Seeding of Abdomen with Primary Intracranial Hemangiopericytoma by Ventriculoperitoneal Shunt: Case Report

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Key words
- Endoscopic third ventriculostomy
- Hemangiopericytoma
- Obstructive hydrocephalus
- Ventriculoperitoneal shunt

Abbreviations and Acronyms
CNS: Central nervous system
ETV: Endoscopic third ventriculostomy
SFT/HPC: Solitary fibrous tumor/hemangiopericytoma
VPS: Ventriculoperitoneal shunt
WHO: World Health Organization

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BACKGROUND: **Ventriculoperitoneal shunt (VPS) placement has been implicated in extraneural metastasis of many primary central nervous system tumors. Reported cases include, but are not limited to, medulloblastoma, germ cell tumor, astrocytoma, oligodendroglioma, lymphoma, ependymoma, melanoma, and choroid plexus tumors. However, a literature review reveals no reported cases of extraneural metastasis of solitary fibrous tumor/hemangiopericytoma (SFT/HPC).**

CASE DESCRIPTION: **Here we report the case of a 34-year-old man with recurrent intracranial malignant SFT/HPC who had undergone surgical tumor resection and subsequent placement of a VPS for obstructive hydrocephalus in 2004. Subsequently, the patient presented in 2011 and again in 2013 with abdominal SFT/HPC metastasis likely caused by the presence of the VPS.**

CONCLUSION: **The case raises concern regarding placement of a VPS in patients with obstructive hydrocephalus caused by SFT/HPC. To avoid spread of SFT/HPC to the abdomen, we propose that patients with intracranial SFT/HPC and obstructive hydrocephalus be treated primarily by endoscopic third ventriculostomy.**

INTRODUCTION

Hemangiopericytoma (HPC) is a rare malignancy known to occur in the musculoskeletal system while less frequently affecting the central nervous system (CNS).[1] HPC arises from fibroblasts with pathologic features resembling those of solitary fibrous tumors (SFT).[2-5] Previously, in the World Health Organization (WHO) classification of CNS tumors, meningeal HPCs were classified as distinct entities from SFTs.[2,6] Meningeal SFT was considered a benign WHO I tumor, whereas an intracranial HPC was classified as a malignant (WHO II or III) tumor, likely requiring adjuvant treatment, typically radiotherapy.[1,2] More recently, the 2016 WHO classification system groups intracranial HPCs with SFTs, using the terminology SFT/HPC.[5]

Meanwhile, radiographically, SFT/HPC can resemble meningioma.[1] It can be of primary intracranial origin in both adult and pediatric populations, with average age of presentation ranging from 38 to 42 years.[1,7,8] Surgical resection is standard treatment.[1] Some SFT/HPC patients suffer from obstructive or postsurgical hydrocephalus requiring cerebral spinal fluid diversion, and there is known delayed metastasis to many organ systems.[7]

It has been reported that neurosurgical procedures including ventriculoperitoneal shunt (VPS) placement are implicated in extraneural metastasis of primary intracranial tumors.[9] As such, the authors performed a U.S. National Library of Medicine PubMed search seeking publications regarding abdominal metastases in the setting of a VPS. The goal was to further identify trends in tumor pathology, extent of resection, timing of VPS placement in relation to development of abdominal metastases, and overall outcome. The search was limited to human studies and reports with 1 or more patients.

Reported cases have included medulloblastoma, germ cell tumor, astrocytoma, oligodendroglioma, lymphoma, ependymoma, melanoma, and choroid plexus tumors, with both pediatric and adult populations affected.[10-26] However, to our knowledge, there has been no previously reported case of extraneural metastasis of HPC in the setting of a VPS.

CASE PRESENTATION

History and Examination

A 34-year-old man underwent an extended right parietal and retromastoid craniectomy for gross total resection of a right occipital and cerebellar paratentorial SFT/HPC at an outside hospital in 2001 (original radiology and pathology unavailable). In 2004, 3 years after the initial resection, he presented to the emergency department at our institution with a large intracranial hemorrhage with intraventricular extension secondary to recurrent tumor (**Figure 1**). He was unresponsive with an asymmetrically larger right pupil.

Treatment Course

A repeat (second) surgical resection was performed emergently, as the patient had

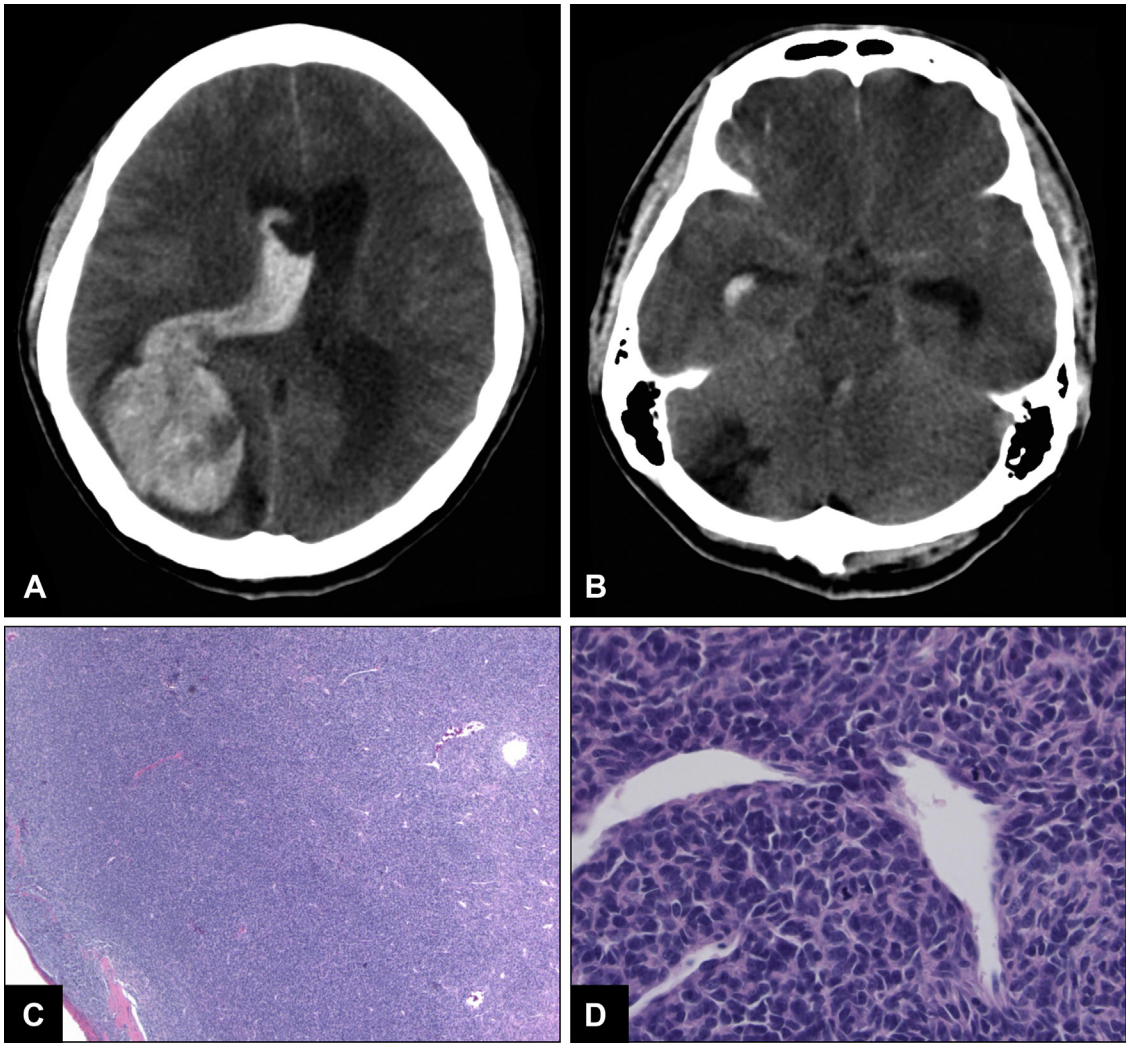

Figure 1. (**A**) Noncontrast head computed tomography demonstrates a large right parietal intracranial hemorrhage with intraventricular extension, hydrocephalus, and midline shift. (**B**) Cerebellar encephalomalacia from prior tumor resection is appreciated. (**C**) 40× magnification, hematoxylin-eosin stain photomicrograph showing a highly cellular, spindled lesion in the brain. (**D**) 400× magnification hematoxylin-eosin stain photomicrograph showing blue ovoid to spindled cells with hyperchromatic nuclei, high nuclear-to-cytoplasmic ratio, and 2 mitoses within this field. The large, irregular vessels are characteristic (though not specific) for solitary fibrous tumor/hemangiopericytoma.

presented in extremis requiring decompression, and pathology was consistent with HPC. He subsequently developed obstructive hydrocephalus necessitating placement of a VPS 1 month following the repeated surgical resection. This was followed by radiation therapy to 54 Gy with conformal technique 5 months later. Almost 5 years later, in 2009, surveillance imaging revealed intracranial recurrence, for which he underwent 30 Gy radiosurgery (Figure 2). In 2011, he developed rapid-onset urinary retention, and computed tomography of the abdomen and pelvis revealed an 8.5 × 10 cm rectoprostatic mass with mass effect on the bladder, encroachment of the prostate and seminal vesicles, and displacement of the sigmoid colon (Figure 3). This was 10 years after his initial diagnosis of intracranial disease and 7 years after placement of a VPS. A low anterior resection was performed to remove the vascular, encapsulated mass consistent with SFT/HPC, after which the patient received adjuvant radiation of 37.5 Gy. Two years later, he underwent a right hemicolectomy and resection of a new 3.0 × 2.6 cm right lower quadrant abdominal mass, embedded in the mesentery adjacent to the right colon and cecum (Figure 4). Pathology was consistent with metastatic SFT/HPC. At the same time, the VPS was removed and an endoscopic third ventriculostomy (ETV) was successfully performed to treat the obstructive hydrocephalus. In 2014, he developed a 5 × 4 mm right occipital parafalcine nodule consistent with recurrence, treated with 20 Gy radiation. Five months later, he was diagnosed with a 2.1 × 1.2 cm right posterior, subpleural, chest wall mass, for which he underwent

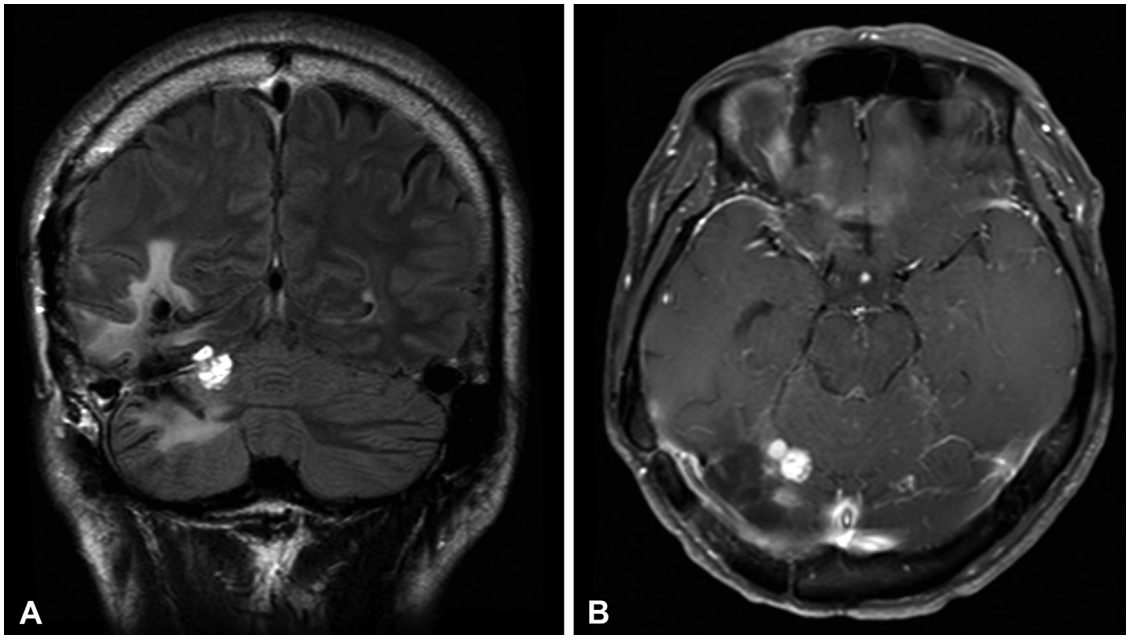

Figure 2. (**A** and **B**) Surveillance magnetic resonance imaging with gadolinium demonstrates interval development of an enhancing mass along each side of the right tentorium.

right thoracoscopic resection, and tissue was consistent with SFT/HPC (Figure 5). In 2015, he developed a right distal humerus lesion and was treated with 25 Gy radiation. More recently, in 2016, he developed a progressing para-aortic abdominal mass. He was treated with Sunitinib and completed 35 Gy palliative radiation therapy for back pain related to his abdominal metastatic disease. He will soon start a course of Avastin.

Pathology

Histopathologic examination of the intracranial lesion removed in 2004 demonstrated a highly cellular tumor with abundant thin-walled vessels showing distinct staghorn configuration, as well as multiple mitotic figures. Pertinent negatives included CD99, TLE-1, BCL2, and cytokeratins. As such, pathologic analysis was consistent with recurrent/residual SFT/HPC (see Figure 1).

Histopathologic examination of the 10-cm rectoprostatic mass, 7 years later, was also found to be a spindle cell neoplasm with staghorn-like intratumoral blood vessels and focal necrosis. There were 20 mitoses per high-power field. Ki-67 proliferation index was between 10% and 20%. Pathologic features were consistent with metastatic malignant SFT/HPC (see Figure 3). The mass involved, but did not invade through, the superficial serosa of the sigmoid colon, suggesting the tumor did not originate from a specific pelvic organ. Superficial location additionally supports the theory of external attachment from tumor cells within the peritoneal space, coming from the distal shunt catheter.

In 2013, histopathologic examination of the right lower quadrant mass was again consistent with metastatic malignant SFT/HPC presenting as a "mesenteric nodule" (see Figure 4). There were no other metastatic foci, nor was there involvement of any lymph nodes found within the surgical specimen. These findings suggested that tumor origin was not abdominal. On the basis of the gross and microscopic characteristics of these 2 masses in relation to the abdominal and pelvic organs, we believe these 2 metastases were each a consequence of tumor seeding the abdomen via the patient's VPS, placed 7 years before diagnosis of the first abdominal metastasis.

The patient later developed a subpleural right lung mass in 2014, also consistent with metastatic SFT/HPC (see Figure 5), and a right distal humerus mass, which was not biopsied, in 2015.

DISCUSSION

Hemangiopericytoma comprises only 0.4% of primary CNS tumors and most often originates from the meninges of the brain and spinal cord.[27-29] It is a vascular neoplasm that arises from the fibroblasts and thus has many possible sites of origin throughout the body.[5] Hemangiopericytoma is an aggressive tumor, resistant to chemotherapy, with up to a 90% reported local recurrence rate and a 20% rate of metastasis.[30] Intracranial recurrence is common and strongly correlated with metastasis.[7] Metastases to many different organs have been reported including bone, lung, liver, breast, thyroid, adrenal glands, kidney, lymph nodes, and pancreas.[12,29,31,32] In 1 retrospective study of 94 cases of CNS SFT/HPC, the abdominal cavity was reported among the most common sites of metastasis.[7] This occurs through both hematogenous and lymphatic routes.[12] A pelvic location is, however,

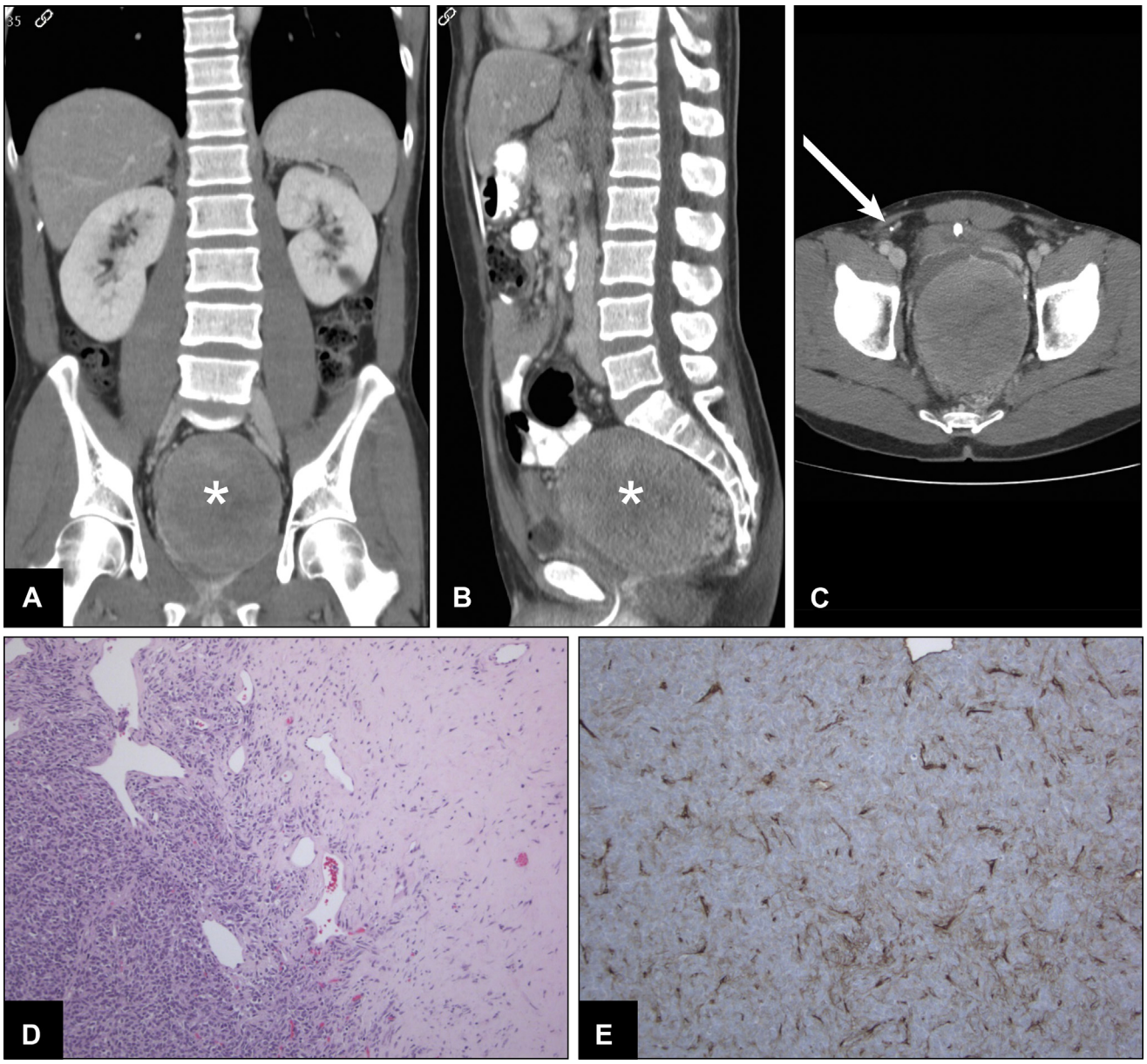

Figure 3. (**A**–**C**) Computed tomography abdomen/pelvis with contrast demonstrates an 8.5 × 10 cm pelvic rectoprostatic mass (*asterisks*) with mass effect on the adjacent structures and rightward displacement of the rectosigmoid colon. There is no periosteal reaction or cortical break to suggest bony involvement. The *arrow* demonstrates the distal tip of the ventriculoperitoneal shunt tubing. (**D**) 100× hematoxylin-eosin stain shows tumor on the left, with spindled to ovoid cells, and characteristic irregular vessels (staghorn). At the right is the involved fibrosed serosa. (**E**) 100× CD34 immunostain outlines the vasculature throughout the tumor and, as shown here, is focally positive in some of the tumor cells themselves.

exceptionally rare.[13] As in the case we report, metastases are typically discovered years after initial treatment. One review of 38 cases reported mean time to metastasis discovery, without a VPS in place, was 8 years[33] but has been delayed up to 24 years.[32] To our knowledge, there are no previously reported cases of extraneural SFT/HPC metastasis in the setting of a VPS.

Upon review of the literature, extraneural metastases associated directly with ventriculoperitoneal shunting include medulloblastoma, germ cell tumor, astrocytoma, oligodendroglioma, lymphoma, ependymoma, melanoma, and choroid plexus tumors, in both pediatric and adult populations (**Table 1**).[10-26,34-36] Time from VPS placement to identification of distant metastases ranged from 1 month to 3 years. Although the extent of resection was not always documented, treatment

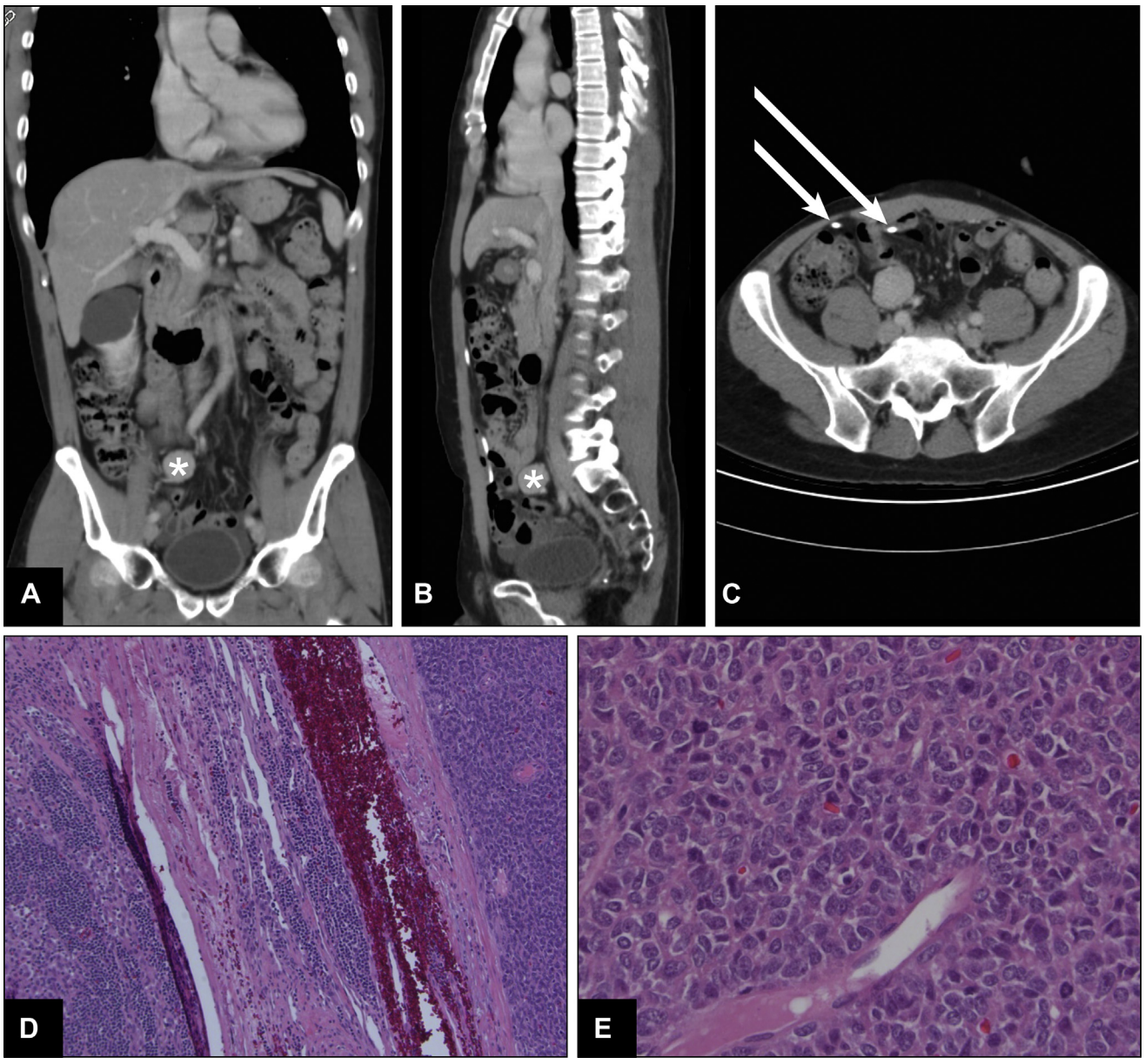

Figure 4. (**A**–**C**) Computed tomography chest/abdomen/pelvis with contrast demonstrates a 3.0 × 2.6 cm mass in the right lower quadrant (*star*). The *arrows* point out the distal tubing of the ventriculoperitoneal shunt. (**D**) 100× magnification hematoxylin-eosin stain shows tumor (far right) abutting the capsule of an uninvolved lymph node (at left) from the ileocecectomy specimen. (**E**) 400× hematoxylin-eosin stain shows ovoid cells with mitoses, morphology is similar to prior specimens.

predominantly involved biopsy only, radiation only, or subtotal resection, of those reported.[7,8,11,13,15,16,20,21,24,26,28-30] Furthermore, outcomes typically resulted in death within weeks to months after distant metastases were identified.[2,14,21,23,24,28,30]

The case we present here is unique in comparison with previously reported extraneural metastases via a VPS.[1-31] The case involves intracranial SFT/HPC, completely resected twice, with much later diagnosis of distant metastases (7 and 9 years following VPS placement), as well as improved outcome (alive at recent follow-up, 15 years after initial diagnosis, and 5 years after the occurrence of metastatic spread).

However, the patient's metastatic disease progression has followed a course similar to that of other reports of primary intracranial SFT/HPC where there was no VPS.[27-29] The patient experienced local recurrence before metastatic disease was detected, 10 years after initial diagnosis of intracranial pathology. It is possible that he would have developed pelvic

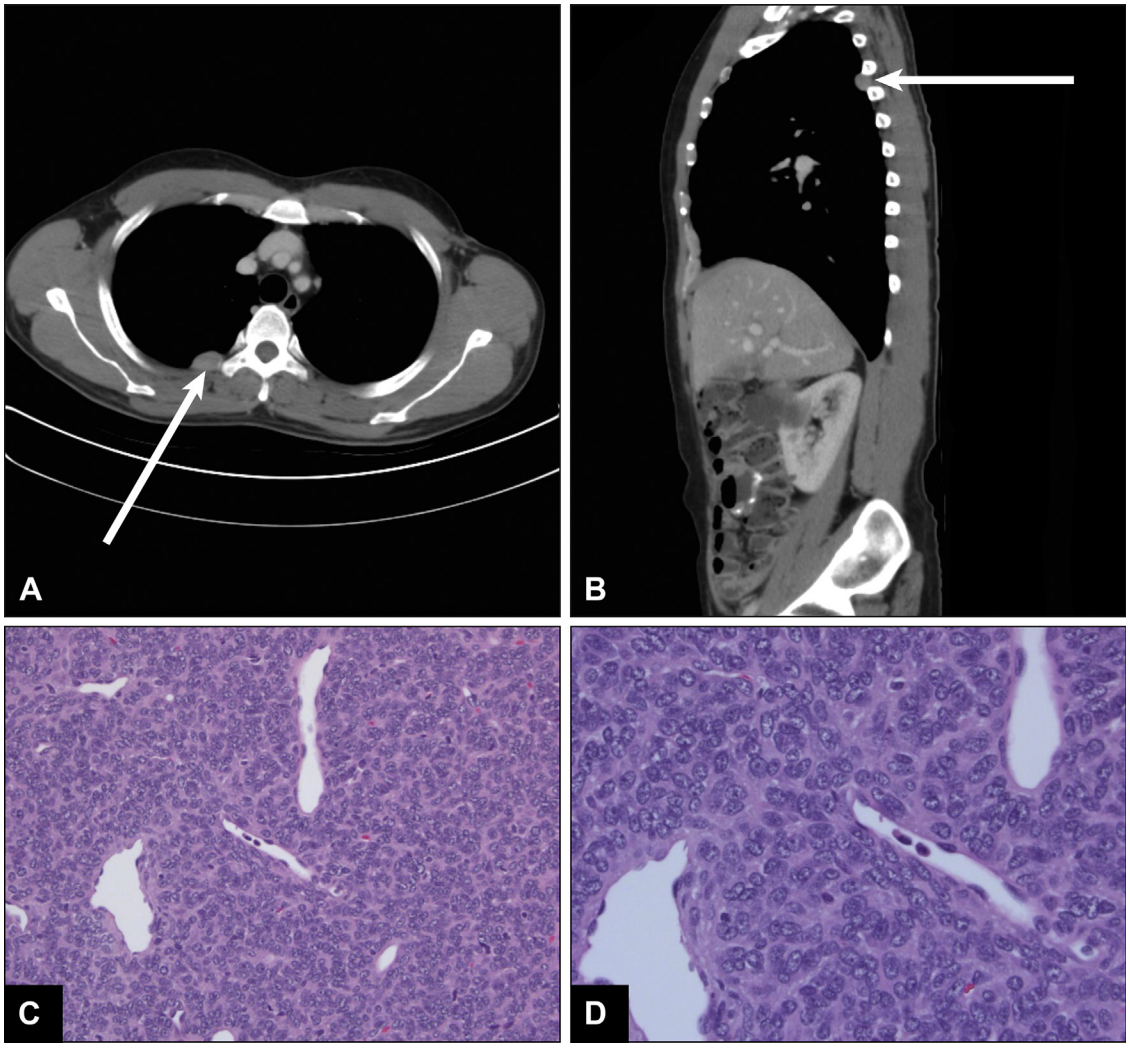

Figure 5. (**A** and **B**) Computed tomography chest/abdomen/pelvis with contrast demonstrates a 2.1 × 1.2 cm enhancing soft tissue mass at the posterior aspect of the right lung apex (*arrows*). (**C**) 200× hematoxylin-eosin stain shows the pleural mass with similar morphology as prior specimens, a highly cellular lesion forming irregular vascular spaces. (**D**) 400× hematoxylin-eosin stain shows ovoid cells with mitotic figures and irregular vascular spaces.

metastases in this time frame regardless of the presence of the VPS. However, the histologic assessment of the relationship to the bowel serosa the first adherent to the superficial serosa of the sigmoid colon (rather than transmural or deep) and the second embedded in the colonic mesentery raises strong suspicion for abdominal seeding of SFT/HPC by VPS. This is corroborated by the lack of lymphatic involvement and absence of other metastases at the time of diagnosis. Furthermore, the patient's better-than-expected condition and prolonged survival are inconsistent with the natural history of extraneural metastatic SFT/HPC. Therefore the neurosurgeons, general surgeons, and pathologists feel this lesion is more consistent with spread via the VPS. As such, the authors propose that patients with intracranial SFT/HPC and obstructive hydrocephalus be primarily treated with ETV when deemed safe to prevent the potential possibility of extraneural spread via a VPS.

CONCLUSION

Ventriculoperitoneal shunt placement has been implicated in extraneural metastasis of many primary CNS tumors yet never previously reported to include SFT/HPC until this case report.[1-31] We suggest that patients with intracranial SFT/HPC requiring permanent cerebral spinal fluid diversion should be considered first for ETV to minimize risk of extraneural metastasis.

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Table 1. Summary Literature Review of Case Reports Discussing Extraneural Metastases in Setting of Ventriculoperitoneal Shunt Placement

Source	Patient Age	Gender	Tumor Pathology	Time from VPS Placement to Identified Distant Metastases	Extent of Resection	Outcomes Noted
Pettersson et al., 2012[22]	5 years	Male	Anaplastic medulloblastoma	3 months	Subtotal	Hospice 5 months later, quickly deceased
Magtibay et al., 2003[17]	37 years	Female	Medulloblastoma	5 years	Subtotal	Doing well 10 months after bone marrow transplant
Jimenez-Jimenez et al., 1991[13]	4 years	Female	Brainstem (pontomesencephalic) glioma	5 months	Radiation only	NR
Loiacono et al., 2006[16]	35 years	Female	Medulloblastoma	5 years	Subtotal	Palliative laparotomy for excision
Mori et al., 1977[18]	2 years	Female	Medulloblastoma	1 month	NR	NR
Shibasaki et al., 1977[24]	9 years	Male	Ependymoblastoma	18 months	Subtotal	Deceased 22 months after mets identified
Becker et al., 1978[10]	16 months	Female	Oligodendroglioma	3 months	NR	Deceased <1 month after mets identified
Neuwelt et al., 1979[19]	16 years	Male	Germinoma	2.5 years	Radiation only	Alive at 12 months (VPS removed)
Oberbauer et al., 1979[21]	14 months	Female	Ependymoblastoma	5 months	NR	Deceased <1 month after mets identified
Wood et al., 1979[26]	11 years	Male	Germinoma	41 months	Radiation only	Alive at 5 years
Wood et al., 1979[26]	13 years	Female	Germinoma	17 months	Radiation only	Deceased <1 month after mets identified
Trigg et al., 1983[25]	3 years	Male	Astrocytoma WHO I (Optic glioma)	10 months	1/3 resected	Alive at 18 months later
Nishio, et al., 1988[34]	19 months	Male	Optic chiasm hypothalamic pilocytic astrocytoma	4 years	NR	Deceased 5 years after mets identified
Pfletschinger et al., 1986[35]	12 years	Female	Pineoblastoma	1 year	NR	NR
Newton et al., 1992[20]	9 years	Female	Glioblastoma WHO IV	2 months	Biopsy	Deceased 4 months after mets identified
Newton et al., 1992[20]	13 years	Male	Glioblastoma WHO IV	3 months	Biopsy	Deceased 4 months after mets identified
Pollack et al., 1994[23]	6 months	Male	Astrocytoma WHO II	2 months	Subtotal followed by "nearly complete"	Alive at 9 years
Gattuso et al., 1995[36]	16 years	Female	Melanoma	NR	NR	NR
Fiorillo et al., 2001[11]	22 months	Male	Medulloblastoma	2 years	Gross total	Alive at 20 months after mets identified
Kun et al., 1981[14]	14 years	Male	Malignant germinoma	Approximately 17 months	Biopsy	Alive 38 months after mets identified
Haimovic, et al. 1981[12]	27 years	Male	Germinoma	3 years	Radiation only	NR
Lewis et al., 1973[15]	46 years	Male	Medulloblastoma	26 months	NR	Deceased 7 months after diagnosis

VPS, ventriculoperitoneal shunt; NR, not reported; WHO, World Health Organization.

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