



# A rare case of dumbbell shaped ancient schwannoma found inside thoracic spine in a psychologically challenged patient

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**Introduction and importance:** Ancient schwannoma undergoes a degenerative process leading to hemorrhage, cystic necrosis, and calcification. The head and neck, thorax, and retroperitoneum are the most common locations; the spinal location is extraordinary.

Case presentation: A 50-year-old man presented with muscle weakness in his lower limbs following 2 years of lower back pain, tingling sensation and numbness. He suffers from moderate depression that was refractory to treatment and was put on selective serotonin reuptake inhibitor. MRI showed an intradural extramedullary dumbbell-shaped mass at the T11–T12 level with compression of the spinal cord. Furthermore, it was located on the right side of the spinal canal in close relation to the right T12 spinal root. He underwent a T11–T12 laminectomy with complete en bloc resection and the nerve root was sacrificed. Histopathological exam confirmed the final diagnosis. Follow-up showed a rapid improvement of the neurological and psychiatric symptoms.

Clinical discussion: Shwannomas are slow growing tumors arising from Schwann cells of the peripheral nerve sheaths. Spinal ancient schwannomas are particularly rare because of the compression of the spinal cord leading to the early presentation of symptoms. The risk of developing depressive disorders is increased by physical diseases; moreover, chronic back pain is strongly associated with depression.

**Conclusion:** Ancient schwannoma is a slow-growing tumor and best pre-operatively diagnosed by MRI. Diagnostic approach in patient with psychiatric disorders presenting with physical symptoms, especially neurological symptoms should be done carefully depending on objective tools such as imaging or detailed neurological examination.

Keywords: ancient schwannoma, case report, depression, MRI, schwannoma, spinal cord

#### **Background**

Schwannoma is a non-cancerous tumor arising from Schwann cells<sup>[1]</sup>, making up about one-third of all primary spinal cord tumors<sup>[2]</sup>. Ancient schwannoma is believed to undergo a long-term retrogressive process which includes: hemorrhage, cystic necrosis, and calcification. The exact incidence rate of ancient schwannoma remains uncertain, The head and neck, thorax, and retroperitoneum are among the most common sites for

### HIGHLIGHTS

- The location and shape of the tumor.
- The relation with back pain.
- The psychiatric condition of the patient the led to neglecting his complains.
- The patient's mental health status improvement following the surgery.

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ancient schwannoma; spinal location of ancient schwannoma is extraordinary, with only few reported cases in the literature, and back pain is the usual complain<sup>[2,3]</sup>. Herein, we are presenting a rare case of intradural-extramedullary ancient schwannoma of the thoracic spine in an 50-year-old patient with major depressive disorder.

#### **Case presentation**

A 50-year-old man from Arab-ethnicity background presented to the general practitioner (GP) with lower limb weakness which had been progressing for the past 3 months. The patient had also been complaining of numbness, pain, and a tingling sensation in his legs, in addition to a dull pain in his lower back for the previous 2 years. The patient was diagnosed with moderate depression that was refractory to treatment with a Patient Health Questionnaire (PHQ-9) score fluctuating between 9 and 11 for several follow-up sessions. Sleep difficulty,

concentrating problem, restless feelings, and low energy was also mentioned; therefore, he was put in selective serotonin reuptake inhibitor. Laboratory values were all within the normal range including: CBC, kidney function, blood fasting glucose, CRP, and liver function. There was no history of trauma and the patient had no significant medical history apart from his depression.

MRI revealed a mass at the T11–T12 level. The neurosurgeon performed a detailed neurological exam; muscle power was estimated to be 4/5 on the right side upon testing hip movements (flexion, extension, abduction, adduction) and also muscle power on knee flexion and extension and on ankle plantarflexion and dorsiflexion was slightly impaired and estimated to be 4/ 5. On the other hand, testing the muscle power of the same movements on the left lower limb revealed a mild weakness and was estimated to be about 4+/5. The neurosurgeon examined carefully his patellar and ankle reflexes on both limbs and his deep tendon reflexes were estimated to be normal. Of course, the physician also tested the muscle power and deep tendon reflexes in the upper limbs but they exhibited no abnormalities. Moreover, the patient was suffering from a dull, aching pain in his lower back (3/10 on Visual Analogue Scale) which was also radiating down to his right limb and was therefore presumed to be radicular in nature. Impaired sensation was also demonstrated below the T12 dermatome level as fine touch, vibratory, and pinprick sensation were completely absent. There were no complaints relating to the upper limbs, bowel and bladder function were also intact. The neurosurgeon proceeded to study the MRI which showed an intradural extramedullary dumbbellshaped mass at the T11–T12 level with marked compression of the spinal cord (Fig. 1A). Furthermore, the mass demonstrated homogeneous low-intensity signal on T1-weighted and high signal intensity on T2-weighted images in addition to marked peripheral enhancement following administration of gadolinium (Fig. 1B and C). The mass was located on the right side of the spinal canal in close relation to the right T12 spinal root. Furthermore, the tumor consisted of solid and cystic compounds (Fig. 2A and B).

Based on the clinical and radiological findings, surgery was decided and the patient underwent a T11–T12 laminectomy with complete en bloc resection of the tumor, it was performed by the guarantor of this work, the tumor appeared to be cystic in nature and was firmly attached to the adjacent nerve root, therefore, the nerve root was sacrificed. Microscopic H&E staining revealed a spindle-cell tumor with alternating hypercellular (Antoni A) and hypocellular (Antoni B) regions, areas of cystic degeneration, areas of hemorrhage, hemosiderin deposit, nuclear palisading, and hyalinized vessels' walls were also visualized (Fig. 3). Nuclear atypia and mitotic figures were absent. Based on these histologic findings, a diagnosis of ancient schwannoma was made.

The patient experienced excellent recovery following the surgery and his neurological symptoms improved rapidly. On a 3-month follow-up the patient had no detectable neurologic deficits and he had also regained muscle strength in his legs. Moreover, the patient reported a significant improvement in his mental state as the symptoms of his depression had diminished following his recovery from surgery. The patient was advised to

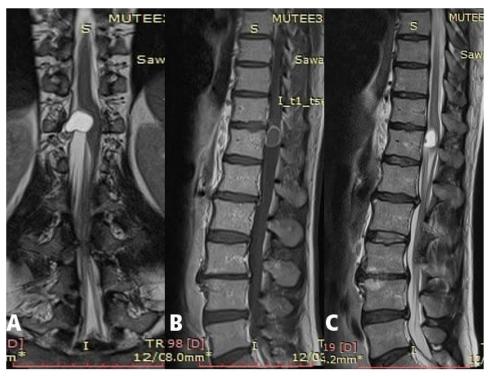


Figure 1. (A) Coronal T2-weighted image reveals a hyper-intense, intradural-extramedullary tumor at the T12 level with marked compression and displacement of the spinal cord to the left side. (B) Sagittal T1-weighted image with Gadolinium contrast, demonstrating rim enhancement of the lesion. (C) Sagittal T2-weighted image with contrast showing high signal intensity.

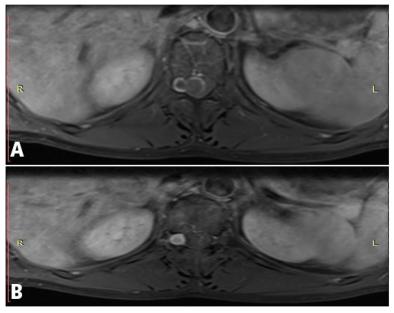


Figure 2. (A) Axial T1-weighted image with contrast showing the cystic compound and dumbbell shape of the tumor. (B) Axial T1-weighted image with contrast showing the solid compound of the tumor.

follow up with his psychiatrist to further evaluate his mental condition. One-year follow-up MRI showed no recurrence or any residuals of the original mass (Fig. 4). Moreover, significant improvement in his mental condition was reported with a PHQ-9 score of 5; activity level and sleep quality had also back to normal.

This case has been reported in line with SCARE 2023 guidelines<sup>[4]</sup>.

#### **Discussion**

Shwannomas (also called neurilemmomas) are non-cancerous, slow growing tumors arising from Schwann cells of the peripheral nerve sheaths<sup>[3]</sup>. Histologically, they are encapsulated

tumors arranged in different cell proportions, making multiple arrays of special tissue indicated as Antoni-A and Antoni-B. Antoni-A region is characterized by a dense accumulation of spindle-shaped cells, in comparison with Antoni-B region which display hypocellularity with a loose myxoid matrix<sup>[3,5]</sup>. Ancient schwannomas, originally mentioned by Ackerman and Taylor in 1951<sup>[6]</sup>, are rare variant of schwannomas, comprising about 0.8% of soft tissue tumors<sup>[5]</sup>. The term "ancient" is used for demonstrating its degenerative and cystic changes, including hemorrhage, calcification, decreased connective tissue cellularity (relative loss of Antoni-A tissue), and degenerative nuclei<sup>[2,3]</sup>. These degenerative features are believed to be the consequence of its long-term slow-growing course<sup>[3]</sup>. Microscopic examination of the tumor revealed spindle cells, round cells with hemorrhage, hyalinization, multiple cystic formations, and nuclear

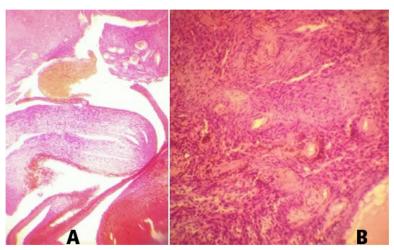


Figure 3. (A) Histopathological findings: areas of cystic degeneration, hemorrhage and hemosiderin deposit (H&E stain, × 10). (B) Histopathological findings: spindle-cell tumor with hypercellular (Antoni A) and hypocellular (Antoni B) regions, nuclear palisading and vessels with hyalinized walls (H&E stain, × 20).



Figure 4. Sagittal T2-weighted image at 1-year follow-up showing no recurrence or residuals of the tumor.

palisading. Ancient schwannoma is predominantly located in the head and neck region, retroperitoneum, pelvis, and thorax, manifesting as a soft tissue mass usually with elderly patients<sup>[5]</sup>. Spinal ancient schwannomas are particularly rare most likely because of the compression of the spinal cord leading to the early presentation of symptoms in case of spinal schwannoma. It means the patient is usually hospitalized and the tumor excised before it can progress to ancient schwannoma at the T11–T12 level which is considered such a rare position for ancient schwannoma. To the best of our knowledge, there are only a few cases in the literature. Omichi *et al* summarized 10 cases in their literature review<sup>[3]</sup>.

The masses of the spine are divided into two branches, primary and secondary. In regard of primary masses, non-malignant lesions are seen more frequently than malignant ones. The differential diagnosis includes, but not limited to osteoid osteoma, osteoichondroma, and hemangoima<sup>[7]</sup>. Other intradural-extramedullary tumors include spinal meningioma and neurofibroma<sup>[8]</sup>. Based on imaging findings, ancient schwannomas can be misdiagnosed or confused with sarcomas and other

soft tissue tumors such as the following: Ewing's sarcoma that usually presents with lytic effect on the vertebral body; Meningioma which locates preferably lateral to the spine<sup>[2,7,8]</sup>. Despite the degenerative changes, these tumors behave in a similar way to their conventional counterparts and considered benign tumors with a rare malignant transformation<sup>[5]</sup>. Isobe et al explored in their study the clinical and radiologic features of ancient schwannoma in seven patients. The enhancement at a peri-degenerative area and sometimes at a capsule on CT and MRI was a significant differentiating radiologic characteristic from typical schwannoma patterns<sup>[9]</sup>. Hayashi et al also reported in their case on calcification in ancient schwannoma<sup>[10]</sup>. The radiologic presentation of our case mostly confirmed with the aforementioned typical characteristics of schwannoma and its ancient variant. While not showing any calcifications, the peripheral rim-enhancing pattern was clearly visible.

The major part of vertebral column tumors is secondary to internal organs' malignancy. However, both malignant and benign lesions of the spine account for a tiny percentage of the causes of backache<sup>[7]</sup>. Pain, which can be localized or radicular, is the most common symptom for ancient schwannoma in the spinal cord. However, they may produce vague symptoms considering its degenerative and slow-growing features<sup>[3,5]</sup>. Back pain requires extended and full neurological scanning to determine any features of myelopathy or reflexes abnormalities<sup>[11]</sup>. The patient in our case has been complaining of back pain with a tingling sensation and numbness in his legs for 2 years before the muscle weakness was developed. Notably, there were no abnormal reflexes. These features were read properly by the GP who decided to further investigate and discovered a spinal mass on MRI which led him to refer the patient to a neurosurgeon.

Medical history is a very important step in patients complaining of physical illness but their complaints were neglected due to their psychiatric history<sup>[12]</sup>. In our case the family of the patient intentionally neglected the patient complaint of back pain and feeling of discomfort for 2 years attributed to his psychiatric condition.

We conducted a literature review trying to identify related cases and articles. We searched PubMed and Google Scholar using keywords, after that we filtered the results by two-step screening: title and abstract, and full text screening. The literature showed that complete surgical resection of the tumor is the preferred treatment in case of spinal schwannoma. Moreover, several series reported recurrence of the tumor in case of subtotal resection<sup>[13]</sup>. Although sparing of the involved nerve is generally attempted in cases of spinal nerve schwannoma. Sometimes, the total resection of the tumor is unachievable without sacrificing the nerve root<sup>[14]</sup>. According to Ohnishi et al, only 3 among total of 15 patients who underwent surgical resection of nerve root had neurological deficits including feeling of numbness and weakened motor movements. Furthermore, Kim et al found that neurological deficits tend to be limited loss of muscles power and sensory deficits. There is a risk of neurological impairment following resection of the nerve root, even though the spinal roots giving origin to the schwannoma are mostly thought to be nonfunctional. Preservation of the root is advised, especially if the nerves involved are located at a critical level like the cervical or lumbosacral segments. However, postoperative neurological deficits do not always occur even when sacrificing roots in these critical segments<sup>[14,15]</sup>. In our case, the right T12 nerve was sacrificed but no neurological deficits were identified following the surgery.

Conti *et al* have reported in their series that the rate of improvement following surgery is better for tumors located in the dorsal tract compared to the cervical or lumbosacral tract<sup>[13]</sup>. In our case, the patient's tumor was located in the lower dorsal region and he experienced a full and speedy recovery following surgery. There is a risk of spinal deformity following multilevel laminectomy. Yasuoka *et al* reported that the risk is higher in younger patients and the more cephalad the laminectomy<sup>[16]</sup>. We searched and reviewed the literature in order to enhance our understanding of this case. Table 1 shows a literature review of related papers including ours as a latest updated on the literature.

It is well-known that the risk of developing serious depressive disorders is increased by physical diseases. Both discomfort and chronic pain are listed as common factors associated with depression<sup>[17]</sup>. Furthermore, the relationship between depression and pain is not that simple. It reflects a deep biological relation as both norepinephrine and serotonin affect pain and mood<sup>[18]</sup>. Moreover, chronic back pain is strongly associated with depression<sup>[19]</sup>. According to Merker et al, 62% of the patients with diagnosed schwannoma had to take drugs for chronic pain and 30% of them were clinically diagnosed with depression<sup>[20]</sup>. Another study found that pain and symptoms of depression were significantly correlated in patient diagnosed with schwannoma<sup>[21]</sup>. In our case, the point-of-start of depression is not well specified. However, it's believed that his complaints including backache among tingling sensation and numbness had increased the severity of his mental condition. Spinal surgery might increase or decrease the severity of psychiatric condition. Those who feel better after spinal surgery might be subject to preoperative depression caused by their spinal disease<sup>[22]</sup>. The patient had depression at least for the last 2 years accompanied with spinal ancient schwannoma, which in turn increased the depressive status of the patient due to back pain and other neurological symptoms. Furthermore, the family report of improvement of his psychiatric symptoms after the surgery should be considered as a piece of evidence on the link between his physical and psychiatric conditions.

Wen et al<sup>[3]</sup> also reported ancient schwannoma in a schizophrenic patient which is considered a psychological disorder and highlighted the probability of physical illnesses in

patients with psychiatric disorders. Moreover, doctors should avoid distraction by the patient's presenting complaint of feeling anxious, depressed, or changes in personality<sup>[12]</sup>. Doctors should let the patient describe his complaints by his own words as family members may underestimate his complaints due to his psychiatric disorder, leading to a delayed diagnosis of his organic illness.

#### Conclusion

Ancient schwannoma is a slow-growing tumor, and the best preoperative diagnostic measure is MRI. Diagnostic approach in patient with psychiatric disorders presenting with physical symptoms and especially neurological symptoms should be done carefully with the help of objective tools such as imaging or detailed neurological examination to avoid misdiagnosing the underlying organic disease as chronic back pain is strongly associated with depression.

#### **Ethical approval**

None.

#### Consent

Written informed consent was obtained from the patient for publication and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

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None.

#### **Author's contribution**

All authors approved the final version of the manuscript. A.N. led the team and wrote the final draft. B.S., A.I., I.S., and G.D. equally participated in data collection and writing the introduction, clinical presentation, and discussion. I.S. supervised the work of the team, critical revision, and patient care.

Table 1

#### Review of literature on spinal ancient schwannoma

Study ID	Sex/age (years)	Location/spinal level	Clinical outcome
Antonopoulos 2009	Male/42	The posterior side of the left psoas muscle/L4–L5	Nerve root was completely excised. The patients discharged in good condition; however, they suffered sensory impairment on the lateral side of his left femur
Wen 2016	Male/58	Intradural extramedullary/T9-T10	Nerve root was completely excised. There were no detectable sensory impairment or movement limitations.
Kumar 2017 <sup>[23]</sup>	Male/60	Intradural extramedullary/T4-T5	The attached nerve root was preserved. No recurrence was detected in the follow-up that lasted for 2 years and 9 months.
Omichi 2021	Male/71	Extradural/T1-T2	Wound infection 3 weeks after surgery. In the follow-up 1 year later the patient was completely recovered with no recurrence.
	Male/58	Extradural/C1–C2	Seven-year follow-up showed small degree of difficulty coordinating the movement of his left hand and sensory impairment.
Bozyigit 2022 <sup>[24]</sup>	Female/66	Spinal canal/T8-T9	There were no postoperative complications.
Our case	Male/50	Intradural extramedullary/T11–T12	The attached nerve root was excised. The patient recovered quickly after 3 months and he had no recurrence on 1-year follow-up.

#### **Conflicts of interest disclosure**

The authors declare no conflict of interest.

## Research registration unique identifying number (UIN)

None.

#### Guarantor

Dr. Issam Salman.

#### Provenance and peer review

The paper is not commissioned, externally peer-reviewed.

#### **Data availability statement**

The datasets generated and/or analyzed during the current study are not publicly available due to the study design (case-report) as there is no datasets generated or analyzed. However, further data and information on the patient condition are available from the corresponding author on reasonable request.

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