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CASE REPORT

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Cerebrospinal fluid leakage complicated by intracranial hematoma and cervical infection following resection of dumbbell schwannoma in the cervical canal: a case report and literature review

Zhen-Shan Yuan[1*], Lian-Song Lu[1] and Yong Hu[1]

Abstract

Background Haemorrhagic cerebral infarction with cervical infection is a severe and complex complication in spinal surgery. This paper report a case of intracranial hematoma secondary to cerebrospinal fluid leakage after surgery for dumbbell tumor of cervical spine.

Case presentations The patient suffered from postoperative cerebrospinal fluid leakage followed by hemorrhagic cerebral infarction, unilateral limb sensorimotor dysfunction and language dysfunction, and was treated conservatively. Combined with cervical infection, the treatment was anti-inflammatory and lumbar puncture drainage. The infection of the patient was cured, and the symptoms related to hemorrhagic cerebral infarction were better than before.

Results The case showed cerebrospinal fluid leakage after the operation of dumbbell tumor of cervical spine, which caused hemorrhagic cerebral infarction with typical clinical symptoms.

Conclusions The risk complications of intracranial hemorrhage and cervical infection should be paid attention to when cerebrospinal fluid loss occurs in patients with cervical dumbbell tumor after surgery.

Keywords Dumbbell tumors, Cerebrospinal fluid leakage, Hematoma, And schwannoma

*Correspondence:
Zhen-Shan Yuan
yuanzhenshan2011@hotmail.com
[1]Department of Spinal Surgery, Ningbo No.6 Hospital, NingBo 315040, Zhejiang Province, People's Republic of China

Background

Dumbbell tumors of the cervical spinal canal are a special type of cervical spinal canal tumor. They are more complex and risky than simple cervical spinal canal tumors and have a higher residual and local recurrence rate after surgery. Therefore, treating these tumors is challenging in spinal surgery and neurosurgery. After the resection of a large tumor, the dura mater defect often becomes large that easily leads to a large loss of cerebrospinal fluid, decreased intracranial pressure, and reduced ventricular system and brain volume. Furthermore, brain tissue declines because of the pressure gradient, leading to pulling and damage of intracranial blood vessels and hematoma formation [1, 2]. Few cases of cerebral infarction and intracranial infection after the resection of dumbbell-shaped cervical tumors have been reported. This report aimed to present a rare case of cervical spinal schwannoma complicated by acute intracranial hematoma and cervical infection, which was treated at our hospital.

Case report

A 51-year-old woman was admitted to our hospital on August 6, 2013, for distending pain in the left side of her neck for 1 year, which had worsened 20 days before admission. Preoperative imaging revealed a tumor inside and outside the cervical spinal canal that penetrated the intervertebral foramen and extended to the adjacent paravertebral soft tissue. The lesion involved two intervertebral foramen. Fig. 1). Routine examinations before surgery, including electrocardiography, revealed no obvious abnormalities in coagulation function or biochemical series. The patient was admitted to hospital due to progressive aggravation of the condition, and a limited-time operation was planned. No clinical symptoms related to cerebral hemorrhage were found in the preoperative

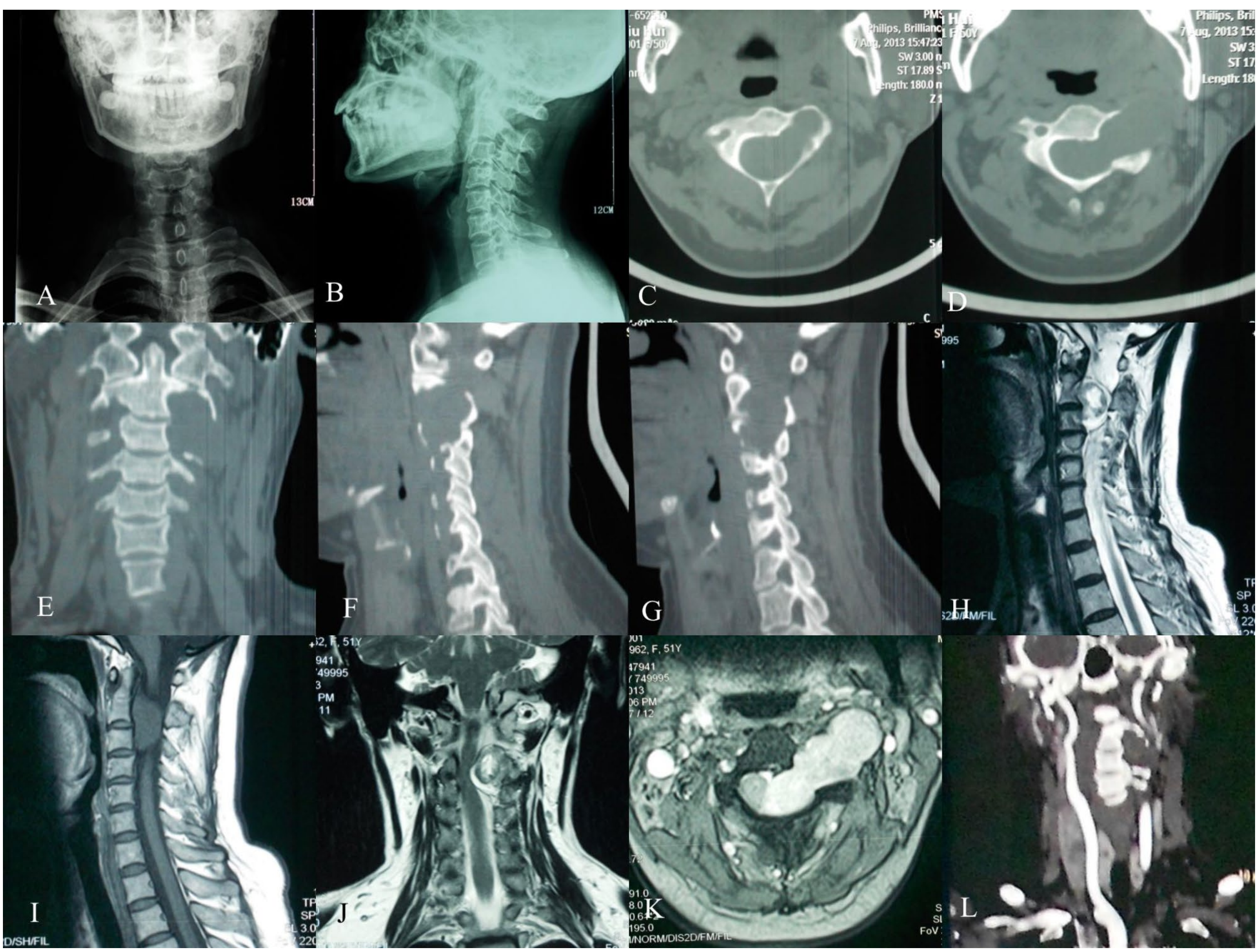

Fig. 1 Preoperative imaging examination: **A**-**B** preoperative cervical anterior and lateral X-ray films; **C**-**G** preoperative CT showed that space occupying in spinal canal and left anterior bone destruction could be seen at cervical 2 level; The sagittal plane of MRI before **H**-**K** showed that space occupying in spinal canal could be seen at cervical 2–3 level, and it was dumbbell-shaped in cross section; **L** Preoperative CTA showed that the left vertebral artery was obviously compressed by tumor

physical examination evaluation, so the preoperative head MRI examination was not completed. The study was approved by the ethics committee of Ningbo No.6 Hospital. The number: CHEC2009-066.

C2 and C3 hemilaminectomy and partial lateral mass resection were performed using the posterior and left lateral approaches of the cervical spine. Removal of the tumors from inside and outside the spinal canal and internal fixation using C1 bilateral pedicle, C2 right pedicle, and C3 bilateral lateral mass screws were performed under general anesthesia in the prone position. The posterior arch, C2–C4 spinous process, lamina, and lateral mass of the atlas were routinely exposed through a posterior cervical midline incision. Based on preoperative magnetic resonance imaging (MRI) showing the involved area of the tumor, the lamina and lateral mass of the affected side of the C2 and C3 segments were removed, and the dural sac and nerve root of the affected side were fully exposed.

During surgery, a dark red oval tumor with a diameter of ~ 1.5 cm was found in the left front of the spinal cord, with a complete capsule, adhesion of the arachnoid membrane and left C2 and C3 nerve roots, and poor mobility. The tumor was carefully peeled off using a 2.5× surgical magnifying glass, and double-click electrocoagulation was performed to stop bleeding. Attention was paid to protect the cervical spinal cord and nerve roots. The tumor was separated completely, and attention was paid to protect the vertebral artery and vein when separating the tumor at the medial wall and outlet of the intervertebral foramen. Because the tumor was completely resected and the dura mater defect was large and difficult to repair, one drainage tube was indwelled. The operation time was 4 h, the volume of blood lost was 2,000 ml, the volume of autologous blood transfused was 300 ml, the volume of allogeneic red blood cells transfused was 4.5 U, and the volume of plasma transfused was 540 ml. During surgery, blood pressure and heart rate was maintained at ~ 110/70 mmHg and 85 beats/min, respectively, and the rhythm was uniform.

The patient developed severe headache and vomiting 12 h after surgery. Approximately 300 ml of light bloody fluid was drained from the neck using negative pressure. Blood pressure and heart rate at this time was 150/70 mmHg and 78 beats/min, respectively. Considering cerebrospinal fluid leakage and intracranial low pressure, mannitol (125 ml) was administered to relieve brain edema. Consequently, head computed tomography (CT) was urgently performed, which revealed a low-density focus in the right temporal lobe (Fig. 2A). The patient's headache symptoms were relieved, and aspirin was administered to prevent cerebral infarction.

On day 3 after the operation at 2:00 pm, the patient developed progressive right hemiplegia with aphasia and blurred consciousness. CT and MRI revealed hemorrhagic cerebral infarction in the left basal ganglia (Fig. 2B–D). The estimated hemorrhage volume based on CT was 6–7 ml, with no evidence of brain surgery. Aspirin was discontinued, and nutritional support was provided. Subsequently, nerve function improved. To promote recovery, symptomatic treatment and rehabilitation exercises were performed. On day 5 after the surgery, the patient's conscious language function gradually recovered, limb function recovered generally, and right muscle strength was recorded at grade 3. Currently, the patient required assistance from family members to perform certain activities of daily living.

From days 2 to 13 postsurgery, the patient experienced persistent fever, excluding the possibility of infection in the lung, urinary system, and other systems. The cerebrospinal fluid was turbid, and the initial pressure was 110/20 mmHg. Routine cerebrospinal fluid test revealed 105 × 106/L of nucleated cells, with 65% lymphocytes, 33% neutral cells, and 2% mononuclear giant cells. The cerebrospinal fluid analysis revealed a glucose level of 4.13 mmol/L, lactate dehydrogenase level of 99 μ/L, high-sensitivity C-reactive protein level of 0.8 mg/L, chlorine

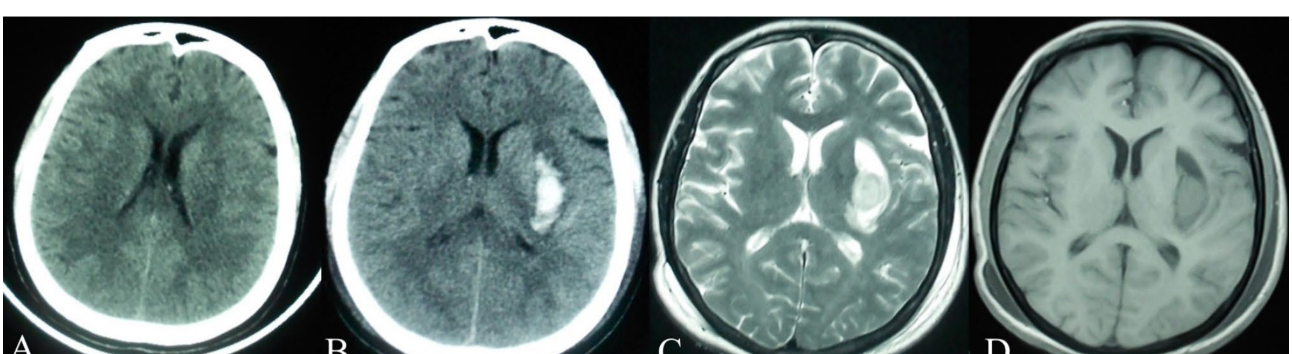

Fig. 2 The right temporal lobe-shaped low-density focus (**A**) on the first postoperative head CT; The second CT and MRI (**B**-**D**) showed hemorrhagic cerebral infarction in left basal ganglia

level of 114 mmol/L, and cerebrospinal fluid protein level of 70.6 mg/dL.

Meropenem, an antibiotic that easily passes through the blood–brain barrier, was administered on day 5 after the surgery for an internal infection. The patient continued to experience a higher body temperature (37.6 ℃–39.1 ℃). On day 13 after postsurgery, the patient was treated with continuous drainage of cerebrospinal fluid in the lumbar cistern combined with meropenem and vancomycin. On day 23 postsurgery, the patient's body temperature recovered to normal levels, and their cerebrospinal fluid was tested consecutively thrice with all the results being negative. Consequently, the lumbar cistern drainage tube was removed. No clear bacteria were detected in this patient, which may be related to the use of antibiotics before and after surgery, which interfered with the results of blood culture and secretion culture. However, cerebrospinal fluid examination results showed that cerebrospinal fluid leakage combined with infection was more likely. Infection may occur in the cervical spine, but due to the large dural defect, ascending bacterial infection to the intracranial cannot be ruled out. Therefore, lumbocisternal drainage is recommended in conjunction with antibiotic treatment. Anterior and lateral X-ray films of cervical spine were reexamined and they showed that the posterior cervical internal fixation was in place (Fig. 3A–B). After the surgery, pathological examination confirmed a schwannoma (Fig. 3C). Cervical MRI showed that the tumor had been completely removed, but a large area of cerebrospinal fluid leakage remained (Fig. 3D–I). One year after the surgery, X-ray and MRI of the cervical spine were performed, and they revealed that the internal fixation was in place, tumor was absent, and cerebrospinal fluid was absorbed into the subcutaneous tissue (Fig. 4A–F). Head MRI revealed chronic intracranial hematoma in the left basal ganglia (Fig. 4G–H). The patient still complained of occasional dizziness and other sequela, and her right muscle strength recovered to grade 4–5.

Discussion

The adjacent anatomical structures of dumbbell tumors inside and outside the cervical spinal canal are complex. Therefore, most surgeons can only resect tumors in the spinal canal or a part of the tumors in the intervertebral foramen, which leads to patients often living with tumors, easy recurrence, or accelerated malignancy [3]. It is extremely difficult and risky to completely remove tumors under the naked eye, which requires the surgeon to have superb and skilled surgical skills and experience. In our case, although the tumor was completely removed, the dura mater defect was too large to repair, so cerebrospinal fluid leakage occurred after surgery.

The ideal repair material should have the following properties: easy to handle and no stitching required; With a high degree of tightness and excellent mechanical properties; Good biocompatibility, easy to be absorbed and promote tissue regeneration; Can reduce the formation of scars, and does not adhere to the surrounding tissue; Widely available and affordable [4–6]. Homologous materials, as the "gold standard" for dural repair, cover membranous materials from the human body such as fascia lata and periosteum. However, due to the scarcity of donor resources and the increase in surgical incisions, its application has been significantly limited [7, 8]. Synthetic materials have attracted attention due to their low production costs and excellent mechanical properties, including non-absorbable materials such as polytetrafluoroethylene and polyurethane, as well as absorbable materials such as polycaprolactone (PCL) and Poly (L-lactic acid) (PLLA) [9]. However, the poor biocompatibility of these materials limits their potential applications in promoting tissue infiltration growth [10]. Natural materials and acellular materials show excellent biocompatibility. The former includes natural biological products such as collagen and bacterial cellulose (BC); [11] The latter included animal-derived membrane materials such as small intestinal submucosa (SIS) and porcine peritoneal acellular matrix (PPAM) after acellular treatment [6, 12]. All of these materials showed good histocompatibility and could promote cell regeneration. However, they also have certain limitations, such as higher brittleness and less than ideal sealing properties. By integrating the characteristics of different materials, composite materials realize the incomparable advantages of a single material, thus overcoming the limitations of traditional single materials. At present, the research of dural repair composite materials mainly stays in the preclinical stage, and only a few materials have progressed to the clinical trial stage [9]. In this case, artificial dural membrane (Tianyifu, China) was used for repair. The repair material belongs to the category of biological materials, but its brittleness is high, resulting in its sealing performance is not ideal, so it can not effectively reduce the risk of cerebrospinal fluid leakage.

Intracranial hematoma may occur in different segments following intraspinal tumor surgery, and its causes are complex. Therefore, no definite conclusion can be drawn about its causes at present. In addition to trauma, abnormal coagulation function, unstable circulating blood pressure, and other factors, cerebrospinal fluid loss related to spinal surgery is also recognized as an important factor [13]. In the present case, owing to the opening of the dura mater during tumor resection, the use of nerve dehydrating agent (mannitol), and catheter drainage after the surgery, a large amount of cerebrospinal fluid was lost, which contributed to the

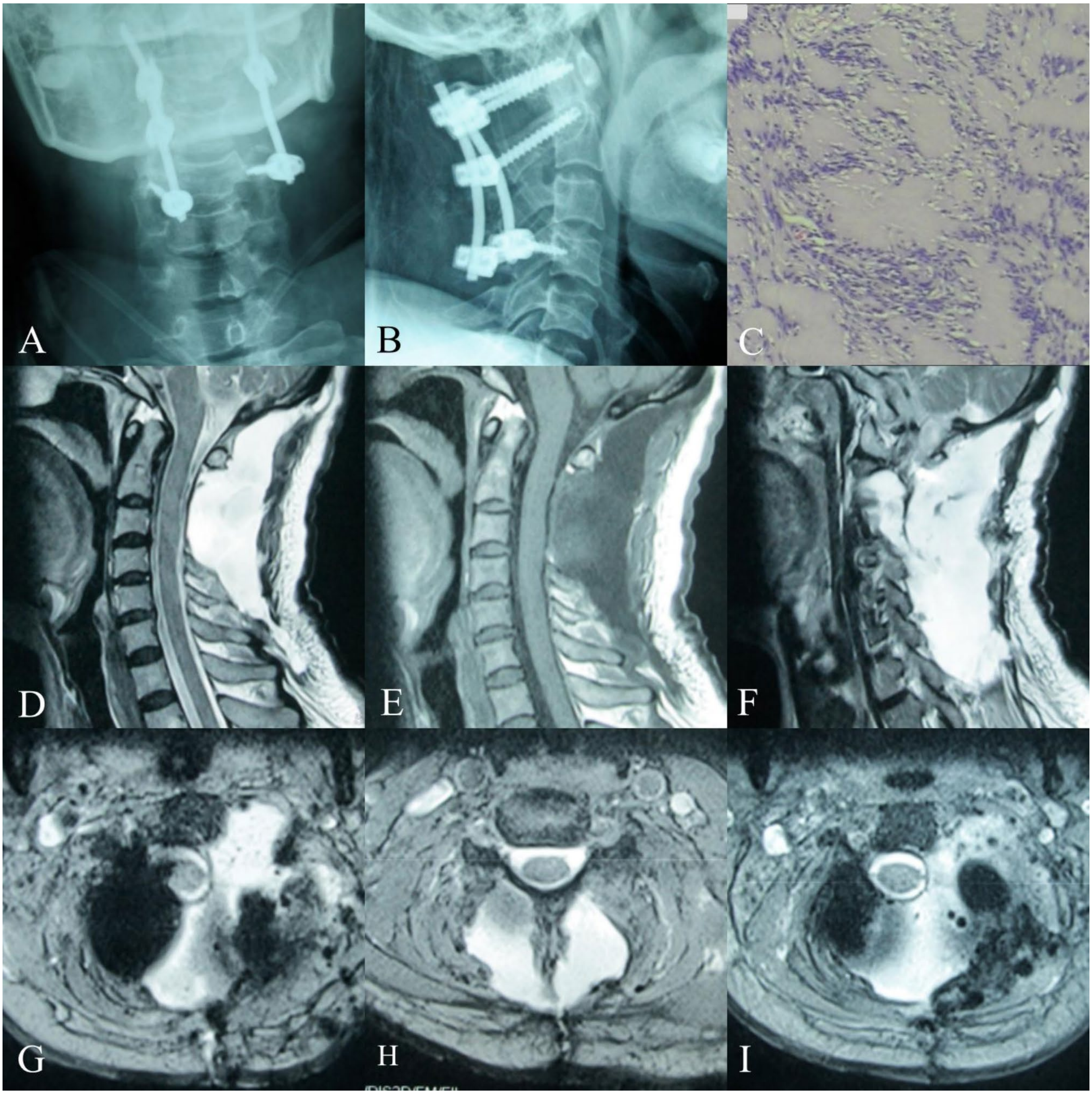

Fig. 3 Postoperative imaging and pathological data: **A**-**B**: postoperative X-ray film shows posterior cervical internal fixation in place; **C**: Neurilemmoma indicated by pathology; **D**-**I**: After cervical spine surgery, MRI showed that the tumor had been completely removed, but a large area of cerebrospinal fluid leakage remained

formation of acute intracranial hematoma. Additionally, the special tumor growth location, difficult surgical resection, long operation time, and factors such as bleeding during the operation may have played a certain role in the formation of a hematoma. Moreover, reportedly, intracranial hematoma may form when a patient is in a sitting or an overextended position that compress the jugular vein during surgery [14]. However, our patient was in the prone position during the operation, and no obvious head hyperextension was observed. Theoretically hypothesize the reason why CSF might have been the cause of hemorrhage.

Most cerebrospinal fluid leaks can be closed and healed naturally in ~1 week. However, if the dura mater defect is too large, it may lead to prolonged healing, which may result in intracranial secondary infection and recurrent meningitis [15]. The lumbar cistern drainage method can be adopted to change the circulation of cerebrospinal fluid, partially discharge it out of the body, reduce the level of cerebrospinal fluid in the subarachnoid space,

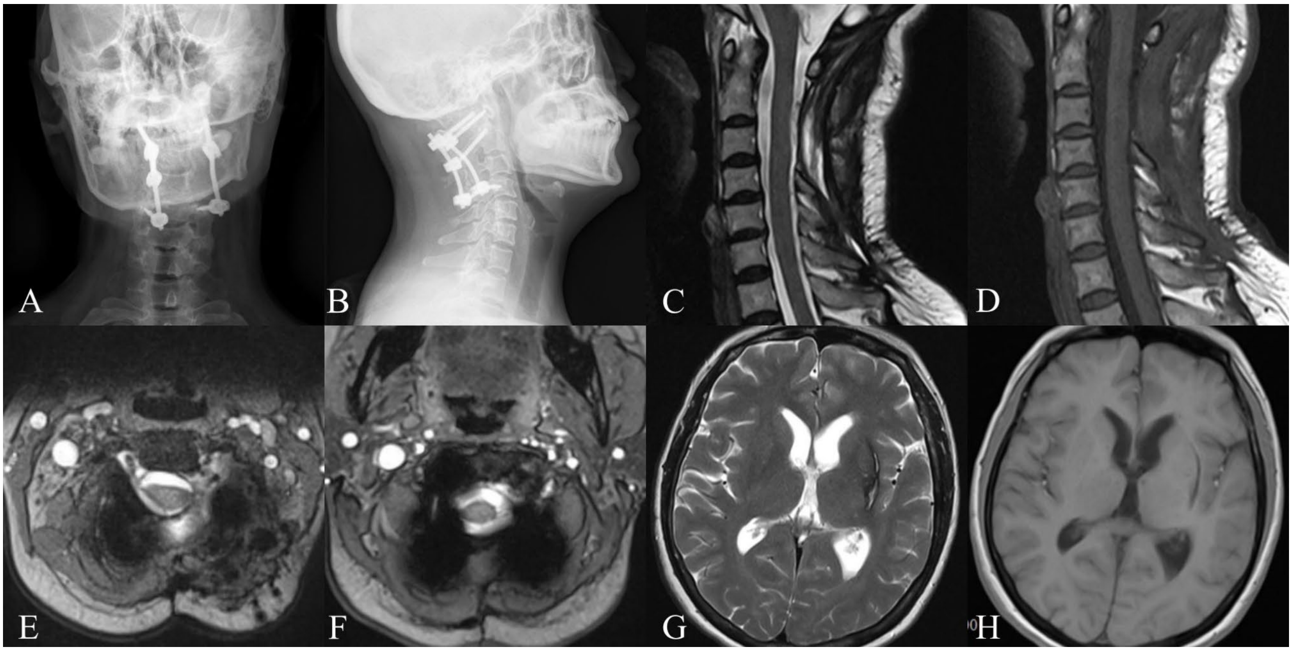

Fig. 4 Imaging data of 1 year follow-up after operation: **A**-**B**: postoperative X-ray film shows posterior cervical internal fixation in place; **C**-**F**: One year after operation, cerebrospinal fluid leakage cavity was obviously absorbed and closed; **G**-**H**: Chronic intracranial hematoma in left basal ganglia

relatively dry the damaged part of the dura mater, and promote growth and healing [16, 17]. For patients with cerebrospinal fluid leakage complicated by intracranial infection, the condition often deteriorates and may even lead to death. In our case, meropenem, an antibiotic that can easily pass through the blood–brain barrier, was used in combination with vancomycin, and lumbar cistern drainage was performed at the same time to avoid deterioration and death. However, continuous external drainage via a lumbar cistern catheter has certain complications, such as secondary intracranial hemorrhage, foramen magnum hernia, and intracranial hypotension syndrome [18, 19].

Some points should be considered when implementing lumbar cistern drainage to avoid these situations. (1) The drainage volume, color, and whether the drainage switch and connector are unobstructed to ensure continuous and gradual cerebrospinal fluid drainage should be closely observed. The drainage speed should be controlled at approximately 2–5 drops/min, 10 ml/h, and 200 ml/day. (2) Whether the drainage tube is discounted, dislocated, and compressed and the official cavity is blocked should be observed. (3) The principle of aseptic operation should be followed, and drainage bags should be changed daily to avoid iatrogenic retrograde infection. Furthermore, comprehensive treatment measures should be taken for patients with cerebrospinal fluid leakage, including antibiotic treatment and lumbar cistern introduction. During treatment, closely observing disease progression and drainage and strictly abiding by the principle of aseptic operation are necessary to ensure the patient safety and treatment effect.

Conclusions

Cerebrospinal fluid leakage combined with intracranial hematoma and cervical infection after cervical dumbbell tumor resection is a severe and complex complication that should be paid great attention.

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Author contributions
Zhenshan Yuan designed and wrote the main manuscript; Liansong Lu and Yong Hu were responsible for studies collection and assessment; All authors read and approved the final manuscript;

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Data availability
The data supporting the fndings of this study are available upon request from the corresponding author (Zhen-shan Yuan, M.D.; E-mail: yuanzhenshan2011@hotmail.com).

Declarations

Ethics approval and consent to participate
The study was approved by the ethics committee of Ningbo No.6 Hospital. The number: CHEC2009-066.

Consent for publication
Informed written consent was obtained from the patient to publish their personal or clinical details information.

Competing interests
The authors declare no competing interests.

Abbreviations
Not applicable.

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