Epidural angiolipoma is histologically distinct from its cutaneous counterpart in the calibre and density of its vascular component; a case report with review of the literature

H Hattori

Angiolipoma of the spine is a rare tumour and no studies have investigated a sufficient number of cases to reach general conclusions. Therefore, as yet, the pathological definition of this entity is not well established. The case of epidural angiolipoma reported here and a review of the literature revealed that this entity is distinct from cutaneous angiolipoma in that the calibre of its predominant vascular component is far greater than that of the fat cells. Therefore, epidural angiolipoma can be regarded as not only topographically, but also histologically, distinct from its subcutaneous counterpart.

The dura of the spine is composed of an outer and inner lobe, between which lies the epidural space, which contains fatty tissue and blood vessels. Several types of mesenchymal tumour can arise in this space, with most of them naturally being lipomas or haemangiomas. The peculiar and not yet well characterised entity loosely designated as angiolipoma can also arise in this space. This report describes such a tumour, the pathological aspects of which have not fully been described previously.

CASE
A 71 year old woman had been followed because of lumbago, the symptoms of which had recently become worse, with intermittent lameness. Computed tomography and magnetic resonance imaging revealed an extradural tumour located on the posterior aspect of L 3/4, compressing the cauda equina. The tumour was hyperintense both in T1 weighted and T2 weighted magnetic resonance images, with gadolinium enhanced cord-like T1 hypointense areas scattered. The resected tumour measured 25 x 40 x 12 mm. The cut surface was yellowish in colour, in places haemorrhagic, firm, and spongy (fig 1). Microscopically, the tumour was composed of mature adipose tissue and abundant vascular channels, the calibre of which was variable, ranging from nearly capillary sized to cavernous, but mostly several times larger than the fat cells (fig 2). There was no cytological atypia or structural abnormalities in the vascular channels. Lipoblasts were not seen.

DISCUSSION
Angiolipoma of the skin is a relatively common tumour that has been well characterised. It consists of mature adipose tissue and vascular proliferations, which by definition are of capillary size. Angiolipoma of the spine is a rare tumour. No studies have investigated a sufficient number of cases to reach general conclusions, so that the pathological definition of this entity is not well established yet. For example, in the second edition of the World Health Organisation monograph it is classified as vascular lipoma, whereas in the latest series it has been stated, in accordance with its cutaneous counterpart, that the vessels in angiolipoma should be by definition of capillary type.

Our case of epidural angiolipoma was distinct from cutaneous angiolipoma in that its predominant vascular component was not of capillary size. In addition, it contained too many vessels to be regarded as an extremely vascular lipoma. Furthermore, it could not be regarded as a haemangioma that had arisen in the fatty tissue because of the close mixture of both the elements. The vascular proliferation seen in our case is beyond the definition of cutaneous angiolipoma, and has never been mentioned as a variant feature of subcutaneous angiolipoma.

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By searching MEDLINE or the database of medical literature in Japanese (Igaku chuo zasshi) it was possible to find case reports containing photomicrographs so that the calibre and density of vessels could be compared with the fat cells. Twenty of the available 32 English literature cases and 17 of the 24 Japanese cases were possibly similar to our present case, with eight of the 20 English cases3–10 and 12 of the 17 Japanese cases clearly similar to our case. The remaining cases were regarded as lipoma or haemangioma, or the figures did not enable the predominant histological feature to be ascertained. There were no microphotographs...
where the tumour was clearly similar in histology to cutaneous angiolipoma.

Thus, epidural angiolipoma should be regarded as not only topographically, but also histologically, distinct from its subcutaneous counterpart. Cutaneous angiolipoma is reported to carry no chromosomal abnormalities, in contrast to other lipomatous tumours.11 This fact and the familial occurrence of angiolipomatosis suggests that cutaneous angiolipoma is a hamartomatous lesion rather than a tumour.11 Such a combination of fatty and angiomatous proliferation is extremely rare in organs other than the epidural space and subcutis. Therefore, there may be certain histogenetic links between cutaneous and extradural angiolipoma, although the clinical features and the histological appearance of the two diseases are different. This hypothesis is supported by cases of familial angiolipomatosis with Lisch nodules12 and Proteus syndrome with epidural angiolipoma.13 Cytogenetic analysis of extradural angiolipoma might help solve the problem.

**Take home messages**

- Angiolipoma of the spine is a rare tumour that has not been well characterised.
- Our case of epidural angiolipoma and a review of the literature revealed that this entity is distinct from cutaneous angiolipoma in that the calibre of its predominant vascular component is far larger than that of the fat cells.
- Epidural angiolipoma appears to be topographically and histologically distinct from its subcutaneous counterpart.

Correspondence to: Dr H Hattori, Department of Pathology, Kariya General Hospital, Sumiyoshicho 5-15 Kariya, Aichi 448-0852, Japan; hhattorih1@yahoo.co.jp

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