CASE REPORT

Spinal Angiolipoma

Daryl R. Fourney, Karen A. Tong, Robert J.B. Macaulay and Robert W. Griebel

ABSTRACT: ***Background:*** Spinal epidural angiolipoma is a rare cause of spinal cord compression. We present a case and review the clinical presentation, radiological appearance, pathological aspects and treatment of this distinct clinico-pathological entity. ***Methods:*** A case of a 46-year-old woman with a five-month history of progressive myelopathy affecting her lower extremities is presented. CT and MRI revealed a large epidural fat-containing mass compressing the spinal cord dorsally at the T7-T8 level. A laminectomy was performed with gross total resection of the lesion. ***Results***: The patient's neurologic symptoms improved postoperatively. A two-year follow-up period has revealed no signs of tumor recurrence and no neurological deficit. ***Conclusion:*** The diagnosis of spinal angiolipoma should be considered in the differential diagnosis of spinal cord compression. Magnetic resonance imaging is the investigation of choice. The surgical objective is complete excision but, for anterior lesions involving bone, an overly aggressive approach should be tempered by an awareness of the overall indolent natural history of so-called "infiltrating" spinal angiolipomas that are only partially excised.

RÉSUMÉ: Angiolipome spinal. ***Introduction:*** L'angiolipome épidural spinal est une cause rare de compression de la moelle épinière. Nous présentons un cas et nous revoyons la présentation clinique, les aspects radiologiques et anatomopathologiques ainsi que le traitement de cette entité clinico-pathologique distincte. ***Méthodes:*** Nous décrivons le cas d'une femme de 46 ans ayant une histoire de myélopathie progressive affectant ses membres inférieurs, évoluant depuis 5 mois. La tomodensitométrie et la résonance magnétique nucléaire ont montré une grosse masse épidurale contenant de la graisse et comprimant la moelle épinière au niveau de D7-D8. Une laminectomie a été effectuée avec exérèse de la lésion. ***Résultats:*** Les symptômes neurologiques de la patiente se sont améliorés après la chirurgie. Deux ans après, il n'y a aucun signe de récidive de la tumeur et pas de déficit neurologique. ***Conclusion:*** Le diagnostic d'angiolipome devrait faire partie du diagnostic différentiel dans la compression de la moelle épinière. L'imagerie par résonance magnétique est l'investigation de choix. L'objectif chirurgical est l'excision totale de la tumeur mais, quand il s'agit de lésions antérieures impliquant les structures osseuses, une approche agressive devrait être tempérée par la notion que les angiolipomes spinaux soit disant infiltrants ont une histoire naturelle plutôt indolente s'ils ne sont pas réséqués complètement.

Can. J. Neurol. Sci. 2001; 28: 82-88

Spinal angiolipomas are benign tumors composed of both mature adipose and abnormal vascular elements that represent a distinct clinical and pathological entity.[1,2] They are very rare, estimated to account for between 0.04% and 1.2% of all spinal axis tumors and two to three percent of extradural spinal tumors.[1,3,4] The first case, clearly identifiable as a spinal angiolipoma was described by Liebscher[5] in 1901, although as early as 1890, Berenbruch[6] described a case of multiple angiolipomatosis which included an extradural spinal mass causing back pain and lower extremity weakness. There have been 75 cases reported to date (Tables 1-3). Another case of spinal epidural angiolipoma at our institution was the subject of a previous report.[24]

Spinal angiolipomas may be subdivided into two types. The commonest form is usually confined to the posterior epidural space. The other less common variety of spinal angiolipomas is termed "infiltrating" in the literature since it invades bone.[55] These tumors typically reside anterior or anterolateral to the spinal cord. Both types have an excellent prognosis following surgical removal. Five cases of intramedullary angiolipomas have also been reported.

Case Report

A 46-year-old Caucasian woman had previously been in good general health with the exception of a four-year history of fibromyalgia. She originally presented with numbness in her feet and legs and was given a course of intravenous pulse steroids by a neurologist for probable multiple sclerosis. The numbness, however, progressed proximally over the next five months and she subsequently developed difficulty walking and bladder incontinence.

From the Division of Neurosurgery (DRF, RWG), Department of Radiology (KAT), and Department of Pathology (RJBM), Royal University Hospital, Saskatoon, Saskatchewan, Canada.

Received May 13, 2000. Accepted in final form September 13, 2000.

Reprint requests to: Robert W. Griebel, Division of Neurosurgery, Royal University Hospital, 103 Hospital Drive, Saskatoon, Saskatchewan, Canada S7N 0W8

Table 1: Spinal Epidural Angiolipomas Without Bone Involvement

Author (year)	Sex	Age (years)	Site	Duration of Symptoms	Treatment	Result
Berenbruch (1890)[6]	M	16	Thoracic	Unknown	Surgery	Necropsy
Liebscher (1901)[5]	F	56	T7-8	4.5 years	No surgery	Necropsy
Frazier & Allen (1918)[7]	F	55	Thoracic	Unknown	Unknown	Unknown
Elsberg (1928)[8]	F	33	T8-9	Unknown	Unknown	Unknown
Kasper and Cowan (1929)[9]	M	6	C2-8 & L3-S3	1 week	No surgery	Necropsy
Petit-Dutaillis & Cristoph (1931)[10]	F	44	T6-10	1 year	Surgery	Recovery
Ehni & Love (1934)[1]	F	33	T6-T8	2.5 years	Surgery	Recovery
	M	68	T8-T9	13 months	Surgery	Recovery
Bucy & Ritchey (1947)[11]	M	33	C6-T5	10 weeks	Surgery & RT	Improved
Taylor et al. (1951)[12]	F	51	T3-T6	9 years	Surgery	Improved
	F	44	T11-L1	2 years	Surgery	Improved
Lo Re & Michelacci (1969)[13]	M	16	Caudal	Unknown	Surgery	Recovery
	F	35	Lumbar	Unknown	Surgery	Recovery
Pearson et al. (1970)[14]	F	44	T7-T10	3 years	Surgery	Recovery
Henry et al. (1971)[15]	M	63	Cervico-thoracic	4 months	No surgery	Necropsy
Bender et al. (1974)[16]	F	50	T6-T8	4.5 years	Surgery	Recovery
	M	58	T7-T10	3 years	Surgery	Recovery
Occhiogrosso & Vailati (1977)[17]	F	34	T6-T8	Unknown	Surgery	Recovery
Obrador et al. (1977)[18]	F	43	T7-T9	Unknown	Surgery	Recovery
Cull et al. (1978)[19]	F	50	T8-T9	17 years	Surgery	Recovery
	F	45	T6-T8	5 years	Surgery	Improved
Shuangshoti & Hongsaprabhas (1979)[20]	M	45	T10-T11	Unknown	Surgery	Unknown
Schiffer et al. (1980)[21]	F	48	T10-L1	3 years	Surgery	Recovery
Miki et al. (1981)[22]	F	46	T3-T5	2 years	Surgery	Recovery
Padovani et al. (1982)[23]	M	50	T4-T6	1 year	Surgery	Recovery
Haddad et al. (1986)[3]	M	34	T5-T6	1.5 years	Surgery	Recovery
	M	22	T7-T8	2.5 years	Surgery	Recovery
Griebel et al. (1986)[24]	F	53	T5-T6	2 months	Surgery	Recovery
Nishiura et al. (1986)[25]	M	42	T3-T6	5 months	Surgery	Improved
	M	24	L5-S1	3 years	Surgery	Recovery
	M	45	T3-T6	6 months	Surgery	Improved
Matsushima et al. (1987)[26]	F	41	T9-T10	2 months	Surgery	Recovery
Poon et al. (1988)[27]	F	65	T8-T9	6 months	Surgery	Recovery
Anson et al. (1990)[28]	F	58	T3-T5	3 months	Surgery & RT	Recovery
	F	65	T1-T6	2 days	Surgery	Unchanged
Mascalchi et al. (1991)[29]	F	42	T5-T6	1 year	Surgery	Recovery
Weill et al. (1991)[30]	F	46	T7-T9	1 year	Surgery	Recovery
Stranjalis et al. (1992)[31]	F	68	T5-T6	1 year	Surgery	Improved
Pagni et al. (1992)[32]	F	59	L4-L5	27 years	Surgery	Recovery
Rubin et al. (1992)[33]	F	58	T8-T10	2 years	Surgery	Recovery
Preul et al. (1993)[4]	F	45	T7-T11	2 years	Surgery	Improved
Shibata et al. (1993)[34]	F	38	T4-T6	6 months	Surgery	Improved
Yamashita et al. (1993)[35]	M	57	T3-T9	2 years	Surgery	Recovery
Bouramas et al. (1995)[36]	F	27	T2-T8	2 months	Surgery	Recovery
O'Donovan et al. (1996)[37]	M	54	T3-T9	1 month	Surgery	Recovery
Provenzale et al. (1996)[38]	F	38	Lumbar	3 years	Unknown	Unknown
	F	61	Thoracic	2 years	Unknown	Unknown
	F	42	Thoracic	2 years	Unknown	Unknown
Trabulo et al. (1996)[39]	F	26	T2-T9	8 months	Surgery	Recovery
Bala Krishnan et al. (1996)[40]	F	55	T6-T9	2 years	Surgery	Recovery
Boockvar et al. (1997)[41]	F	34	T3-T8	5 months	Surgery	Recovery
Shuangshoti & Lerdlum (1997)[42]	M	11	T1	2 weeks	Surgery	Improved
Oge et al. (1999)[43]	M	72	T2-T5	4 days	Surgery	Recovery
Labram et al. (1999)[44]	F	40	C7-T2	6 years	Surgery	Recovery
	M	17	C5-T3	12 hours	Surgery	Recovery
Turgut (1999)[45]	F	54	T4-T9	5 months	Surgery	Recovery
Fourney et al. (2000) (present case)	F	46	T7-8	5 months	Surgery	Recovery

Abbreviations: M = male; F = female; RT = radiotherapy

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Table 2: Spinal Epidural Angiolipomas Involving Bone

Author (year)	Sex	Age (years)	Site	Bone involvement	Duration of Symptoms	Treatment	Result
Gonzalez-Crussi et al. (1966)[46]	F	20	L3	L3 body & lamina	3 years	Surgery & RT	Recovery
Pearson et al. (1970)[14]	F	17	T2-T5	T4 pedicles	1 month	Surgery	Recovery
	M	22	T3-T9	T5 body & pedicle	1 year	Surgery	Recovery
Schiffer et al. (1980)[21]	F	48	T10-L1	L1 body	3 years	Surgery	Recovery
von Hanwehr et al. (1985)[47]	M	35	T6	T6 body & pedicle	1 year	Surgery	Recovery
Rivkind et al. (1986)[48]	M	52	T6-T8	T7 body & pedicle	2 weeks	Surgery	Recovery
Kuroda et al. (1990)[49]	F	73	T4	T4 body & pedicle	Unknown	Surgery	Improved
Pagni et al. (1992)[32]	F	56	L3	L3 body & pedicle	12 years	Surgery	Recovery
Preul et al. (1993)[4]	M	58	T3	T3 lamina	Unknown	Surgery	Unchanged
Trabulo et al. (1996)[39]	M	72	T5-T6	T6 body & pedicle	6 months	Surgery	Improved
Sakaida et al. (1998)[50]	M	72	T3-T5	T3-T5 bodies & pedicles	4 months	Surgery	Recovery
Labram et al. (1999)[44]	F	68	T5-T10	T7 body	6 weeks	Surgery	Recovery
Kujas et al. (1999)[51]	F	67	T6	T6 pedicle	6 months	Surgery	Worsened

Abbreviations: M = male; F = female; RT = radiotherapy

Table 3: Spinal Intramedullary Angiolipomas

Author (year)	Sex	Age (years)	Site	Duration of Symptoms	Treatment	Result
Palkovic et al. (1988)[52]	M	27	C6-T2	5 months	Surgery	Recovery
Weill et al. (1991)[30]	F	27	T5-T8	Several weeks	Surgery	Improved
Preul et al. (1993)[4]	F	36	T5-T8	1 year	Surgery	Recovery
Maggi et al. (1996)[53]	F	8	T11-L2	3 months	Surgery	Improved
Klisch et al. (1999)[54]	F	34	C6-T4	8 weeks	Surgery	Improved

Abbreviations: M = male; F = female

Physical examination demonstrated diminished pain and light touch to the T10 dermatome as well as poor joint position sense in the lower extremities. Lower limb reflexes were symmetrically brisker than in the upper extremities. Bilateral Babinski responses were present. Tone was within normal range. Motor weakness was mild, affecting only the right knee flexors (+4/5). No atrophy or fasciculations were noted. Spinal examination was unremarkable with no local tenderness or deformity. The remainder of the systemic examination was normal.

Plain computerized tomography (CT) showed a dorsal epidural hypodense mass extending from T3-T11 with the maximum canal compromise at the T7-8 level (Figure 1). The measured Hounsfield value was approximately -31, indicating a predominantly lipomatous structure. There were no associated bony abnormalities. Head and whole-spine magnetic resonance (MR) imaging further showed that the lenticular-shaped mass extended laterally into the neural foramina from T6 to T8 predominantly on the right side and, to a lesser degree, on the left. On T1-weighted images the lesion was homogeneously hyperintense, similar to epidural fat (Figure 2). The T2 signal on a turbo spin-echo sequence was also hyperintense, again similar to epidural fat. The mass enhanced with Gadolinium administration.

A T6-T8 bilateral laminectomy using a high-speed drill was performed. A dark yellow, globular, firm mass with several small vessels on its surface was seen overlying and compressing the dural sac. It was easily dissected from the dura but bled profusely from multiple small epidural feeding vessels. Blood loss was estimated to be 1000 cc. The mass extended into normal-appearing epidural fat at its poles. The dura was not opened. The specimen, which was removed in a single piece, took the mold of two spinal foramina bilaterally (Figure 3). It measured 5.5 cm in length, up to 3.5 cm in width, and was 1.2 cm in maximal thickness.

Histologic examination of the tumor showed mature adipose tissue interlaced with numerous blood vessels ranging from distended capillaries, venules and blood-filled lakes to small arteries (Figure 4). Endothelial cells showed mild atypia. No mitotic figures were seen and Ki-67 immunostaining revealed a low proliferation rate, with some regional variablility. There was minimal perivascular collagen deposition and only subtle condensation of collagenous connective tissue at the surface, with no definitive encapsulation. A pathological diagnosis of angiolipoma was made.

The postoperative course was uneventful. The patient showed improvement in strength and sensation in the days following surgery. Two years later, the patient is asymptomatic with normal muscle tone, strength and sensation. Plantar responses have reverted to normal.

Discussion

Spinal angiolipomas are rare benign tumors containing mature fat cells with an excessive degree of vascular

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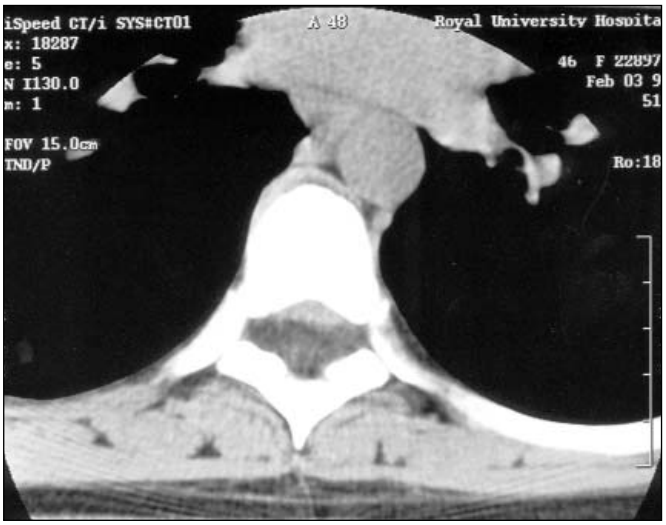

Figure 1. *Nonenhanced axial CT scan at the T7 level. A fat density mass in the dorsal epidural space causing marked spinal compression is seen.*

proliferation. In the past, various terms including vascular lipoma, hemangiolipoma and fibromyolipoma have been used to describe these lesions.[3] They are distinct from extradural spinal lipomas, which are either iatrogenic (due to exogenous corticosteroids) or associated with congenital myelovertebral malformations.[56]

A thorough review of the world literature yielded 75 cases of spinal angiolipoma, including our report (Tables 1-3). There were 57 cases (76%) of spinal epidural angiolipoma without bone involvement and 13 cases (17%) associated with bone infiltration. We identified five cases (7%) of intramedullary angiolipoma. Angiolipomas also occur intracranially, although this is even more rare.[57]

There were 26 males (35%) and 49 females (65%) with a female-to-male ratio of 1.9:1. A hormonal influence on the development, maintenance, or progression of spinal angiolipomas is theorized to account for the female preponderance.[4] The mean age at diagnosis was 44.0 years (range 17-73 years), with women generally older than men (mean ages 45.4 years and 41.2 years, respectively). Patients with intramedullary angiolipomas were younger (mean age 26.4 years).

All patients presented with some degree of neurological deficit in the lower limbs, ranging from a mild sensory deficit to complete motor loss. The average duration of symptoms was 27.6 months (range 12 hours to 27 years) in the 64 patients for which data was available. Vascular factors may contribute to a more rapid onset of symptoms in some cases, possibly due to the gradual enlargement of anomalous vessels, intralesional thrombosis, hemorrhage or steal phenomena.[23,28,44] A few cases with a rapidly deteriorating course have also been reported in pregnancy.[4,19]

Most spinal angiolipomas involve the thoracic spine with 53% of cases involving the midthoracic (T6-T9) region.[4] The reason for this is unknown and is out of proportion to the relative length of each vertebral segment. Almost all noninfiltrating epidural angiolipomas are posterior or posterolateral in location, with four exceptions reported.[5,25,32,44] One of these tumors was diagnosed on pre-operative CT scan as a herniated disc.[32] In another case the tumor extended into the chest cavity over the apex of the right lung through the C7/T1 and T1/T2 neural foramina.[44] Infiltrating angiolipomas are almost always anterior or anterolateral in position with two exceptional cases.[4,46]

Plain x-ray findings in spinal angiolipomas include erosion of the pedicles and widening of the spinal canal. In tumors infiltrating bone, trabeculation of the affected vertebral body may be identified, simulating vertebral hemangioma.[38,49] Myelogram may show a complete or partial block.[33] CT scan usually demonstrates a hypodense lesion consistent with fat density, however some tumors are isodense,[26] probably related to the extent of vascularity. In earlier reports, investigators claimed that spinal angiolipomas exhibited little or no contrast enhancement, allowing a reliable way to distinguish angiolipoma from hemangioma.[49] Contrast enhancement has since been shown to be nonspecific.[4,39]

MR imaging was performed in 34 cases.[4,26,28-31,34-45,48-54] Spinal angiolipomas are typically hyperintense on noncontrast

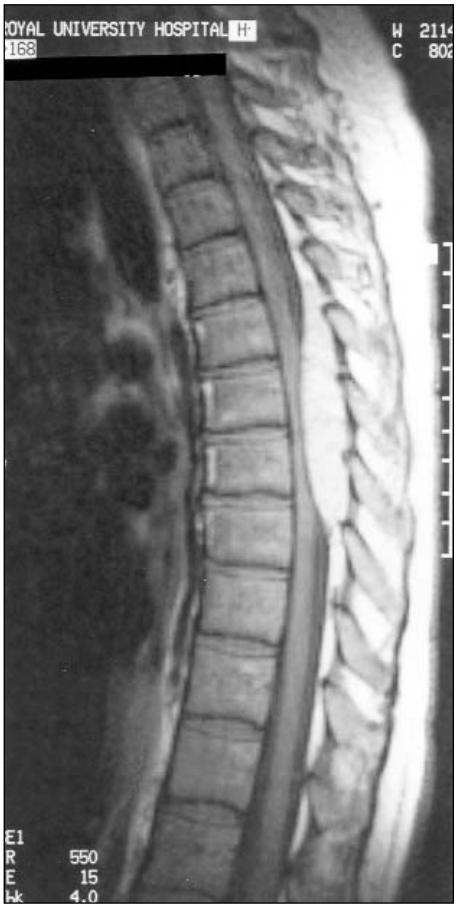

Figure 2. *Nonenhanced sagittal T1-weighted MRI. A homogeneously hyperintense lenticular-shaped mass, dorsal to the spinal cord, is seen at T3-T11 with most marked cord compression at T7-8.*

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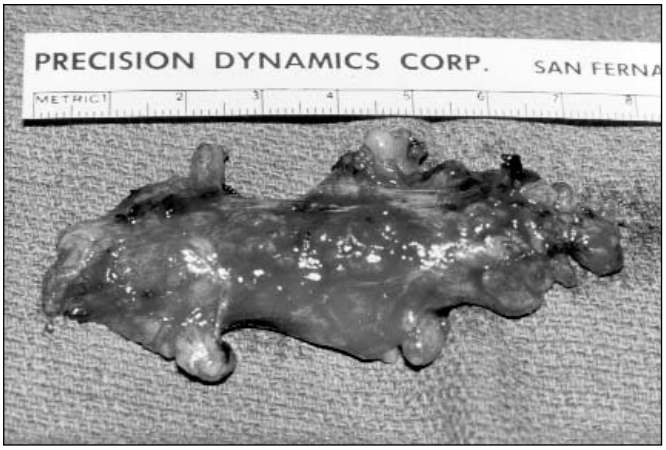

Figure 3. *Gross appearance of the entire surgical specimen demonstrating nipple-like extensions where the tumor took the shape of the intervertebral foramina.*

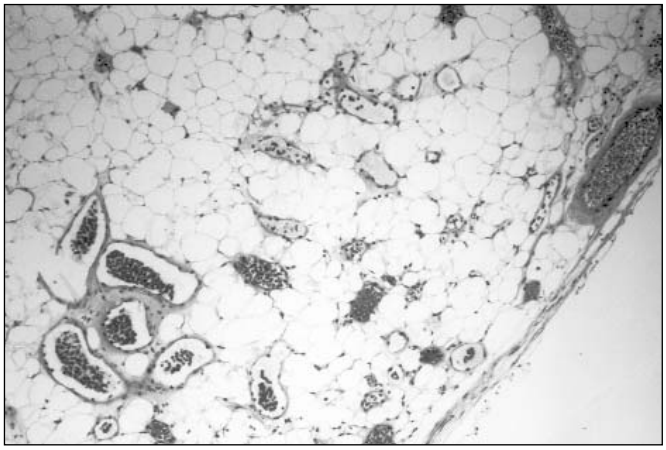

Figure 4. *Photomicrograph of the tumor shows mature adipose tissue interspersed with blood channels of various sizes, indicative of angiolipoma. Note the condensation of collagenous tissue at the surface (hematoxylin-eosin, original magnification X 200).*

T1-weighted images owing to their fatty content.[52] Large hypointense foci on noncontrast T1-weighted images correlate with increased vascularity.[38] Most spinal angiolipomas enhance and fat suppression MR in conjunction with contrast administration better defines the borders of the tumor and aids in surgical planning.[38]

Angiolipomas are histologically characterized by mature adipose tissue and prominent abnormal blood vessels.[1,2] The ratio of fat to vessels is variable and ranges from 1:3 to 2:3.[3] The blood channels range from capillary, to sinusoidal and venular, to arterial in size with irregularly thickened walls. Tumors with an abundance of smooth muscle proliferation are further subclassified as angiomyolipomas.[14,47] Vessels may lack a definitive adventitia, with smooth muscle cells merging into the surrounding adipose tissue.[4] The vascular stroma may be localized or diffuse and is occasionally interrupted by bands of fibrosis. The presence of cartilage and osteoid tissue has been observed in some cases.[4,14] A thin capsule, often defective in many areas, may surround the lesion. The degree of nuclear atypia is minimal. Low proliferation rates are found on Ki-67 immunostaining. Mitotic figures are rare and there are no reported cases of malignant transformation.

The histiogenesis of these tumors is unknown but several theories have been advanced. They may originate from primitive pluripotential mesenchymal cells which normally gives rise to adipose tissue, smooth muscle and vascular endothelium.[1,16] Others have considered the tumor to be a congenital malformation[11] or a benign hamartoma.[58] Hemangiomas and lipomas may represent a spectrum within which angiolipomas constitute an intermediate entity.[2] The more invasive infiltrating type of spinal angiolipoma would then represent a shift towards the hemangioma end of the spectrum.[23]

Spinal lipomas differ from angiolipomas in that they are most commonly found in the lumbosacral (rather than mid-thoracic) region and may be associated with dysraphic abnormalities.[20] Ossification of the neural arch begins in the thoracic region, taking place between the 6th week and 4th month of fetal development. An early inclusion of pluripotential stem cells in this area may be a prerequisite for spinal angiolipoma, while a developmentally later inclusion of mature adipocytes in the lumbosacral region (the region of the last closure site of the embryonic neural arch) may lead to the formation of spinal lipoma.[34] This hypothesis may help to explain the differences in location between spinal lipoma and angiolipoma and the association of lipomas with dysraphism. Further, since ossification of the vertebrae first develops in the ventral and lateral spinal canal, a relatively more immature pluripotential stem cell (which is thought to have more infiltrative potential) is more likely to be included that region.[50] This may help explain the strong predilection of the infiltrative spinal angiolipomas to be located anteriorly.

The treatment of spinal angiolipomas is surgical removal and the prognosis after surgical management is very good. Either recovery or improvement in neurological function was seen in 62 of 65 operated patients (95.4%) for whom results were reported (recovery in 47/65 = 72.3%; improvement in 15/65 = 23.1%). Two patients[4,28] were unchanged postoperatively (2/65 = 3.1%). The postoperative course worsened "transiently" in one patient[51] (1/65 = 1.5%) who underwent laminectomy for an infiltrating spinal angiolipoma. This patient was later subjected to a second operation. However the results of this procedure were not stated.

An anterior transthoracic approach is recommended for infiltrating spinal angiolipomas involving the vertebral body.[47,48] However, in all but three reports[47,48,50] these tumors were approached with a laminectomy. As a result, only three authors reported a gross total resection.[4,47,48]

In three cases, postoperative radiotherapy was given following a partial excision because of concerns for potential malignancy.[11,28,46] No adjuvant radiation should be given to patients with this benign pathological entity, since even in the infiltrating group, the prognosis is very good. Only one recurrence of an angiolipoma has ever been reported,[16] with a successful re-operation 12 years after the first surgery. There were a total of four deaths, with three occurring in very early

reports.[5,6,9] More recently, a patient who had never been operated on, died from a cervical glioblastoma.[15]

Conclusions

In summary, spinal angiolipoma is a rare but well-defined entity with distinct clinicopathological and imaging features. Outcome following surgery is very good overall. Improved imaging techniques have increased the number of reported cases over the past few years. Like their "noninfiltrating" counterparts, spinal angiolipomas with an associated bony component are benign tumors with indolent growth, hardly deserving the aggressive connotation suggested by the term "infiltrating".[39] Although complete removal is certainly preferred, outcomes remained favorable despite incomplete resections in a number of ventral "infiltrating" tumors.[14,21,32,39,44,46,49,50]

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