

# Paraganglioma in the Lumbar Spinal Canal Treated by Surgery Assisted by MRI Neuroimaging and 3D Printing Reconstruction: A Case Report and Literature Review

**Ya Zhang**

The Third Affiliated Hospital of Kunming Medical University: Yunnan Cancer Hospital

**Dongqi Li**

The Third Affiliated Hospital of Kunming Medical University: Yunnan Cancer Hospital

**Kun Li**

The Third Affiliated Hospital of Kunming Medical University: Yunnan Cancer Hospital

**Lijuan Ye**

The Third Affiliated Hospital of Kunming Medical University: Yunnan Cancer Hospital

**Cao Wang**

The Third Affiliated Hospital of Kunming Medical University: Yunnan Cancer Hospital

**Yihao Yang**

The Third Affiliated Hospital of Kunming Medical University: Yunnan Cancer Hospital

**Qiuyun Chen**

The Third Affiliated Hospital of Kunming Medical University: Yunnan Cancer Hospital

**Yan Liu**

The Third Affiliated Hospital of Kunming Medical University: Yunnan Cancer Hospital

**Tiying Wang**

The Third Affiliated Hospital of Kunming Medical University: Yunnan Cancer Hospital

**Kecheng Li**

The Third Affiliated Hospital of Kunming Medical University: Yunnan Cancer Hospital

**Zunxian Tan**

The Third Affiliated Hospital of Kunming Medical University: Yunnan Cancer Hospital

**Yanan Zhu**

The Third Affiliated Hospital of Kunming Medical University: Yunnan Cancer Hospital

**Jiaxiang Chen**

The Third Affiliated Hospital of Kunming Medical University: Yunnan Cancer Hospital

**Xinchao Yu**

The Third Affiliated Hospital of Kunming Medical University: Yunnan Cancer Hospital

**Zuozhang Yang** (✉ [yangzuozhang@163.com](mailto:yangzuozhang@163.com))

The Third Affiliated Hospital of Kunming Medical University: Yunnan Cancer Hospital <https://orcid.org/0000-0002-0763-5805>

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## Case report

**Keywords:** spinal canal, paraganglioma, Tumor resection.

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# Abstract

**Background:** Ganglion cell paraganglioma is a subtype of neuroendocrine tumors, which is rare in clinical practice. Paraganglioma in the spinal area, especially in the spinal canal, is even rarer.

**Case presentation:** The case we reported was a 46-year-old female who was admitted to the hospital due to "physical examination found a mass in the lumbar spinal canal for more than 2 months." After admission, the relevant examinations and preoperative preparations were perfected, and the posterior L2-3 spinal mass resection + spinal decompression + nail rod system internal fixation was performed. The patient recovered well after the operation, and there was no tumor recurrence in follow-up.

**Conclusions:** Surgical treatment assisted by MRI neuroimaging and 3D printing reconstruction technology may be an effective way to treat paraganglioma in the lumbar spinal canal.

## 1 Background

Gangliocytic paraganglioma (gangliocytic paraganglioma) belongs to the neuroendocrine tumor subtype and is clinically rare. Paraganglioma is a tumor that originates in the neuroectoderm and mainly occurs in the adrenal medulla. Approximately those occurring outside the adrenal gland 10%, it is rare in the spine and spinal canal<sup>[1]</sup>. At present, there are few reports in the literature, with more than 200 cases reported abroad<sup>[2]</sup>, and only more than 20 cases reported in China. Paraganglioma in the spinal canal (especially paraganglioma in the lumbar spinal canal) is mainly manifested as symptoms related to nerve compression, which is often misdiagnosed as a herniated disc in clinical practice. We reported a case of paraganglioma in the lumbar spinal canal, combined with relevant literature reports to summarize and analyze this type of case, and then improve the understanding of this disease.

## 2 Case Presentation

### 2.1 Introduction to medical records

The patient, female, 46 years old, came to our hospital on February 26, 2019 due to "physical examination found a mass in the lumbar spinal canal for more than 2 months". The patient went to the "First Affiliated Hospital of Kunming Medical University" in December 2018, and found a mass in the lumbar spinal canal, occasional touch of the left lower extremity, no pain, and no mobility disorders. The patient was admitted to our hospital for further diagnosis and treatment. There is no history of hypertension, no history of exposure to tuberculosis, and no family history of tumors. Physical examination: Physiological curvature of the spine exists, no scoliosis deformity, no tenderness, no percussive pain in the spinous process of cervical, thoracic and lumbar vertebrae, no percussion pain, lumbar spine movement, no obvious abnormalities in muscle strength and muscle tension of both lower limbs, and physiological reflexes exist. Pathological reflex is not elicited. DR showed that the density of the soft tissue around the L2 and L2/3 intervertebral space was increased. It is recommended that the lumbar spine be combined with MRI (Fig. 1A, 1B). MRI showed: L2/3 intervertebral disc level intravertebral canal extramedullary intradural round nodules, clear boundaries, size about 1.6cm × 1.5cm × 2.0 cm, T1 is slightly longer T2 signal, enhanced scan is significantly enhanced, and the final The filaments are closely related, and the neuroimaging shows better. Consider neurogenic tumors, please combine with the clinical; 2. The shape, structure and signal of the remaining spinal cord are not abnormal; 3. The proximal nerve roots on both sides of the neck, thorax and lumbar are small nodules, Small cystic enlargement changes, long T1 long T2 signal, no obvious enhancement in enhanced scan, observation on neuroimaging images is clearer and more intuitive, the nerve root runs distally, and the shape is not obvious. Consider multiple nerve root sheath cysts. (Fig. 1D-G). The preoperative NSE value was 17.51 µg/L↑ (reference range is 0-16.3 µg/L), and other laboratory tests showed no abnormalities.

### 2.2 Admission to hospital

After admission, complete various preoperative examinations. Under general anesthesia, "L2 and 3 laminectomy decompression + spinal mass removal + nail rod system internal fixation", L2 and 3 segments can be seen after L2 and 3 lamina The dural sac is full. After opening the dura, a mass about 2.0cm × 2.0cm × 1.5 cm can be seen. The envelope is intact, dark red, and the surface is smooth. The tumor is connected to the terminal filaments. The superficial part of the tumor is connected to the cauda equina Nerve adhesions are mild, the boundary between the tumor and the dural sac is clear, the adhesions are carefully separated, and the tumor is completely removed when the nerve terminal filaments are disconnected. Carefully stop the bleeding and suture the dura mater. The internal fixation system is installed in the conventional L2 and 3 segments, drainage tube is placed, and suture is layer by layer. On the 7th day, he recovered and was discharged.

### 2.3 Postoperative pathology and review

Postoperative pathological examination showed that the epithelioid cells constitute the tumor parenchyma, the cell morphology is uniform, the cytoplasm is abundant, the nucleolus is not obvious, the mitoses are rare, and the arrangement is acinar; the interstitial is sinusoidal and a little fibrous (Fig. 3A). Immunohistochemistry showed that the tumor cells were strongly positive for CgA, NSE, and Syn (Fig. 3BCD). Pathological diagnosis: Paraganglioma in L2 and 3 spinal canal. Postoperative DR showed that the physiological curvature of the lumbar spine became flat, and the lumbar 1–3 vertebral and paravertebral metal internal fixation were in normal position (Fig. 2C-F). The postoperative function recovered well, and no tumor recurrence was found in the 14-month follow-up.

### **3 Literature Review**

In order to review the cases of paraganglioma in the spinal canal at home and abroad, we use the keywords "spinal cannal' and 'paraganglioma' to search for cases in Medline for nearly 40 years. Using this strategy, we found 26 articles and 47 cases were reported. The reported case and the age of onset, location, clinical characteristics and follow-up of this case are summarized in Table 1.

Table 1  
Literature review of intraspinal paraganglioma

References	Sex	Age(year)		Disease site	Benign / Malignant	Clinical symptoms	Surgery	Complication	Publishtime
		Onset	Diagnosis						
Zhang Qian ying et al. <sup>[3]</sup>	NM	25	25	L <sub>3-4</sub>	Benign	Backpain Radiation pain	NM	NM	2016
Han Songbo et al. <sup>[4]</sup>	M	< 54	59	L2-3	Benign	Backpain Radiation pain	TR	NM	2012
Zhou Guomin. <sup>[5]</sup>	M	< 31	31	T11-L1	Malignant	Backpain Numbness	TR	Paralysis	2017
Zhang zhao et al. <sup>[6]</sup>									
Case 1	F	< 41	42	L4	Benign	lumbago	TR	NM	2019
Case 2	M	35	35	L2-3	Benign	lumbago Numbness	TR	NM	2019
Zhang Lihua et al. <sup>[7]</sup>									
Case1	NM	NM	NM	T12-L1	Benign	lumbago Radiation pain	NM	NM	2007–2015
Case2	NM	NM	NM	L2	Benign	lumbago Radiation pain	NM	NM	2007–2015
Case3	NM	NM	NM	L3-4	Benign	lumbago Radiation pain	NM	NM	2007–2015
Case4	NM	NM	NM	L3	Benign	lumbago Radiation pain	NM	NM	2007–2015
Case5	NM	NM	NM	L3	Benign	lumbago Radiation pain	NM	NM	2007–2015
Case6	NM	NM	NM	L3	Benign	lumbago Radiation pain	NM	NM	2007–2015
Case7	NM	NM	NM	S1	Benign	lumbago Radiation pain	NM	NM	2007–2015
Case8	NM	NM	NM	T6	Malignant	lumbago Radiation pain	NM	NM	2007–2015
Case9	NM	NM	NM	T6-7	Metastasis	lumbago Radiation pain	NM	NM	2007–2015
Case10	NM	NM	NM	L2	Benign	lumbago Radiation pain	NM	NM	2007–2015
Case11	NM	NM	NM	S1-2	Metastasis	lumbago Radiation pain	NM	NM	2007–2015
Tang jianwei et al. <sup>[8]</sup>	M	< 49	51	L2-3	Benign	Numbness	TR	urinary incontinence	2016
Wang tieyan et al. <sup>[9]</sup>									
Case 1	F	< 29	29	L2-3	Benign	lumbago Mobility disorder	TR	No	2008

References	Sex	Age(year)		Disease site	Benign / Malignant	Clinical symptoms	Surgery	Complication	Publishtime
		Onset	Diagnosis						
Case 2	F	< 32	39	T12-L3	Benign	lumbago	TR	No	2013
Wang yin et al. <sup>[10]</sup>	F	< 50	54	L1	Benign	Lumbago Weakness	TR	NM	2005
Cheng anyuan et al. <sup>[11]</sup>	F	< 29	30	T12-L1	Benign	Lumbago Weakness	TR	No	2005
Dai zhehao et al. <sup>[12]</sup>	M	< 16	16	T6-7	Benign	Paraplegia	TR	NM	2006
Hu xueyi et al. <sup>[13]</sup>	F	< 50	51	L4-5	Benign	lumbago Radiation pain	TR	NM	2015
Chu Meihua et al. <sup>[14]</sup>	M	< 50	50	L3-4	Benign	lumbago	TR	NM	2014
Feng Yanqi et al. <sup>[15]</sup>	M	< 75	75	L2-3	Benign	Lumbago Numbness	TR	No	2013
Han yunfeng et al. <sup>[16]</sup>									2017
Case 1	F		43	L2-3				NM	
Case 2	F		62	T12-L3				NM	
Case 3	M		59	L2-3				NM	
Case 4	M		73	S1				NM	
Lu hongsheng et al. <sup>[17]</sup>									
Case 1	M		58	cauda equina		Pain		NM	2000–2004
Case 2	M		64	C4-6		Numbness		NM	2000–2004
Case 3	M		77	L3-4		Lumbago Mobility disorder hypertension		NM	2000–2004
Case 4	F		20	L1-2		Paraplegia		NM	2000–2004
Case 5	F		50	L1-2		Lumbago		NM	2000–2004
Ren qi et al. <sup>[18]</sup>									
Case 1	M		64	L2-3		Pain urinary incontinence		NM	2015
Case 2	M		50	L2		Lumbago		NM	2016
Wang Junhong et al. <sup>[19]</sup>	M		36	T2		Paraplegia	Biopsy	NM	1980
Zhao jian et al. <sup>[20]</sup>									

References	Sex	Age(year)		Disease site	Benign / Malignant	Clinical symptoms	Surgery	Complication	Publishtime
		Onset	Diagnosis						
Case 1	M		43	cauda equina		lumbago Radiation pain	TR	NM	2007
Case 2	F		49	L3-4		lumbago	TR	NM	2007
Wang ZH et al. <sup>[21]</sup>	M		36	L1		electrical pain in the waist and buttock		NM	2018
Gusmão MS et al. <sup>[22]</sup>	M		26	T10		Paraplegia		NM	2009
Alexander Reddy et al. <sup>[23]</sup>	F		34	T6-7		Paraplegia	TR	NM	2017
Shuzhong Liu et al. <sup>[24]</sup>	M		14	T4	malignant	elevated blood pressure and back pain	Partial resection	NM	2018
Alexander Richter et al. <sup>[25]</sup>	F		16	L1	malignant	cramp like pain in the right lower abdomen	Chemotherapy ,radiotherapy and resection	NM	2011
Słowiński J et al. <sup>[26]</sup>	F		46	L3	Benign	a right-sided lumboischialgia	NM	NM	2005
Houten JK et al. <sup>[27]</sup>	M		41	T1	malignant	Paraplegia	NM	NM	2002
Castel JP et al. <sup>[28]</sup>	M		68	cauda equina	Benign	urinary retention	NM	NM	1995

## 4 Discussion

Paragangliocytoma originates from sympathetic nerve sheath cells, 85%~90% occurs in the adrenal gland, about 10% occurs outside the adrenal gland, such as carotid body, jugular bulb, heart, retroperitoneum, bladder, hepatic portal, thoracic cavity<sup>[28-29]</sup>, Pick named pheochromocytoma that occurred in the adrenal gland in 1912, and named paraganglioma that occurred outside the adrenal gland. There are no accessory ganglia in the normal spinal canal and vertebral body, so the paraganglioma in the spinal canal and vertebral body is very rare, the incidence is about 0.07/1,000,000. The origin of paraganglioma in the spine has not been determined. Some scholars believe that it originated from the sympathetic cells of the lateral spinal cord of the thoracolumbar segment or the ectopic sympathetic trunk branches<sup>[30]</sup>, which mainly occurred in the dura mater of the spinal canal. It is rare to occur in the epidural. Paraganglioma can synthesize, store, and secrete catecholamines. Some tumors have neuroendocrine function. Few cases that occur in the spinal area are accompanied by neuroendocrine function. There are reports in the literature that may be associated with hypertension<sup>[31]</sup>, most of which are concentrated in the lumbosacral area, cauda equina, and terminal filaments in the spinal canal<sup>[32-33]</sup>, and non-functional tumors account for the vast majority. The prevalence of this disease is about 50 years old. Because most tumors have no functional lesions, non-specific symptoms caused by tumor compression are the first symptoms in clinical practice. Therefore, the misdiagnosis rate and missed diagnosis rate of this disease are extremely high.

MRI is currently the best examination method for the diagnosis of paraganglioma in the spinal canal<sup>[34]</sup>. It is mainly manifested as soft tissue masses with clear boundaries, T1WI and other signals, T2WI high signals, abundant internal nourishing blood vessels, and a few tumors have spotted hemorrhage. The tumor is obviously strengthened on the enhanced scan, and there are often thick "empty blood vessels" around it, and the enhanced scan shows vascular-like enhancement. Mainly differential diagnosis with neurogenic tumors in the spinal canal, epidermoid cysts in the spinal canal and other rare diseases<sup>[35-36]</sup>. Hemosiderin deposits can be seen at the edge of the tumor, which is helpful to prompt diagnosis. Hemosiderin deposits are mainly seen in vascular-rich lesions<sup>[37]</sup>. Preoperative MRI neuroimaging and 3D reconstruction in this patient have important guiding significance for the diagnosis of tumor, the formulation of surgical plan and the recovery of postoperative nerve function.

Pathological examination is the only way to diagnose paraganglioma. The tumor's main cells are uniform in size, round or almost round, with nuclei located in the center, chromatin punctiform, thinner, and nucleoli are usually inconspicuous. They are distributed in nests and look normal. Paraganglia: Sertoli cells arranged in a single layer around the nest, fusiform or cylindrical, sometimes difficult to identify under light microscope. The nested principal cell and surrounding supporting cells constitute a typical "Zellballen" structure<sup>[21]</sup>; Immunohistochemistry is mainly used to distinguish it from other neuroendocrine tumors. Chromaffin A (CgA), synapse protein (Syn), neuron-specific enolase (NSE), S-100 protein, glial fibrillary acidic protein (GFAP) and keratin (CK), etc. To a certain degree of diagnostic significance, the expression of Syn, CgA, and NSE among these indicators in this patient was positive.

Total surgical resection is the first choice for treatment of paraganglioma in the spinal canal, and most patients have a good prognosis. The surgery uses the posterior median approach of the vertebral body segment where the tumor is located, and the lamina and dura must be opened. Paraganglioma is located in the subdural epimedullary, mostly closely related to nerve tissue, and has abundant blood supply<sup>[38]</sup>. Paraganglioma has a rich blood supply and shares a large and deformed blood supply vessel with the conus medullaris. Disconnection of this blood vessel may cause abnormal blood supply to the conus medulla<sup>[42]</sup>. It is often seen in literature reports that patients have complications such as urinary incontinence after surgery. The possible reasons are not only the damage of the cauda equina during the operation, but also the influence of the blood supply of the conus medullarius after the vessel is broken. Therefore, the blood supply artery should be blocked during surgical resection. In order to reduce bleeding and reduce the tumor volume; the priority is to separate the tumor from the adhering nerve roots, if the nerve roots cannot be separated, then perform a total resection; then strictly stop the bleeding, close the dura mater, and fix the lamina.

Paraganglioma in the spinal canal is a benign tumor, but there are still a few reports of malignant metastasis of the tumor after surgery<sup>[39]</sup>. Surgery may have a curative effect, but the spread of the tumor limits the chance of radical resection<sup>[41]</sup>. A small number of tumors that cannot be completely removed may have cerebrospinal fluid dissemination and metastasis. For patients whose tumors are not completely removed, reoperation and combined treatment with radiotherapy and chemotherapy are feasible<sup>[33]</sup>. According to reports, there are still a few reports that pathological examinations cannot determine the benign and malignant tumors, so long-term follow-up is very important for the prognosis of patients<sup>[40]</sup>. This patient underwent complete tumor resection during the operation. Thanks to preoperative neuroimaging and 3D reconstruction technology, no important nerves were injured during the operation. The patient's neurological function recovered well after the operation. The postoperative patient needs to review B-ultrasound, DR, CT, etc. regularly. To better assess the local recurrence. The patient has been followed up for 1 year and 7 months with good internal fixation and no signs of tumor recurrence and metastasis. The follow-up is still ongoing.

Although neuroendocrine markers such as CgA, Syn, S-100 protein, NSE positive and epithelial-derived markers such as CK, EMA and other negative are helpful to distinguish ependymoma, schwannoma, meningioma, chordoma, etc. Pathological examination is still difficult to judge the benign and malignant tumors. It should be judged according to its biological behavior. If lymph node metastasis or/and distant metastasis occurs, it can be considered as malignant paraganglioma. Therefore, long-term follow-up is of great significance in determining the prognosis of patients.

## 5 Conclusion

Paraganglioma of the spine is a subtype of neuroendocrine tumors and is clinically rare. Because most tumors have no functional lesions, non-specific symptoms caused by tumor compression are the first symptoms in clinical practice. Therefore, the misdiagnosis rate and missed diagnosis rate of this disease are extremely high. This patient was found in a physical examination. After admission, he was actively improved relevant examinations, performed MRI neuroimaging and 3D printing reconstruction technology, formulated a complete surgical treatment plan before the operation, carefully protected the nerves during the operation, and the patient's tumor was completely removed during the operation, Postoperative neurological function recovered well, and no tumor recurrence was found in follow-up examination.

## Abbreviations

F  
female; M: male; NM: not mentioned; TR: total resection

## Declarations

### 6 Informed Consent Publishing

The patient and family members agreed that the case was reported and signed an informed consent form. The scanned copy of the informed consent form can be sent to the publishing house.

## 8 Conflict of interest

The authors declare that they have no competing interests.

## 9 Author' contributions

Ya Zhang and Dongqi Li ,Zunxian Tan collected the data of the case, reviewed the literature, and drafted the manuscripts. Lijuan Ye,Kecheng Li, Qiuyun chen andYan liu help to collect the data of the case and pathological pictures as well as modifying the manuscripts. Zuozhang Yang, Cao Wang, and Yihao Yang carried out the operation. Zuozhang Yang and Tiying Wang revised the manuscript. Kun Li and Xinchao Yu,Yanan Zhu,Jiaxiang Chen collected the Imaging data. All authors read and approved the final manuscript.

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## 11 Availability of data and materials

Data will be available upon request to the first author Ya Zhang.

## 12 Ethical approval

Ethical approval was not involved in this study.

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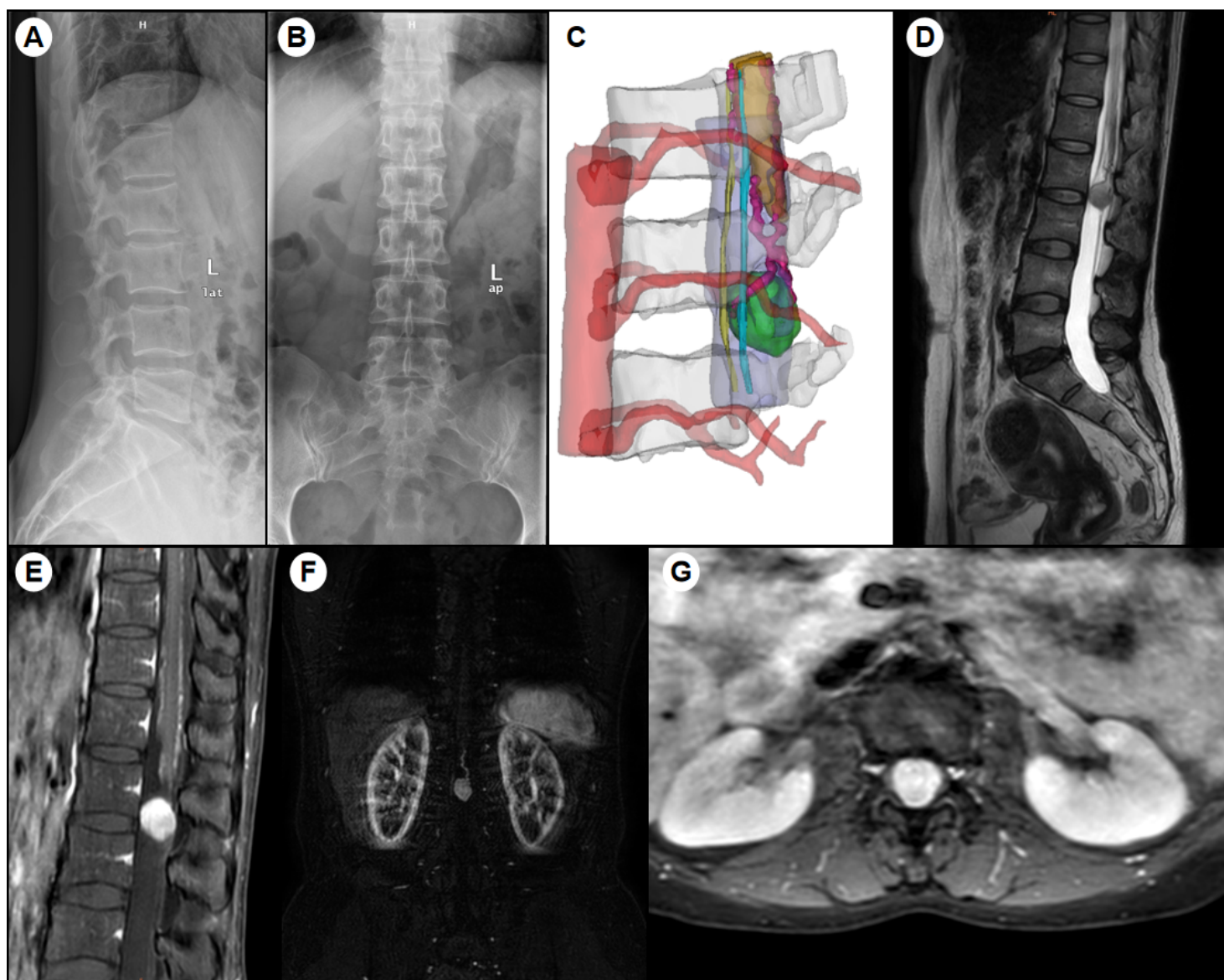
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## Figures



**Figure 1**

A, B: preoperative X-ray of the patient; C: 3D printing three-dimensional reconstruction showing the relationship between the tumor and the surrounding spinal cord and blood vessels; D, E, F: magnetic resonance neuroimaging showing the spinal canal stiffness at the 2/3 level of the waist The submembrane nodules show long T1 and long T2 signals, with clear borders, and seem to be connected to the terminal

filaments. After enhancement, they are uniformly strengthened, and the spinal artery can be seen to participate in the blood supply of the tumor. ¶1¶Preoperative DR, CT, MRI;

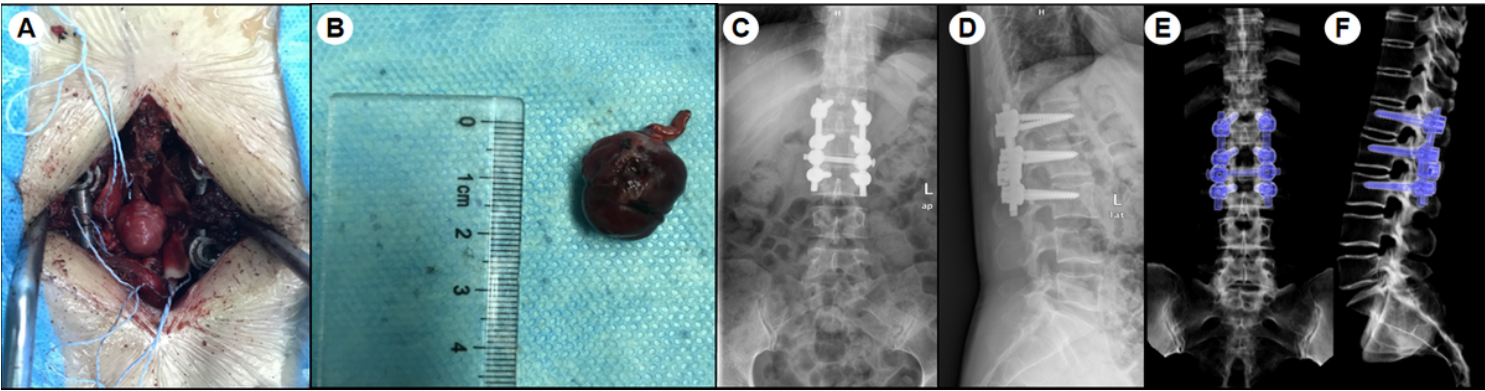


Figure 2

A: Intraoperative exploration revealed that the tumor capsule is intact, dark red, smooth surface, the tumor is connected to the terminal filament, and the superficial part of the ventral side of the tumor is slightly adhered to the cauda equina nerve; B: the gross tumor specimen is removed; C, D: X-ray review after operation; E, F: CT image review after operation. ¶2¶Intraoperative findings and postoperative DR, CT;

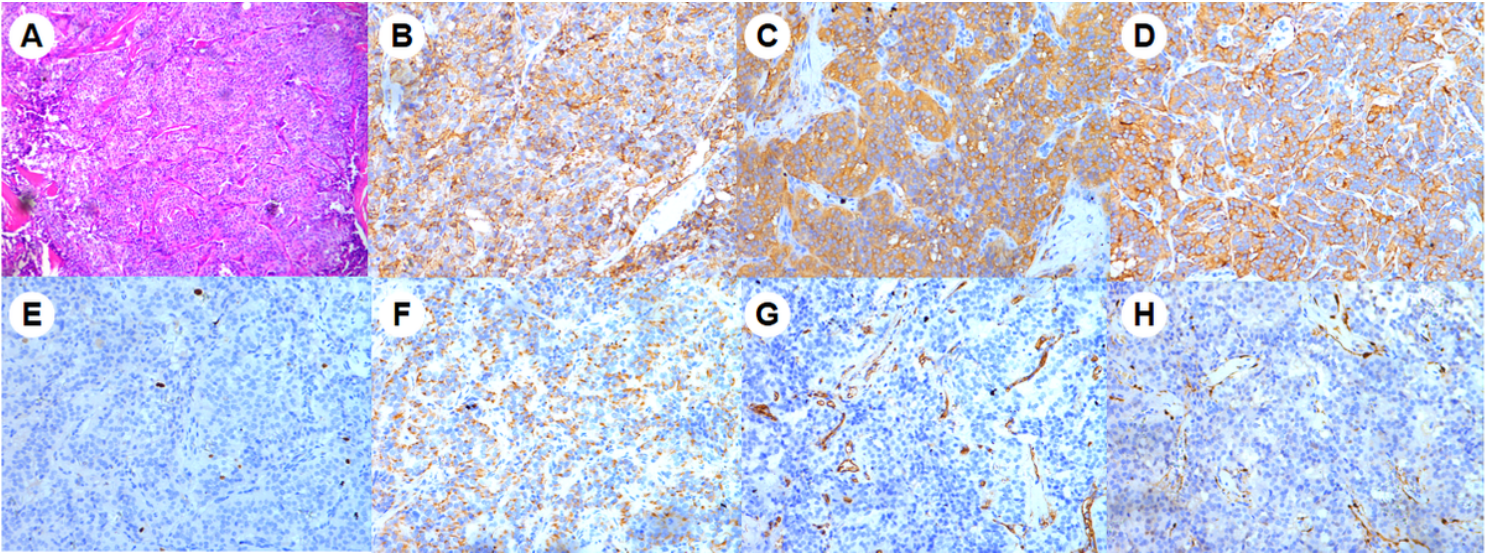


Figure 3

A: HE staining showed that the epithelioid cells constitute the tumor parenchyma, the cell morphology is uniform, the cytoplasm is more, the nucleolus is not obvious, the mitoses are rare, and the arrangement is acinar; the interstitial is sinusoidal and a little fibrous; B: CgA immunohistochemical results showed strong positive; C: NSE immunohistochemical results showed strong positive; D: Syn immunohistochemical results showed strong positive; E: Ki67 immunohistochemical expression was positive (about 5%) ; F: CK immunohistochemistry result showed negative; G: CD34 immunohistochemistry result showed negative; H: S-100 immunohistochemistry result showed negative. ¶3¶Postoperative pathological pictures