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SPINAL EPIDURAL ANGIOLIPOMA: A LITERATURE REVIEW

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Abstract: Spinal angiolipomas are rare benign tumors composed of mature adipocytes mixed with abnormal vessels. They represent 0.0004 to 1.2% of angiolipomas, usually located at the extradural and posterior thoracic level, with multimetameric extension. Spinal angiolipoma is an uncommon form of benign tumor. There are 2 types: non-infiltrating and infiltrating. Its clinical course is slow and progressive, and it can be accelerated by vascular phenomena, intratumoral abscess and pregnancy. Intratumoral bleeding must be considered a cause of acute spinal compression syndrome. Spinal epidural angiolipoma is a rare, slow-growing and progressive benign tumor. MRI is the gold standard for diagnosis. The gold standard treatment must always be surgery, although total resection may not be possible in some cases.
Keywords: Angiolipoma; Spinal cord compression; Spinal cord neoplasms; Paraplegia

INTRODUCTION

Spinal angiolipomas are rare benign tumors. They represent 0.0004 to 1.2% of angiolipomas, usually located at the extradural and posterior thoracic level, with multimetameric extension (UMANA et al 2022). They constitute about 0.04-1.2% of all spinal axis tumors and about 2-3% of all extradural spinal tumors (DRAPKIN et al. 1974; El KHAMLICHI et al. 1989). Clinical symptoms are usually compressive myelopathy and radiculopathy. Most spinal angiolipomas are located at the thoracic level and arise in the posterior extradural space. Pure lumbar localization is rare (EPSTEIN et al. 1989). Surgical resection of tumors is the treatment of choice for spinal angiolipomas, especially when there is myelopathy (ROCCHI et al. 2004).

Angiolipoma is a benign neoplasm that consists of mature adipose tissue and abnormal

vascular structures and affects middle-aged female patients (between 30 and 40 years old), usually located in the mid-thoracic region (AKHADDAR et al. 2008). The location in the lumbar spine is not very common (AKHADDAR et al. 2008; GELABERT-GONZÁLEZ et al. 2009). It usually follows a slow and progressive clinical course, and acute myelopathy is very rare. (MENG et al 2013). There are only 142 cases of extradural spinal angiolipoma reported to date (FUJIWARA et al. 2013). They account for between 0.14% and 1.2% of all spinal cord tumors and 2% to 3% of spinal epidural tumors. They can be classified into two subtypes: non-infiltrating and infiltrating. The treatment of epidural spinal angiolipomas is by total surgical removal (FUJIWARA et al. 2013; LEU et al. 2003).

MATERIAL AND METHODS

A review of the current literature was carried out. The following databases were consulted: MEDLINE (PubMed); base; Web of Science, Google Scholar. Conference abstracts/articles have been deleted from Embase. No other limits have been applied. All retrieved records were organized using Endnote citation management software version 20. For the removal of duplicates, a review software and literature review citation screening software was used.

The search strategy was designed to capture case studies of spinal epidural angiolipoma. The searches were complemented by manual searching and retrieval of any additional articles that met the eligibility criteria.

RESULTS

The article by Berenbruch et al. described the first case of spinal angiolipoma in 1890 in a 16-year-old boy with numerous cutaneous lipomas, while the first pathological report was published in 1960 by Howard and Helwigem who established angiolipoma as a defined clinicopathological entity composed of variable proportions of cells. mature adipose tissue and capillaries, sinusoids, and abnormal venous or arterial elements (FUJIWARA et al. 2013; (HOWARD et al. 1960; BERENBRUCH et al. 1890). Subsequently, angiolipomas were subdivided into two categories by Lin et al., non-infiltrating and infiltrating. The non-infiltrating type is encapsulated and well delimited, without invasion of the dura of the vertebrae. The infiltrative type is more common and usually emerges from the posterior epidural space. Infiltrating spinal angiolipomas are found in the anterior epidural space and may invade the body vertebral column (FUJIWARA et al. 2013; GELABERT-GONZÁLEZ et al. 2009; LIN et al. 1974).

Histologically, the lesion is mainly composed of mature fat cells and blood vessels. The composition of fat is similar to that of adipose tissue, and the vascular components usually consist of capillaries, thin or thick-walled sinusoids with smooth muscle, and occasionally small, well-developed arteries. A diagnostic feature is the presence of fibrin thrombi in the capillary lumen. Degenerative changes (ie, myxoid changes, hyalinization, and fibrosis) may be present in some cases with a long course (FUJIWARA et al. 2013).

CLINICAL PRESENTATION

They occur mainly in women (women: men=22:17) and more frequently during the 4th and 5th decades of life. The presentation may be acute, subacute, or chronic and may include progressive, remitting, or recurrent radicular and paraplegic signs and symptoms. The most common initial symptoms are muster pain, numbness of the lower extremities or paresthesia and weakness in the legs. But the progression can be accelerated through vascular phenomena such as vascular engorgement, venous stasis,

intratumoral hemorrhage and, in rare cases, intratumoral abscess. Bleeding is very rare in angiolipomas (FUJIWARA et al. 2013). Akhaddar et al. described a case in which a spontaneous hemorrhage caused an acute paraplegia similar to the clinical presentation of our patient. As with other vascular lesions, the onset or deterioration of neurological symptoms may occur during pregnancy or with weight gain (AKHADDAR et al. 2008; FUJIWARA et al. 2013; LEU et al. 2003).

Pre- and postoperative clinical assessments are made using the Japanese Orthopedic Association (JOA) scale for thoracic myelopathy (MENG et al 2013)

DIAGNOSTIC EVALUATION

Spinal radiography is normal, but in some cases it shows erosion of the pedicle, increased diameter of the spinal canal and trabeculation of the vertebral body affected by the infiltration of tumors (LEU et al. 2003).

Computed tomography (CT) usually shows a hypodense lesion with fat density, provides information on the degree of bone involvement, and can also show varying degrees of image enhancement after contrast injection (FUJIWARA et al. 2013).

Magnetic resonance imaging (MRI) is considered the gold standard for the diagnosis of spinal angiolipoma, which is typically isointense on T1 and hyperintense on T2 (MENG et al 2013; PROVNZALE et al. 1996; GHANTA et al. 2012). Intravenous injection of gadolinium contrast enhances the vascularity of these tumors.3T2-weighted images with fat suppression techniques can be very useful in distinguishing between angiolipomas and melanomas or subacute hemorrhage (FUJIWARA et al. 2013; PROVNZALE et al. 1996; GHANTA et al. 2012).

Angiolipomas can be detected by fluorodeoxyglucose (FDG) - positron emission tomography (PET). Spinal angiography is advantageous for the differential diagnosis and evaluation of the tumor's supplying vessels. Differential diagnosis includes lipoma, hemangioma, malignant lymphoma, and nerve sheath tumors (MENG et al 2013).

TREATMENT

There is no clear consensus as to the best therapy for the treatment of spinal angiolipoma. The biological behaviors of non-infiltrating and infiltrating angiolipomas require different focuses of treatment. The primary treatment is total surgical resection. Most non-infiltrating extradural tumors are eligible for total surgical resection via laminectomy. The posterior location of a spinal cord tumor facilitates resection by bilateral laminectomy (LEU et al. 2003). For total resections of non-infiltrating angiolipomas that compromise the vertebral body, an anterolateral approach and stabilization of the affected vertebrae are recommended. Although total resection of the lesion is not always easy, recurrence is rare. In cases of recurrence, extensive surgical resection followed by radiotherapy must be considered. Most patients have a good prognosis because the tumors are usually slow growing and do not metastasize (HOWARD et al. 1960).

DISCUSSION

Spinal angiolipomas are rare benign tumors composed of mature adipose tissue and abnormal vascular elements. They represent 2-3% of spinal extradural tumors and 24% of spinal lipomas (DRAPKIN et al.1974; GELABERT-GONZÁLEZ et al. 2009). Two subtypes of spinal angiolipoma were proposed by Lin et al. in 1974. In most cases, spinal angiolipoma is encapsulated and not infiltrative, with a benign prognosis. Unencapsulated spinal angiolipoma is less common, with a poor prognosis. Spinal

angiolipomas are more common among women, with a male-to-female ratio of 1:1.9 (YEN et al. 2008). Spinal angiolipomas usually present in the fourth or fifth decade of life, suggesting a hormonal influence on the development or progression of spinal angiolipomas GELABERT-GONZÁLEZ et al. 2009). They most commonly occur in the thoracic spine and are predominantly found in the extradural space (El KHAMLICHI et al. 1989). Pure lumbar localization is extremely rare. Most non-infiltrating spinal angiolipmas are predominantly found in the extradural space and are located posteriorly or posterolaterally (HUNGS et al. 2008).

Compressive myelopathy and radiculopathy related to the location of tumors are the main presentations of spinal epidural angiolipomas. In 40.6% of patients, the clinical presentation is progressive or sudden weakness of the extremities.

(DRAPKIN et al.1974). Other clinical presentations include back pain, progressive sensory dysfunction of the lower extremities, hyperreflexia, spasticity, and sphincter dysfunction. Most symptoms progress slowly over months and the diagnosis is typically established in less than 1 year GELABERT-GONZÁLEZ et al. 2009). Vascular factors such as gradual enlargement of anomalous vessels, intralesional thrombosis, hemorrhage or stealing phenomena have been reported to contribute to a more rapid development of symptoms in some cases (Epstein et al. 1989; HUNGS et al. 2008; FOURNEY et al. 2001;). Pregnancy and obesity have also been reported to be related to a course of rapid deterioration (FUJIWARA et al. 2013).

Spinal MRI is the most valuable radiological test for diagnosing spinal angiolipomas. As spinal angiolipomas are composed of both adipose and vascular components, your spinal MRI shows the characteristics of these two tissues. Spinal angiolipomas are hyperintense areas on non-contrast T1-weighted images that reflect the degree of vascularity. The presence of large hypointense regions on T1-weighted images is correlated with high vascular content.

Macroscopic total surgical resection of tumors is the treatment of choice for spinal epidural angiolipomas. Most patients present good postoperative neurological improvement (DRAPKIN et al. 1974; PIERRE-KAHN et al. 1997; SCHERPEREEL et al. 1984). Postoperative recurrences of spinal angiolipomas are rare, especially when macroscopic total resection of the tumors is achieved (DRAPKIN et al.1974). Adjuvant radiotherapy is not indicated after the operation. Both of our patients recovered well after total tumor resections and remained tumor-free one year after the operation.

Spinal angiolipomas are rare but well described spinal tumors and must be listed in the differential diagnosis in patients who have both compressive myelopathy and radiculopathy. MRI of the spine can provide information about its vascularity. The more heterogeneous intensity on T1- and T2-weighted images may suggest greater vascularization of the tumors and the possibility of more intraoperative bleeding. Early diagnosis and total resection of spinal angiolipomas are key points for a good therapeutic outcome.

FINAL CONSIDERATIONS

Spinal epidural angiolipoma is a rare, slow-growing and progressive benign tumor. MRI is the gold standard for diagnosis. Therefore, the diagnosis of spinal angiolipomas can initially be a radiological challenge, since it can mimic other spinal injuries.

The gold standard treatment modality must always be surgery, although total resection may not be possible in some cases. The treatment of this pathology is done by

releasing and resection of the tumor, and it has a favorable prognosis, even when the surgery is delayed, as in the case reported. Intratumoral bleeding must be considered as a result of a spinal compression syndrome, as was the case with our patient.

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