous 6 months the sensory disturbance in her face had gradually worsened and had spread to involve her right arm and the entire right side of her body. This sensory disturbance was described as being an “abnormal feeling,” with constant dull pain and an unpleasant tingling sensation whenever the affected area was touched. The right side of the face was predominantly affected, but

the remainder of the right side of her head and the right side of her body were also involved. She described a very discrete midline limit to the symptoms. There were no other symptoms and her only medical history was hypertension, for which she was taking Captopril, and hypercholesterolemia, treated with Lopid.

Examination. Detailed physical examination failed to reveal any neurological deficit; in particular, there was no sensory deficit. A cranial contrast-enhanced computerized tomography (CT) scan revealed an enhancing lesion in the region of the bifurcation of the left internal carotid artery, extending up into the left thalamus (Fig. 1). Cerebral angiography confirmed this to be a pear-shaped aneurysm, 15 to 20 mm in length and 7 mm in diameter (Fig. 2). The bulbous fundus projected superiorly into the region of the left thalamus.

Operation. The aneurysm was clipped via a left frontotemporal approach. The lesion arose from the bifurcation of the left internal carotid artery and its large neck included several millimeters of the origins of both the anterior and middle cerebral arteries. Several perforating arteries were dissected off the deep aspect of the neck prior to placing a scimitar-shaped clip across the neck, reinforced by a 45°-angled ring clip. The aneurysm was



FIG. 1. Contrast-enhanced computerized tomography scans revealing a lesion in the region of the bifurcation of the left internal carotid artery. *Left:* The aneurysm is seen as an enhancing spherical lesion in the region of the bifurcation of the left internal carotid artery. *Center:* The fundus of the aneurysm extends into the left thalamus, just medial to the posterior limb of the left internal capsule. *Right:* Sagittal reconstruction demonstrating the aneurysm projecting up into the left thalamus.

then decompressed by puncturing the fundus; its exclusion from the circulation was confirmed. During exposure of the aneurysm, the oculomotor nerve was seen to be adherent to the medial temporal lobe; it was dissected off the temporal lobe to allow temporal lobe retraction.

Postoperative Course. The patient's recovery was uneventful apart from a temporary slight left ptosis. There was complete resolution of her sensory symptoms immediately following the operation. She continued to suffer some lethargy and depression that resolved within 12 months. She remained pain-free at the final 18 month postoperative follow-up evaluation.

Discussion

Although the mechanism of thalamic pain is not completely understood, Pagni¹¹ identified the following possibilities: direct irritation of sensory pathways and nuclei, loss of descending or ascending nociceptive inhibitory

mechanisms, incorrect integration of nociceptive impulses on remaining neurons or abnormal firing patterns of deafferented central nuclei, activation of the nonspecific (paleospinothalamic) system following selective damage to neospinothalamic fibers, or creation of abnormal reverberating circuits between thalamic nuclei. Although these mechanisms are not mutually exclusive, it is interesting to consider which of them may best explain this case. Pain is but one component of the classic thalamic syndrome described by Déjerine and Roussy in 1906,⁴ and the so-called "hemialgic" form of the syndrome,⁸ in which dysesthesia occurs without other sensory derangements, is rare.

The selective dysesthesia reported in this case might be an important discriminator of its mechanism. Within the thalamus, there is a spatial segregation of sharply circumscribed sensory inputs from the lemniscal and neospinothalamic systems on the one hand, and ill-defined dysesthetic inputs from the paleospinothalamic system on the other. The former relay chiefly on the ventroposterolateral and ventroposteromedial nuclei, the latter on the nonspecific or intralaminar nuclei.^{1,6,11} It is likely that the ventroposterolateral nucleus normally exerts an inhibitory influence over the nonspecific nuclei; surgical evidence in support of this concept has been cited by Miles.⁹ Thalamic pain is most commonly due to acute ischemic or hemorrhagic vascular lesions.¹¹ Given the complexity of the intrathalamic nuclear connections, it is possible that the final common mechanism for these lesions is selective destruction of normally inhibitory nuclei or pathways. This was the conclusion in Déjerine and Roussy's original case and in the more recent case of an intrathalamic arteriovenous malformation described by Waltz and Ehni.¹² Infiltrative primary thalamic tumors, on the other hand, infrequently give rise to pain.²

Aneurysms are a distinctly unusual cause of the thalamic syndrome.^{3,10} Oka, *et al.*,¹⁰ attribute the appearance of a localized contralateral dysesthesia in their patient to compression of a neighboring perforating thalamic vessel by an anterior cerebral artery aneurysm. However, their

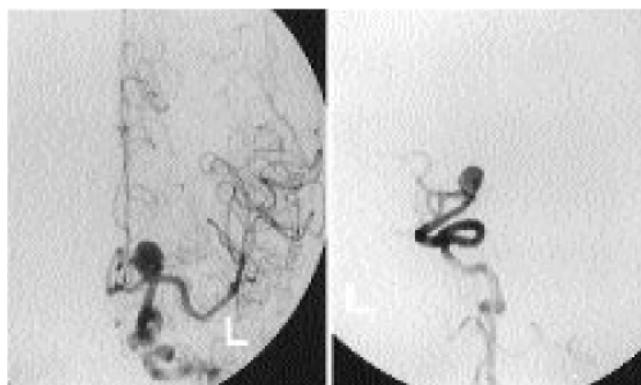


FIG. 2. Left carotid arteriograms demonstrating an anteroposterior (*left*) and lateral (*right*) bulbous aneurysm arising from the bifurcation of the left internal carotid artery. The origin of the aneurysm extended into the middle and anterior cerebral arteries.

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case is not truly comparable with the present one, because their patient also developed contralateral lower limb weakness.

It is difficult to see how an aneurysm could behave as a selectively deafferenting lesion at the level of the thalamus. Direct irritation of thalamic nuclei, presumably by local mechanical distortion, still appears promising as a potential mechanism of dysesthesia. Although direct stimulation of intralaminar nuclei in individuals can reproduce the dysesthetic experience,⁷ this has only been demonstrated in patients already affected by deafferentation pain. On the other hand, indirect evidence of an irritative mechanism operating at the thalamic level to produce an isolated dysesthesia is provided by rare cases of so-called "sensory epilepsy," in which pain may arise from global stimulation of the thalamus by cortical discharges.^{5,11} The prompt and lasting relief of dysesthesia after surgery in this case appears to support a simple explanation of this type.

References

1. Bowsher D: Pain: sensory mechanisms, in Vinken PJ, Bruyn GW, Klawans HL, et al (eds): **Handbook of Clinical Neurology**. Amsterdam: Elsevier, 1985, Vol 45, pp 227-244
2. Cambier J, Gravelleau P: Thalamic syndromes, in Vinken PJ, Bruyn GW, Klawans HL, et al (eds): **Handbook of Clinical Neurology**. Amsterdam: Elsevier, 1985, Vol 45, pp 87-98
3. Clinton GL, Fox MJ: Central pain syndrome: a review of the literature and a case report. **Wis Med J** 57:169-174, 1958
4. Déjerine J, Roussy G: La syndrome thalamique. **Rev Neurol** 12:521-532, 1906
5. Garcin R: La douleur dans les affections organiques du système nerveux central (partie anatomo-clinique). **Rev Neurol** 68: 105-153, 1937
6. Hirato M, Kawashima Y, Shibasaki T, et al: Pathophysiology of central (thalamic) pain: a possible role of the intralaminar nuclei in superficial pain. **Acta Neurochir** 52 (Suppl): 133-136, 1991
7. Levin G: Electrical stimulation of the globus pallidus and thalamus. **J Neurosurg** 24:415, 1966
8. Lhermitte J: Les syndromes thalamiques dissociés. Les formes analgiques et hémialgiques. **Ann Med Intern** 17:488-501, 1925
9. Miles JB: Surgical treatment of pain, in Miller JD (ed): **Northfield's Surgery of the Central Nervous System**. Edinburgh: Blackwell Scientific, 1987, pp 645-685
10. Oka H, Oki H, Ohbayashi M, et al: A case of unruptured A1 aneurysm causing ischemic compression to the medial proximal striate artery. **No Shinkei Geka** 14:1613-1617, 1986
11. Pagni CA: Central pain due to spinal cord and brainstem damage, in Wall PD, Melzack R (eds): **Textbook of Pain**. Edinburgh: Churchill Livingstone, 1989, pp 634-655
12. Waltz TA, Ehni G: The thalamic syndrome and its mechanism. Report of two cases, one due to arteriovenous malformations in the thalamus. **J Neurosurg** 24:735-742, 1966

Manuscript received May 9, 1994.

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