Lateral sellar angiolipoma: a tumor illustrative of the extradural compartment of the neural axis

Case report

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Angiolipomas are rare tumors of the CNS that most frequently develop in the orbit, the cavernous space, and the epidural space of the spine. The authors report the case of a patient who presented with an angiolipoma of the cavernous space. Using data from the published literature and an experimental anatomical approach, they demonstrate that the cavernous space contains adipose tissue. Consequently, they suggest that angiolipomas constitute a characteristic tumor illustrating the interperiosteal-dural concept.

The authors report the clinical, radiological, and histological data of a patient who presented with a tumor of the cavernous space. In addition, they prepared 2 encephalic extremities (4 cavernous spaces) using a special anatomical preparation consisting of an injection of colored neoprene latex followed by a 6-month immersion in a formaldehyde solution enriched with hydrogen peroxide to soften the bone structures (coronal sections) while leaving the fat in the cavernous space intact.

This case report corroborates previously published clinical data and shows that the tumor was a hamartoma comprising mature fat cells associated with vascular proliferation. The tumor developed in the cavernous space, which is an interperiosteal-dural space extending from the sphenoid periosteum (osteoperiosteal layer) to the superior and lateral walls of the cavernous space (encephalic layer). This space represents an anatomical continuum extending from the coccyx to the orbit: the interperiosteal-dural concept. It contains fat tissue that is abundant at the level of the orbit and the epidural spinal space and sparser at the level of the cavernous spaces, as was shown in our anatomical study.

The authors suggest that angiolipomas represent a characteristic tumor that illustrates the interperiosteal-dural concept because they essentially develop in the fat tissue contained in these spaces. (DOI: 10.3171/2010.1.JNS091031)

KEY WORDS • anatomy • angiolipoma • cavernous sinus • fat • skull base surgery

Angiolipomas of the CNS are rare, benign tumors originating in mesenchymal tissue and are constituted by a proliferation of mature fat cells and endothelial cells.1,17,23,33 They most frequently develop in the epidural space of the spine, the cavernous spaces, and more rarely in the orbit.2,5,8,10,12,13,15,16,18,22,24,26,28,31,35–37 Few publications have reported them outside of these locations, in particular at the level of the tectal lamina and the thalamus.21,25 Angiolipomas represent an anatominopathological entity distinct from lipomas, and their anatominopathological substratum and localizations are different (they mainly develop along the midline: the corpus callosum, the quadrigeminal plate, and the interhemispheric fissure).6,32,38 This new case of angiolipoma of the LSC and an extensive review of the literature (Table 1) prompted us to use a novel anatomical technique and apply it to the LSC and its contents. This technique allowed us to soften the bony structures without altering the fat tissue and thus determine whether fat tissue was present in the LSC. The operative findings in this case combined with the anatomical study we performed and a critical review of the published literature lead us to suggest that angiolipomas are a pathological illustration of the interperiosteal dural concept.

Case Report

History and Examination. This 56-year-old woman with no significant medical history presented with headache and partial ophthalmoplegia that had been present for 2 months. Neurological examination revealed an incomplete extrinsic paralysis of CN III with ptosis and diplopia. Facial sensitivity was normal and visual acuity was 6/10° on the right and 10/10° on the left.
Magnetic resonance imaging showed a well-defined tumor measuring 35 × 40 × 35 mm in the left cavernous space with extension into the middle cerebral fossa and hypophysial space. The tumor was hyperintense on T2 FLAIR-weighted MR imaging sequences and strongly enhanced after Gd administration (Fig. 1A and B). The T1-weighted fat saturated MR imaging sequences showed a clear-cut hypointense signal in comparison with the adjacent cerebral tissue, thereby confirming that the tumor consisted of fat tissue (Fig. 1C).

**Operation.** We removed the tumor through a left pterional approach after preparing the patient for a cerebrospinal fluid leak that was colored blue during our anatomical preparation. We dissected 2 cadaveric heads (4 LSCs) after injecting them with neoprene latex, colored red for the arteries following catheterization of the common carotid and vertebral arteries of the neck and colored blue for the veins following catheterization of the internal jugular veins. They were then fixed by immersion for 6 months in a 10% formaldehyde solution containing 5% hydrogen peroxide with regular renewal of the immersion baths. Using this technique, we were able to soften the bony structures without affecting the fat tissue. After the 6-month immersion treatment, we extracted the brains and performed coronal sections of the cavernous spaces using an operating microscope (Zeiss OPMI 9FC). We took photographs with a Hasselblad camera (Hasselblad AB) and developed them using Kodak film (Ektachrome 160T). We obtained the latex (#671) from E.I. Du Pont de Nemours-Dow Elastomers.

The results of the 2 anatomical preparations (4 cavernous spaces) were identical. The cavernous space consisted of an interperiosteal dural space located between the encephalic layer of the dura mater and the osteoperiosteal layer. The encephalic layer formed the lateral wall, the roof, and the superomedial wall of the cavernous space and was contiguous with the encephalic layer of the anterior and middle floor of the skull base. The osteoperiosteal layer adhered to the bone and formed the inferomedial wall of the cavernous space. The cavernous space was filled with islets of fat tissue located lateral to the intracavernous carotid artery and medial to the lateral wall of the cavernous space (Fig. 2). The fat islets were visible between the neurovascular elements that coursed through the cavernous space and were more developed at the level of the anterosuperior compartment of the cavernous space. The cavernous space also contained venous lakes that were colored blue during our anatomical preparation.

**Discussion**

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are different from lipomas. They most frequently appear in the thoracic spine where they are called extradural angiolipomas since they develop between the encephalic layer of the dura mater that forms the dural sheath of the spinal cord and the osteoperiosteal layer that covers the vertebrae. This interperiosteo-dural space is improperly called the epidural space. It contains an important quantity of fat tissue that facilitates the movements of the dural sheath of the spine (spinal prolongation of the encephalic layer) in the spinal canal, which is covered by the osteoperiosteal layer. Angiolipomas develop from this fat tissue. They have also been reported to develop in the cavernous space and the orbit.

The epidural spinal space, the cavernous space, and the orbit are areas in which the interperiosteo-dural space is larger and filled with fat tissue and venous lakes. The interperiosteo-dural concept in the cavernous space was first described by Taptas. He individualized the encephalic and osteoperiosteal layers of the dura mater after closely examining the walls of the cavernous space. Thus, the walls of the cavernous space are formed by the apposition of 2 encephalic layers: an external layer that is continuous with the middle cerebral fossa layer and an internal layer formed by the apposition of the dural sheaths of the oculomotor, ophthalmic, and trochlear nerves as can be seen in our histological sections. The

Fig. 1. A: Axial T2-weighted FLAIR MR image of a left angiolipoma located in the LSC with intrasellar extension. B: Axial T1-weighted MR image obtained after Gd injection. The tumor appears homogeneously hyperintense. The anterior bend of the carotid artery is totally encased in the lateral sellar compartment (arrow). C: Fat saturation MR image showing that the angiolipoma is isointense to gray matter, which is typical of tumors with a fat component. D: Photomicrograph showing typical angiolipoma patterns with mature adipose cells interspersed with thin-walled vascular channels. The tumor is covered by the encephalic layer of the lateral wall of the cavernous sinus (asterisk). H & E, original magnification x 10.
The medial wall is formed cephalad by the hypophysial dural bag (that is, a meningeal bag suspended from the sellar diaphragm), which has been described by Destrieux et al. Caudally, the medial wall is formed by the osteoporoosteal layer that covers the body of the sphenoid. The roof of the cavernous space is formed by the encephalic layer covering the anterior clinoid process. The roof is perforated by the oculomotor and trochlear nerves, carrying the encephalic layer with the nerves and composing the dural sheaths of the encephalic layer. These entry points in the dura constitute areas of least resistance for the extension of tumors in the cavernous space toward the subarachnoid spaces. Indeed, some angiolipomas described in the suprasellar region are in fact tumors of the cavernous space that have extended to the suprasellar region by passing through these openings. Some anatomical data have been confirmed by data obtained with medical imagery, and they have frequently demonstrated the presence of fat tissue in the cavernous space.

As we have shown in our anatomical dissections (Fig. 2), the cavernous space contains venous spaces that communicate with the orbital veins, the superior petrosal sinus, and the petroclival venous confluence. Parkinson has extended this concept of the dura.

**Fig. 2.** A: Superior view of the sellar region with coronal sections (B, C, and D) passing through the left LSC. The LSC is an interperiosteal-dural space limited laterally by a meningeal layer. The medial wall is composed of 2 parts: the sphenoidal part, which corresponds to the endosteal layer, and the hypophysal part, which corresponds to the encephalic (meningeal) layer. B: Anterior coronal section (B in panel A) passing through the anterior bend of the carotid artery. C: Middle coronal section of the cavernous sinus (C in panel A). The adipose tissue (yellow area, indicated by arrowheads) occupies a large island filling most of the space between the arteries, nerves, veins, and dura layers. D: Posterior coronal section (D in panel A) passing through the posterior bend of the carotid artery. A. = artery; Ant. Clin. = anterior clinoid; EL = endosteal layer; ML = meningeal layer; Pit. = pituitary.
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mater of the cavernous space to include an anatomical concept: the interperiosteo-dural concept that constitutes a veritable anatomical continuum extending from the orbit to the coccyx. This concept is illustrated in Fig. 3 and summarizes the distribution of the encephalic and osteoperiosteal layers. In the orbit, the encephalic layer contains the dural sheath of the optic nerve, which is an invagination of the encephalic layer of the anterior floor at the level of the falciform ligament. The osteoperiosteal layer is represented by the periorbit and adheres to the orbit. Caudally, at the foramen magnum, the osteoperiosteal layer is represented by the vertebral periosteum while the encephalic layer forms the dural sheath. At this point, the interperiosteal dural space is wide, just as it is at the level of the orbit and the cavernous space; this corresponds to the epidural spinal space. When the 2 layers are separated from one another, it is apparent that this space contains fat tissue and venous lakes, a primary condition for the interperiosteo-dural concept. Recently, we demonstrated the validity of this concept by using electron microscopy to examine these layers. Indeed, the encephalic layer we observed is formed by a linkage of collagen fibers covered by an amorphous substance. In fact, the encephalic layer comprises a deep cellular layer (dural border cell) and a superficial layer. An amorphous, less resistant substance extends between them. Under these conditions, removal of the encephalic layer exposes the amorphous substance and consequently, only the superficial layer of the encephalic layer is actually removed. The deeper, cellular layer remains adherent to the arachnoid. This aspect is not observed at the level of the osteoperiosteal layer. These morphological findings corroborate the anatomical data pertaining to the distribution of the layers of the dura mater. Thus, angiolipomas electively develop between the 2 layers of the dura mater, in the interperiosteo-dural space, and originate in the fat tissue that is present.

Considering all of these epidemiological, histological, radiological, and anatomical findings, we suggest that angiolipomas are characteristic tumors that illustrate the interperiosteal dural concept.

Disclosure

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Fig. 3. Diagram representing a paramedian section of the neural axis showing the distribution of the encephalic and osteoperiosteal layers of the dura mater. These 2 layers are separated from one another at the level of the orbit, the cavernous space, and the epidural spinal space and delimit the interperiosteal-dural space, a veritable anatomical continuum extending from the coccyx to the orbit. The 2 layers are separated from one another by fat tissue and venous lakes.
interpretation of data: P François. Drafting the article: P François. Critically revising the article: I Zemmoura, AM Bergeron Fouquet, M Jan, S Velut. Reviewed final version of the manuscript and approved it for submission: S Velut.

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