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Case Report

Senile tanycytic ependymoma of the spinal cord: a case report and literature review

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Abstract: Tanycytic ependymoma is a rare subtype of ependymoma that most commonly occurs in the brain. Senile tanycytic ependymoma that arises from the spinal cord is extremely rare. The present study reported the case of a 65-year-old woman who presented with a 4-day history of numbness in both lower limbs accompanied by an increased urinary frequency and constipation. Magnetic resonance imaging (MRI) of the lumbar vertebrae indicated an oval intradural mass at the level of the second lumbar vertebra, which exhibited an equal intensity on the T1-weighted image (T1WI) and a slightly increased intensity on the T2-weighted image (T2WI). A complete resection of the tumor was performed. A histopathological examination indicated that the tumor cells were elongated, bipolar and spindle-shaped with different densities and formed a palisade arrangement that varied in width. The cells were immuno positive for GFAP and S-100 protein, as well as weakly positive for Syn. Furthermore, we reviewed 55 cases of tanycytic ependymoma indexed in PubMed since 1978. We determined that the patients with tanycytic ependymomas in the spinal cord were predominantly female and older than the individuals with tumors in the brain. Tanycytic ependymomas in the brain and spinal cord had a similar appearance via MRI and pathology examinations.

Keywords: Tanycytic ependymoma, brain, spinal cord, pathology, magnetic resonance imaging

Introduction

Tanycytic ependymoma comprises a rare subtype of ependymoma, which was classified as a grade II tumor according to the World Health Organization (WHO) 2000 classification of brain tumors [1, 2]. The histomorphological features of tanycytic ependymoma are characterized by spindle cells; thus, it is difficult to discern them from the cells of other tumors, such as astrocytoma and neurilemmoma [3]. In contrast to other ependymomas, tanycytic ependymomas frequently occur in the spinal cord [4]. The identification of the clinical manifestations and pathological features of these tumors is of substantial significance for an accurate diagnosis. In this article, we reported a rare case of tanycytic ependymoma that arose from the cauda equina of an elderly woman. Moreover, we also reviewed all 55 cases of tanycytic ependymoma indexed in PubMed since 1978.

Case presentation

A 68-year-old woman was admitted to our hospital on November 18, 2014. She had developed a sensation of numbness in her lower limbs 4 days earlier accompanied by an increased urinary frequency and constipation. Hypoesthesia occurred below the second lumbar vertebra. The myodynamia of the lower limbs was grade IV. Magnetic resonance imaging (MRI) demonstrated an oval intradural extramedullary tumor at the level of the second lumbar vertebra. It exhibited an iso-intensity on the T1-weighted image (T1WI), a slightly increased signal intensity on the T2-weighted image (T2WI) and a slightly decreased signal intensity on the T2WI with fat suppression. The nerve roots at the corresponding level in the cauda equina were compressed. The mass was enhanced by gadolinium-diethylene triamino pentaacetic acid (Gd-DTPA), and the angiography indicated significant tumor labeling (**Figure 1**). A clinicoradiologic diagnosis of an intraspinal tumor was made, which appeared to be neurogenic.

Under general anesthesia, resection of the intraspinal extramedullary tumor was performed. There was less epidural fat; however, the spinal

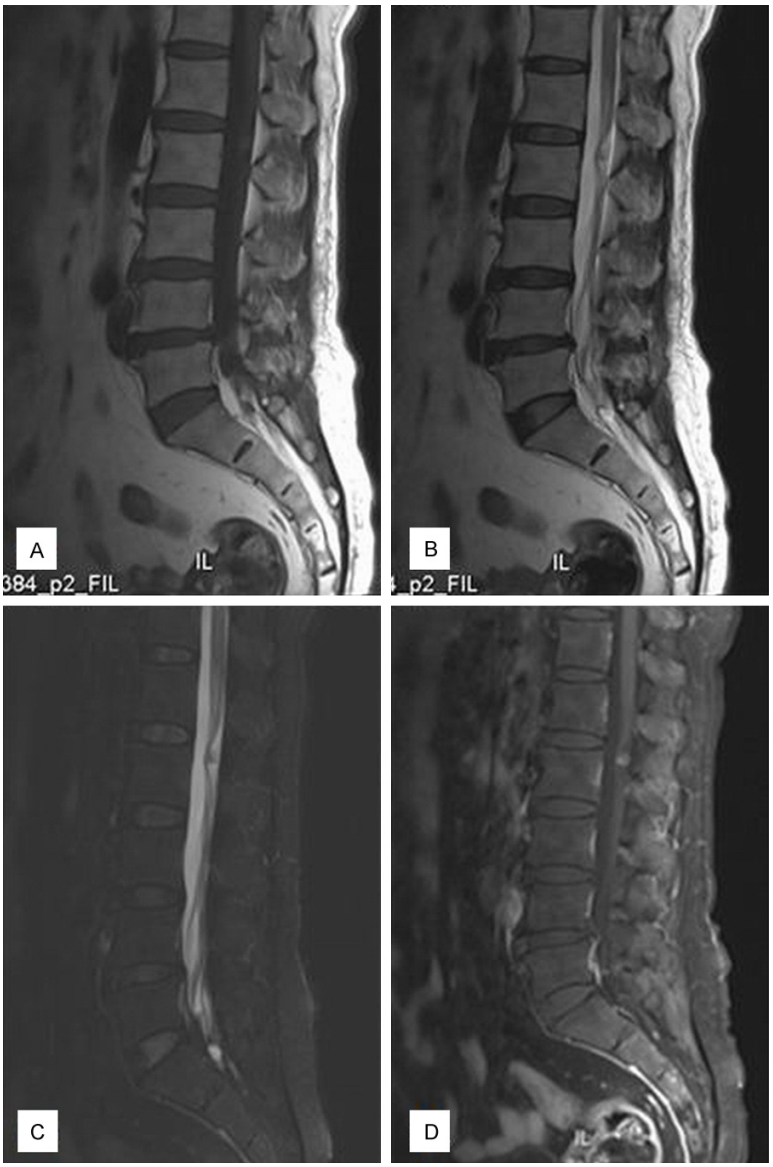

Figure 1. Preoperative MR imaging (A) Sagittal magnetic resonance imaging (MRI) demonstrated an oval intradural tumor at the level of the second lumbar vertebra, which exhibited an equal signal intensity on the T1WI. (B) Sagittal T2WI, in which the lesion was slightly hyperintense. (C) Sagittal T2WI image with fat suppression, in which the lesion exhibited a slightly decreased signal intensity and the nerve roots at the corresponding level in the cauda equina were compressed. (D) The mass was enhanced by Gd-DTPA, and the angiography indicated significant tumor labeling.

dura mater tension was moderate, and no obvious pulsation was identified. Following the incision of the spinal dura mater, a gray, tenacious tumor with an integral capsule and a moderate blood supply was identified. One side of the tumor adhered to a nerve root in the cauda equina and was freed by electrocoagulation. The tumor, which measured 1.0×0.8×0.6 cm, was completely dissected. The postoperative clinical course was uneventful, and the patient was neurologically intact with the exception of substantially relieved hypoesthesia below the level of the knees. She was discharged from the hospital on postoperative day 12. No radiation therapy was administered.

The tumor fragments were 0.6×0.4×0.3 cm. The cross-section of the tumor had a gray and fragile appearance, similar to fresh fish meat. No areas exhibited calcification. Histopathologically, the tumor cells were elongated, bipolar and spindle-shaped with different densities. These cells formed a palisade arrangement, which varied in width. No classic ependymal rosettes were identified. Immunohistochemical staining indicated that GFAP and S-100 protein were positive, whereas Syn was weakly positive (**Figure 2**). The pathological diagnosis was tanycytic ependymoma. The patient was neurologically intact 1 month after surgery. According to the MRI evaluations, she has remained free from a recurrence for 12 months and has a favorable prognosis.

Literature review and analysis

In combination with the 55 cases indexed in PubMed since 1978, we have summarized the clinical data for the reported cases of tanycytic ependymomas (**Table 1**). With the exception of 1 tumor located in both the brain and spinal cord, 21 tumors were located in the brain and 34 tumors were located in the spinal cord. Data regarding the genders and ages of the patients were complete in all reports. The T1WI changes were described in 20 cases,

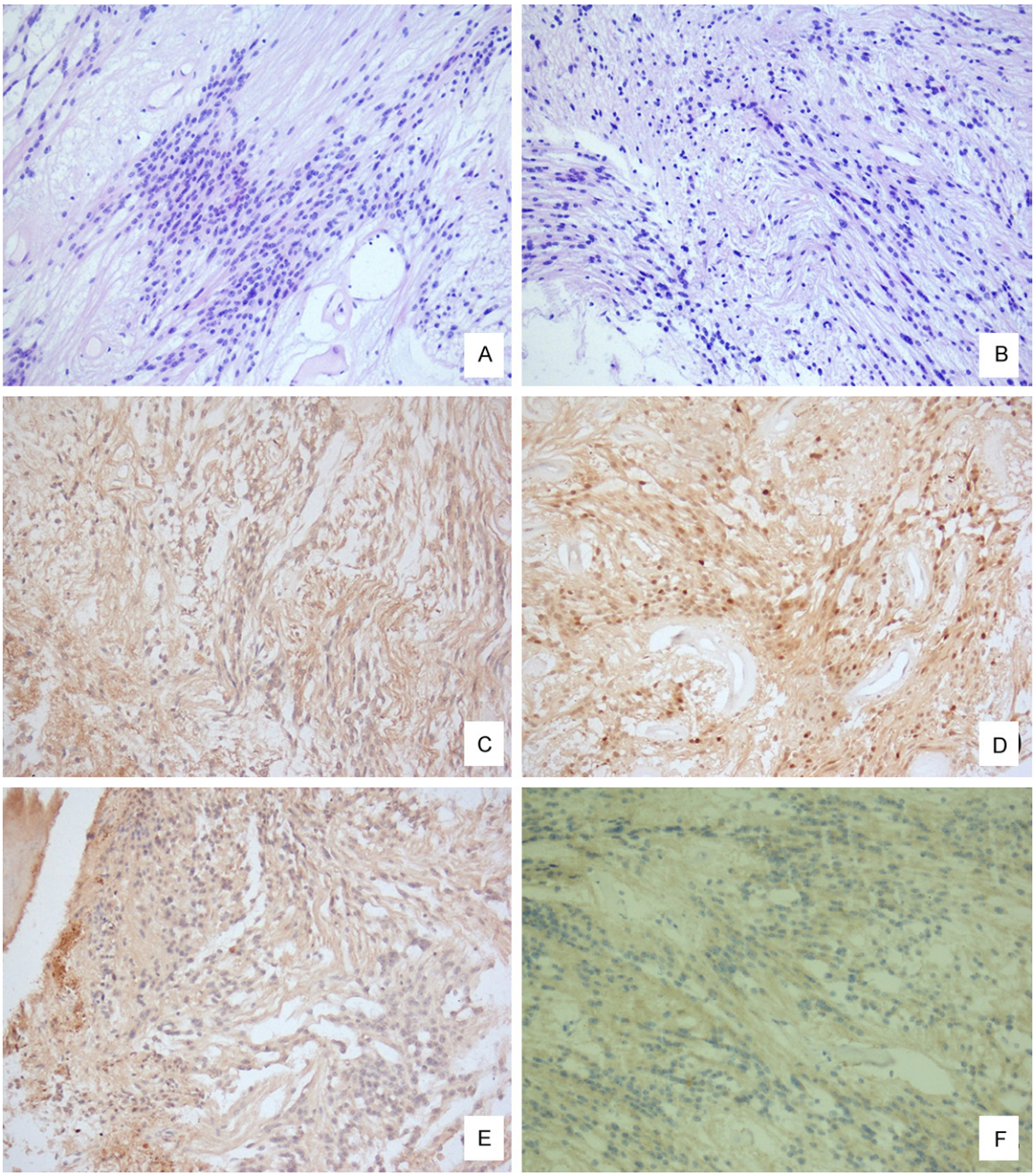

Figure 2. Examination of postoperative pathology. Histopathologically, the tumors consisted of elongated, bipolar and spindle-shaped cells (A, ×200). These cells with different densities formed a palisade arrangement, which varied in width (B, ×200). Immunohistochemical staining indicated tumor cells positive for GFAP (C) and S-100 protein (D), as well as weakly positive for Syn (E), but negative for EMA (F).

which comprised 8 brain cases and 12 spinal cord cases. The T2WI changes were described in 25 cases, which comprised 9 brain cases and 16 spinal cord cases. Cystic degeneration was described in 36 cases, which included 14 brain cases and 22 spinal cord cases. Tumor enhancement was described in 36 cases, which comprised 14 brain cases and 22 spinal cord cases. Rosettes or pseudorosettes were identified in 18 cases, which included 8 brain cases and 10 spinal cord cases. Chi-square and t tests were used to analyze the differences in the gender, age, MRI manifestations and pathological features of these cases.

Table 1. Summary of previously reported tanycytic ependymoma cases

Source [reference]	Gender/age (years)	Location	Radiographic features				Pathological features
			T1WI	T2WI	Cystic	Gadolinium	
Friede et al. [7]	M/10	1	ND	ND	ND	ND	0
Friede et al. [7]	M/34	1	ND	ND	ND	ND	1
Friede et al. [7]	M/6	1	ND	ND	ND	ND	0
Friede et al. [7]	M/3.5	1	ND	ND	ND	ND	0
Langford et al. [25]	F/40	1	ND	ND	ND	ND	0
Langford et al. [25]	M/13	1	ND	ND	ND	ND	0
Hayashi et al. [26]	M/51	1	ND	ND	1	1	1
Daneyemez et al. [27]	M/42	1	ND	ND	1	ND	1
Richards et al. [28]	M/17	1	ND	ND	ND	1	0
Ragel et al. [29]	F/55	1	3	3	2	1	1
Ito et al. [1]	M/59	1	1	3	2	1	3
Zhang et al. [4]	M/38	1	1	3	2	1	0
Vajtai et al. [30]	F/40	1	ND	ND	2	1	0
Luigi et al. [31]	M/14	1	1	3	1	1	1
Agarwal et al. [32]	F/44	1	2	2	2	ND	0
Saumya et al. [33]	M/8	1	2	2	1	1	2
Lopez et al. [34]	M/58	1	2	3	1	1	0
Reis et al. [35]	M/6	1	3	1	1	1	0
Divito et al. [36]	M/57	1	ND	3	2	1	1
Leonidas et al. [37]	M/40	1	ND	ND	ND	ND	0
Kuga et al. [38]	M/24	1	ND	ND	1	1	0
Kambe et al. [6]	M/2	1	ND	ND	2	1	0
Friede et al. [7]	F/75	2	ND	ND	ND	ND	0
Friede et al. [7]	F/38	2	ND	ND	ND	ND	0
Friede et al. [7]	F/46	2	ND	ND	ND	ND	3
Friede et al. [7]	M/45	2	ND	ND	ND	ND	0
Friede et al. [7]	M/17	2	ND	ND	ND	ND	0
Friede et al. [7]	F/36	2	ND	ND	ND	ND	0
Friede et al. [7]	F/35	2	ND	ND	ND	ND	0
Spaar et al. [39]	M/32	2	ND	ND	ND	ND	ND
Langford et al. [25]	F/52	2	ND	ND	ND	ND	0
Dvoracek et al. [12]	F/31	2	ND	ND	ND	ND	0
Kawano et al. [20]	M/45	2	ND	ND	2	1	0
Kawano et al. [20]	F/55	2	ND	ND	1	ND	0
Kawano et al. [20]	F/36	2	ND	ND	1	1	3
Ueki et al. [6]	F/18	2	ND	ND	ND	1	1
Kobata et al. [40]	M/30	2	ND	ND	1	1	2
Boccardo et al. [41]	F/39	2	ND	3	ND	ND	1
Ito et al. [42]	M/62	2	2	2	1	1	0
Sato et al. [43]	M/58	2	3	3	1	1	0
Mohindra et al. [13]	F/10	2	ND	ND	1	1	1
Shintaku et al. [14]	F/55	2	2	3	1	1	0
Shintaku et al. [44]	M/43	2	2	1	2	2	0
Lim et al. [3]	F/16	2	ND	3	2	1	0
Present case	F/68	2	2	3	2	1	0
Rosario et al. [45]	F/57	2	ND	ND	2	2	ND

Ishihama et al. [15]	F/40	2	1	3	1	1	0
Julio et al. [46]	F/25	2	ND	3	2	1	0
Tosun et al. [16]	F/41	2	1	3	2	1	0
Ortiz et al. [47]	M/76	2	ND	ND	ND	ND	0
Navneet et al. [48]	M/30	2	2	3	1	1	1
Khaled et al. [49]	M/50	2	ND	low	2	1	0
Funayama et al. [50]	M/53	2	2	2	1	1	0
Radhakrishnan et al. [51]	M/44	2	2	3	1	1	1
Zane et al. [8]	F/66	2	2	3	2	1	1
Kuga et al. [52]	M/24	2	ND	ND	2	1	0
Santiago et al. [36]	M/20	2	3	2	2	ND	1

ND: not described; Location: intracranial-1, intraspinal-2; TI/T2WI: hypointense-1, isointense-2, hyperintense-3; Cystic/Gadolinium: positive-1, negative-2; Pathology: no pseudorosettes & rosettes-0, pseudorosettes only-1, rosettes only-2, both pseudorosettes & rosettes-3.

The ages of all tanycytic ependymomas patients varied from 2 to 75 years (mean: 36.7), and the ratio of male to female patients was 1.375:1. The ages of the patients with tanycytic ependymomas in the brain varied from 2 to 59 years (mean: 30.1), and the ratio of male to female patients was 4.5:1 (18/4). The ages of the patients with tanycytic ependymomas in the spinal cord varied from 10 to 75 years (mean: 40.9), and the ratio of male to female patients was 1:0.75 (15/20). There were 3 cases (3/8) of tanycytic ependymomas in the brain and 2 cases (2/12) of tanycytic ependymomas in the spinal cord that had a decreased signal intensity on the T1WI and an increased signal intensity on the T2WI; 2 cases (2/8) of tanycytic ependymomas in the brain and 2 cases (2/12) of tanycytic ependymomas in the spinal cord exhibited an iso-intensity on both the T1WI and T2WI; only 1 case (1/8) of tanycytic ependymomas in the brain and 5 cases (5/12) of tanycytic ependymomas in the spinal cord exhibited an iso-intensity on the T1WI and an increased signal intensity on the T2WI; 1 case (1/8) of tanycytic ependymomas in the brain and 1 case (1/12) of tanycytic ependymomas in the spinal cord exhibited an increased signal intensity on both the T1WI and T2WI; 1 case (1/8) of tanycytic ependymomas in the brain exhibited an increased signal intensity on the T1WI and a decreased signal intensity on the T2WI; 1 case (1/8) of tanycytic ependymomas in the spinal cord exhibited an increased signal intensity on the T1WI and an iso-intensity on the T2WI. Via MRI, tumor enhancement was identified in 13 cases (13/14) of tanycytic ependymomas in the brain and 19 cases (19/21) of tanycytic ependymomas in the spinal cord. Cystic degeneration was identified in 7 cases (7/14) of tanycytic ependymomas in the brain and 11 cases (11/22) of tanycytic ependymomas in the spinal cord.

The pathological examination indicated there were 14 cases (14/22) of tanycytic ependymomas in the brain and 23 cases (23/33) of tanycytic ependymomas in the spinal cord without rosettes or pseudorosettes; 6 cases (6/22) of tanycytic ependymomas in the brain and 7 cases (7/33) of tanycytic ependymomas in the spinal cord exhibited pseudorosettes; 1 case (1/22) of tanycytic ependymomas in the brain and 1 case (1/33) of tanycytic ependymomas in the spinal cord exhibited rosettes; and 1 case (1/22) of tanycytic ependymomas in the brain and 2 cases (2/33) of tanycytic ependymomas in the spinal cord exhibited both rosettes and pseudorosettes.

Chi-square and t tests were used to analyze the differences in the gender, age, MRI manifestations and pathological features of these cases. The patients with tanycytic ependymomas in the spinal cord were predominantly female and older than the patients with tumors in the brain ($P<0.05$). The tumors mainly demonstrated an iso-intense signal on the T1WI and an increased signal intensity on the T2WI with a slight enhancement and partially with cysts. Rosettes and pseudorosettes were not commonly identified in the pathological examination; however, there were no significant differences in the MRI or pathological presentations between tanycytic ependymomas in the brain and spinal cord ($P>0.05$).

Discussion

There are several subtypes of ependymomas according to the WHO classification [5]. Tanycytic ependymomas comprise a rare subtype of ependymomas [6]. Because the tumor cells have morphological characteristics that resemble tanycytes with slender bipolar tumor cells as a feature, they were described as an ependymoma variant by Friede and Pollack in 1978 [2] and were classified as a variant of ependymoma in the WHO 2000 classification of brain tumors [5, 7]. Based on previous studies of tanycytic ependymomas, 55 cases have been reported (**Table 1**). Most tanycytic ependymoma cases were female. Male morbidity was increased compared with female morbidity [8]. In addition, the spinal cord was the most frequent location of tanycytic ependymomas, followed by the intraventricular and supratentorial extraventricular regions [6, 8]. In other ependymoma cases, the infratentorial region was reported to be the predominant location (60%), whereas the spinal cord was the secondary location, followed by the lateral ventricles and the third ventricle [9]. The location of tanycytic ependymomas was related to age and gender. Patients with tanycytic ependymomas in the spinal cord are older than patients with tanycytic ependymomas in the brain; most of the former were female. However, most patients with ependymomas in the spinal cord were male, and the location of ependymomas was unrelated to gender [10, 11].

Tanycytic ependymomas most frequently arise in the cervical or thoracic spine, whereas tanycytic ependymomas present in the lumbar region are rare, with only three cases described prior to this report [8, 12, 13]. The substantial majority of spinal tanycytic ependymomas are intramedullary, with only four previously reported extramedullary tumors [8, 12, 14-16]; these latter tumors were identified in the lower thoracic or lumbar spine. The patient described in the current report was the oldest and only the second woman over 65 years of age reported to have an extramedullary tanycytic ependymoma of the spinal cord.

MRI is the preferred imaging method used for the assessment of ependymomas. According to previous reports, in general, ependymomas exhibit an iso-intensity or increased signal intensity on the T2WI and a decreased or iso-intense signal on the T1WI. Tumor enhancement is nearly always present; however, it may be heterogeneous or homogeneous, and 19.3% of ependymomas were cystic [11, 17]. The MRI presentation of tanycytic ependymoma varies substantially and does not always exhibit similar imaging features with other ependymomas according to previous reports. The MRI presentation of the current case was iso-intense on the T1WI, with a slightly increased signal intensity on the T2WI and was similar to the previously reported lumbar extramedullary ependymomas. This finding was similar to the MRI presentation of spinal schwannomas and was considered a neurogenic tumor prior to the operation [18]. Tanycytic ependymomas always exhibit an enhancement on gadolinium, and it was similar to the MRI presentation of ependymomas. Cystic degeneration may be identified in half of tanycytic ependymomas; however, it is substantially reduced in common ependymomas. Moreover, ependymomas with cystic degeneration are always located in the supratentorial extraventricles [10]; however, this feature did not apply to tanycytic ependymomas. We have been unable to differentiate tanycytic ependymomas in the brain from tanycytic ependymomas in the spinal cord using MRI. There were several differences in the MRI presentation between tanycytic ependymomas and common ependymomas; however, these differences are not absolutely specific. The focus should be on distinguishing tanycytic ependymomas from neurogenic tumors.

The typical pathologic characteristics of ependymomas are rosette or pseudorosette formation and the presence of abundant astrocytes [19]. Monopolar or bipolar cells of tanycytic ependymomas are elongated and hair-like. The cell nuclei are round, oval or spindle-shaped with a slight pleomorphism and have abundant chromatin distributed similar to salt-and-pepper, which is commonly identified in tanycytes [12]. These cells form a palisade arrangement of varied widths, which differ from other ependymoma cells. Typical ependymoma rosettes are not common; however, perivascular pseudorosettes may occasionally be identified [20]. Tanycytic ependymomas with pseudorosettes were less common than tumors with rosettes or tumors with both of them. This feature was similar to all types of ependymomas. In general, immunohistochemical staining of tanycytic

ependymoma cells indicated that GFAP, S-100 protein and vimentin were positive [21]. EMA was distributed in a dot-like pattern of positivity; however, Syn and NeuN were negative [22]. The MIB-1 labeling index ranged from 0% to 4.60%. The differential diagnosis of tanycytic ependymomas includes schwannoma, pilocytic astrocytoma, and fibrillary astrocytoma [4]. Pilocytic astrocytoma presents a biphasic pattern, with Rosenthal fibers and granular bodies. Fibrillary astrocytoma has microcysts and mucinous degeneration as its morphological characteristics. Histopathologically, the tumor cells in our case were elongated, bipolar and spindle-shaped with different densities. These cells formed a palisade arrangement, which varied in width. Immunohistochemical staining indicated that GFAP and S-100 protein were positive, whereas Syn was weakly positive. The previously described features are consistent with the features of typical tanycytic ependymomas as previously reported.

Surgery is the accepted treatment for tanycytic ependymomas [23]. According to the previous reports of 56 cases, 2 patients died as a result of postoperative complications and 1 patient died of fever and seizures 8 months after the surgery; the remaining 53 postoperative patients had a favorable prognosis. Among all cases, only 2 cases recurred (one patient had no surgery description and the other patient had a subtotal operation). In general, the patients who had completely removed tumors exhibited a favorable prognosis. According to the previous clinicopathological studies of ependymomas, different subtypes exhibited different expression levels of MIB-1 and p53 [24]. Tanycytic ependymomas have the lowest expression of MIB-1 and p53, which suggested the most favorable prognosis. In our case, the tanycytic ependymoma occurred in the cauda equine, and there was no neurological dysfunction after the adherence was cut off. Thus, tanycytic ependymomas that occur in the spinal cord should be completely resected to obtain a favorable prognosis.

In conclusion, tanycytic ependymomas that arise from the spinal cord were more common, whereas extramedullary tanycytic ependymomas were rare. All patients who suffered from extramedullary tumors were senile, and all tumors were located in the lumber spine. In comparison with the patients who suffered from tanycytic ependymomas in the brain, the patients who suffered from tanycytic ependymomas in the spinal cord were older and most of the patients were female. The MRI presentations of the tanycytic ependymomas substantially varied. There was no significant difference in the MRI presentations between the tanycytic ependymomas in the spinal cord and brain. Cystic degeneration was more common in tanycytic ependymomas compared with ependymomas. The MRI presentations of the tanycytic ependymomas were different from the presentations of ependymomas; however, these differences were not absolutely specific. Thus, the preoperative image detection may mislead the clinical diagnosis. A pathological examination is of substantial significance in the differentiation of tanycytic ependymomas and astrocytomas. However, the pathology changes were not significantly different in tanycytic ependymomas that arose from the brain and spinal cord. Surgery is the principal method for tanycytic ependymoma treatment. The tumor should be completely resected to obtain a favorable prognosis. Senile tanycytic ependymomas that arose from the spinal cord were rare.

Disclosure of conflict of interest

None.

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