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Case Report

Widespread Intra-abdominal Carcinomatosis from a Rhabdoid Meningioma after Placement of a Ventriculoperitoneal Shunt: A Case Report and Review of the Literature

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Abstract

Intra-abdominal metastasis (IAM) of central nervous system (CNS) tumors through ventriculoperitoneal shunt (VPS) is rare but has been previously reported (e.g., germinomas and medulloblastomas). However, there has been no previous reports in literature involving meningiomas. A case of primary rhabdoid meningioma with widespread intra-abdominal carcinomatosis after placement of a VPS in a 36-year-old man is described. The patient underwent preoperative angioembolization of the tumor, craniotomy, and surgical excision, followed by postoperative gamma knife radiosurgery. Five months later, he underwent a decompressive craniectomy and surgical excision for tumor recurrence causing raised intracranial pressure and communicating hydrocephalus, necessitating placement of a VPS. One month after placement of the VPS, the patient developed abdominal distension and confusion. He was treated for a VPS infection and the shunt was explanted. He continued to deteriorate with high output from the peritoneal drain placed at the time of shunt explantation. An exploratory laparotomy revealed multiple diffuse peritoneal and omental nodules which had the same histopathological and immunohistochemical morphology as the primary tumor. We reviewed the current literature on IAM of primary CNS tumors through VPS, which revealed that patients belonging to the pediatric age group of the male gender and with a primary intracranial germinoma or medulloblastoma have a higher incidence of IAM. The majority of IAM occurred within 2 years of VPS placement, and patients most commonly present with abdominal distension and ascites. Treatment after diagnosis is varied, and the prognosis is poor, with more than half of the patients dying within a year. It is vital for clinicians to maintain a high index of suspicion for similar patients as early intervention could potentially improve patient outcomes and patient expectations managed more effectively.

Keywords: *Meningioma, metastasis, ventriculoperitoneal shunt*

Introduction

Meningiomas are the second most common symptomatic primary brain tumors in adults and account for one-third of all primary brain and central nervous system (CNS) tumors.[1,2] They are a diverse set of tumors derived from arachnoid cap cells of arachnoid villi in the meninges. Rhabdoid meningioma is an aggressive and unusual variant of meningioma that was first described and introduced by Kepes *et al*. and Perry *et al*. in 1998 and then subsequently added into the World Health Organization (WHO) classification of CNS tumors as a Grade III (malignant) tumor in 2000.[3-5] The latest 2007 WHO classification identified 15 different types of meningiomas based on morphological criteria and stratified them into three grades.[6] Grade I (benign) tumors form the vast majority, while Grade III (malignant) lesions remain rare.[7] Treatment of Grade III CNS meningiomas usually comprises a combination of surgical excision and radiotherapy.[8]

Any intracranial pathology is a common cause of acquired hydrocephalus. Implantation of a ventriculoperitoneal shunt (VPS) is the most widely used treatment to manage persistent hydrocephalus but can be associated with a number of complications, such as mechanical failures, infections, and dissemination of tumor cells.[9] Intra-abdominal metastasis (IAM) or seeding of CNS tumors through a VPS is rare but has been reported in literature. We describe the first reported case of widespread intra-abdominal carcinomatosis through a VPS in a patient with rhabdoid

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meningioma and present findings from a literature review of patients with primary CNS tumors and subsequent IAM through VPS.

Case Report

Written consent was obtained from the patient's next of kin for publication. The Institutional Review Board approval was not required as this is the case report.

A 36-year-old man presented with left-sided headaches associated with vertiginous giddiness for 3 months. A magnetic resonance imaging (MRI) scan of the brain revealed a 5.7 cm left temporal mass with a significant left cerebral edema and midline shift [Figure 1a]. The patient underwent preoperative angioembolization of the tumor followed by a left pterional craniotomy and excision of the tumor. Histopathological examination of the resected specimen revealed a meningothelial tumor predominantly consisting of cells with eccentrically placed nuclei, occasional prominent nucleoli, and abundant eosinophilic cytoplasm, consistent with a diagnosis of a meningioma with predominant rhabdoid features [Figure 2a]. Extensive tumor necrosis was also noted [Figure 2b]. Immunohistochemically, the tumor cells showed diffuse strong positivity to vimentin with focal expression of epithelial membrane antigen (EMA) and cytokeratins AE1/3 [Figure 2c]. Placental alkaline phosphatase, activin receptor-like kinase 1, and desmin were not expressed by tumor cells. The patient recovered well postoperatively and received gamma knife radiosurgery 2 months later for treatment of residual tumors.

Five months following the initial surgery, a routine follow-up MRI scan of the brain showed tumor recurrence in the left middle cranial fossa measuring 7.9 cm with a midline shift, marked sulcal effacement, basal cistern distortion, and left uncal herniation [Figure 1b]. An emergency left decompressive craniectomy and excision of the recurrent tumor were performed. On postoperative day 3, the patient developed wound dehiscence over the lateral aspect of the craniectomy incision with a significant cerebrospinal fluid (CSF) leak. Computed tomography (CT) scan of the brain revealed increasing ventricular dilatation with intracranial herniation and a VPS was implanted. The patient subsequently recovered well.

The patient represented 1 month later with abdominal pain and distension associated with anorexia, lethargy, and confusion. He was disoriented and his abdomen was distended with generalized tenderness. A CT scan of the brain revealed recurrence of a 7.9 cm left frontotemporal tumor with associated perilesional edema [Figure 1c] while a CT scan of the abdomen revealed moderate amounts of ascites with diffuse peritoneal enhancement suggestive of ongoing peritonitis [Figure 3]. The VPS was explanted with placement of separate extraventricular and peritoneal drains in view of a possible VPS infection. The patient remained septic and critically ill even after the removal of the VPS and administration of broad-spectrum antibiotics, requiring increasing inotropic and ventilatory support. CSF and peritoneal fluid cultures were negative. There was 2–3 L of serous fluid drainage from the peritoneal drain daily and multiple samples of peritoneal fluid sent for cytology and culture did not show any malignant cell or infective organisms.

Diagnostic laparoscopy was attempted to elucidate the cause of persistent sepsis but conversion to a laparotomy was necessary due to dense intra-abdominal adhesions. Intraoperatively, the peritoneal surfaces, greater omentum, serosal surfaces, and mesentery of the small and large bowel were studded with multiple flesh-colored tumor nodules. Histopathological examination of a segment

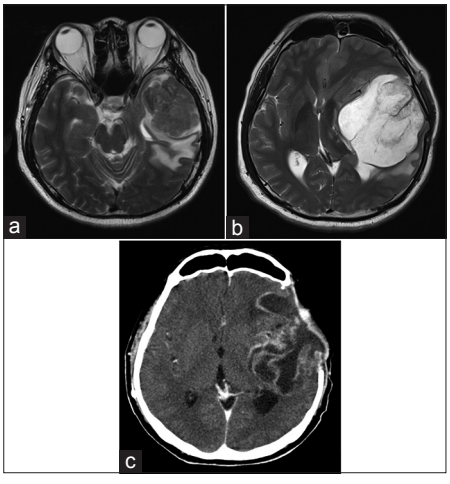

Figure 1: (a) Magnetic resonance imaging scan of the brain showing the initial left temporal mass before surgery. (b) Magnetic resonance imaging of the brain showing tumor recurrence 5 months after initial surgery. (c) Computed tomography of the brain showing tumor recurrence 6 months after diagnosis and after two separate tumor resections

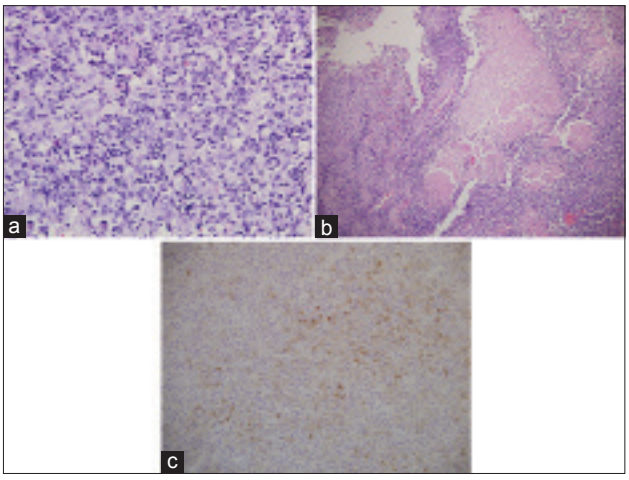

Figure 2: (a) Histology of brain tumor showing meningothelial tumor composed predominantly of cells with abundant eosinophilic cytoplasm and enlarged eccentrically placed nuclei, with occasional prominent nucleoli, consistent with rhabdoid meningioma (H and E, ×400). (b) Extensive geographical tumor necrosis in tumor (H and E, ×100). (c) Positive immunostaining of the tumor cells with epithelial membrane antigen (×200)

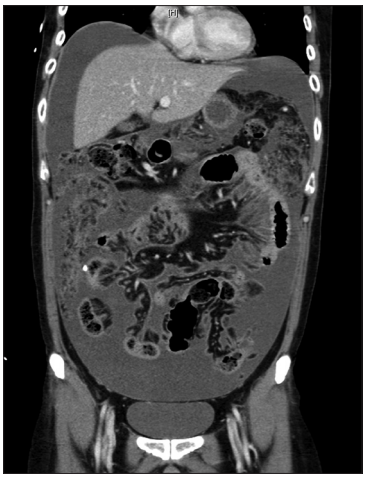

Figure 3: Computed tomography of the abdomen showing ascites with diffuse peritoneal enhancement with diffuse omental thickening and stranding

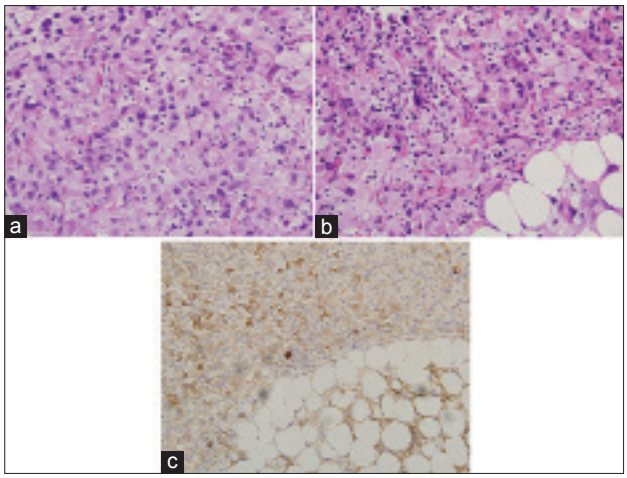

Figure 4: (a) Histology of omental biopsy showing tumor composed of predominantly epithelioid cells with moderate amount of clear or eosinophilic cytoplasm and markedly pleomorphic, irregular, hyperchromatic nuclei with prominent nucleoli, consistent with anaplastic meningioma (H and E, ×400). (b) Cells with rhabdoid appearance characterized by eccentric nuclei displaced by rounded intracytoplasmic eosinophilic inclusions (H and E, ×400). (c) Positive immunostaining of the tumor cells with epithelial membrane antigen (×200)

of greater omentum revealed adipose tissue coated and infiltrated by an extensively necrotic tumor which was composed of predominantly epithelioid cells with moderate amount of clear or eosinophilic cytoplasm and marked pleomorphic, irregular, hyperchromatic with prominent nucleoli [Figure 4a]. Some tumor cells had a rhabdoid appearance [Figure 4b], with eccentric nuclei displaced by rounded intracytoplasmic eosinophilic inclusions, and some had a spindled appearance. The tumor cells stained strongly with vimentin and EMA [Figure 4c]. Morphologically and immunohistochemically, the omental biopsy was similar to that of the primary brain tumor which supports the diagnosis of IAM from the primary brain tumor through the VPS. The patient continued to deteriorate and subsequently died 4 days after laparotomy, secondary to massive bilateral pulmonary embolism.

Discussion

Rhabdoid meningioma is a highly aggressive tumor with a high recurrence rate and poor prognosis. A recent systematic review showed that out of 48 patients with rhabdoid meningioma, almost 50% died within 1 year of surgery and two-thirds had tumor recurrence, with calculated 3- and 5-year recurrence-free survival rates of 41.4% and 27.6%, respectively. It also showed that metastasis most commonly disseminates through the CSF to the spinal cord or other intracranial locations, but extraneural metastasis is rare.[10] Extraneural metastasis to the lung, parotid gland, and liver has been reported and is most likely due to hematogenous seeding.[10-12] Tumor dissemination through VPS to the peritoneal cavity causing widespread intra-abdominal carcinomatosis has been reported in CNS tumors of other histological cell types, but to our knowledge, not in rhabdoid meningioma. In our case, we believe that the VPS was the conduit for transcoelomic spread of malignant cells that were present in the CSF into the peritoneal cavity leading to widespread intra-abdominal carcinomatosis. The patient's abdominal symptoms only started 1 month after placement of the VPS, suggesting that IAM developed during this time period. Furthermore, there was no evidence of other synchronous primary malignancies that could have accounted for the intra-abdominal carcinomatosis and the metastatic lesions found in the abdomen had similar histopathological and immunohistochemical morphology as that of the primary rhabdoid meningioma.

A survey of the literature published from 1979 to 2014 pertaining specifically to patients diagnosed with primary CNS tumors with subsequent IAM from VPS was conducted. Thirty-eight articles with a total of 43 patients were reviewed [Table 1].[13-50] The mean age of patients was 15 years and 3 months, with 69.8% (30 out of 43) of the patients in the pediatric age group (<18 years of age) and 67.4% male (29 out of 43). The overall predominant histological subtypes of primary CNS tumors with VPS-related metastasis were germinomas (11 out of 43, 25.6%) and medulloblastomas (5 out of 43, 11.6%). Other common histological subtypes include endodermal sinus tumors (4 out of 43, 9.3%), glioblastomas (4 out of 43, 9.3%), and atypical teratoid/rhabdoid tumors (3 out of 43, 7.0%). In the adult age group (≥18 years of age), germinomas and medulloblastomas remain the predominant histological subtype at 38.5% (5 out of 13) and 15.4% (2 out of 13) of patients, respectively. The mean time interval from VPS insertion to the occurrence of IAM was 21 months, with 74.3% (26 out of 35) occurring within 2 years of VPS insertion. The most common presenting symptoms of IAM were abdominal distension (41.7%, 15 out of 36), abdominal pain (36.1%, 13 out of 36), constipation (19.4%, 7 out of 36), and vomiting (19.4%, 7 out of 36). The diagnosis of IAM was made after postmortem examination

Table 1: Review of 43 cases of ventriculoperitoneal shunt-related intra-abdominal metastasis reported between 1979 and 2014

Author	Primary central nervous system tumor	Age/ gender	Presenting symptoms of IAM	Time to IAM after VPS insertion (months)	Nature of IAM	Treatment for IAM	Survival after diagnosis of IAM
Yoo *et al*.[13]	Melanotic neuroectodermal tumor of infancy	28 months/ female	Right groin pain and swelling	16	Ascites, peritoneal thickening	Chemotherapy	5 months
Han *et al*.[14]	Atypical teratoid/ rhabdoid tumor	8 years/ female	Abdominal pain, constipation	11	Ascites, multiple abdominal masses	Nil	1 month
Pettersson *et al*.[15]	Anaplastic medulloblastoma	5 years/ male	Fever, nausea, vomiting, thoraco-abdominal wall mass adjacent to VPS	3	Thoracoabdominal wall mass along VPS tract, peritoneal nodules, thickened omentum	Chemotherapy	5 months
Belongia and Jogal[16]	Mixed malignant germ cell tumor with predominant embryonal carcinoma component	7 years/ male	Asymptomatic, incidental finding on spinal imaging	5	Multiple intra-abdominal masses	Chemotherapy	10 months
Murray *et al*.[17]	Pineal germinoma	11 years/ female	Abdominal distension	17	Ascites, pelvic mass, peritoneal nodules	Chemotherapy, resection of mass	>34 months
Boyd *et al*.[18]	Primitive neuroectodermal tumor	23 months/ male	Abdominal distension, vomiting	7	Multiple abdominal masses	Nil	1 month
Cajaiba *et al*.[19]	Neurocutaneous melanocytosis	7 years/ male	NS	84	Ascites, peritoneal nodules	Nil	NS
	Neurocutaneous melanocytosis	4 months/ male	Abdominal distension	32	Ascites, peritoneal nodules	Chemotherapy	5 months
Yokosuka *et al*.[20]	High-grade glioma	12 years/ male	Abdominal pain and fullness	92	Ascites, multiple abdominal masses	Chemotherapy (systemic and intraperitoneal)	13 months
Loiacono *et al*.[21]	Medulloblastoma	35 years/ female	Abdominal pain, constipation	54	Ascites, pelvic, and abdominal mass	Palliative resection of mass	NS
Ingold *et al*.[22]	Atypical teratoid/ rhabdoid tumor	45 years/ female	Postmortem diagnosis	N/A	Multiple abdominal and peritoneal nodules	Nil	12 days
Donovan and Prauner[23]	Choroid plexus carcinoma	3 years/ male	Vomiting, poor nutritional intake	8	Fluid collection	Nil	NS
Yasuhara *et al*.[24]	Glioblastoma	47 years/ female	Postmortem diagnosis	N/A	Splenic nodule	Nil	NS
Magtibay *et al*.[25]	Medulloblastoma	37 years/ female	Urinary frequency	60	Pelvic, retroperitoneal, and right upper abdominal masses	Chemotherapy, resection of mass	>10 months
Altundag *et al*.[26]	Pineal germinoma	23 years/ male	Abdominal distension	24	Ascites, pelvic mass, multiple liver nodules	Chemotherapy	>36 months
Thambi dorai *et al*.[27]	Medulloblastoma	4 years/ male	Right inguinoscrotal swelling	24	Spermatic cord mass	Resection of mass, chemotherapy	6 months

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Table 1: Contd...

Author	Primary central nervous system tumor	Age/ gender	Presenting symptoms of IAM	Time to IAM after VPS insertion (months)	Nature of IAM	Treatment for IAM	Survival after diagnosis of IAM
Araki *et al*.[28]	Anaplastic ganglioglioma	53 years/ female	Abdominal distension	14	Ascites, peritoneal and omental nodules	Nil	1 month
Korones *et al*.[29]	Atypical teratoid/ rhabdoid tumor	4 years/ female	Abdominal pain	13	Ascites, pelvic mass, peritoneal nodules	Nil	1 month
Rickert *et al*.[30]	Cerebral teratocarcinoma	24 years/ male	Acute ileus	7	Abdominal mass	Resection of mass	1 month
Back *et al*.[31]	Pineal germinoma	10 years/ male	Abdominal pain and distension	13	Abdominal mass	Chemoradiotherapy, resection of mass	>4 months
Wong *et al*.[32]	Pineal germinoma	23 years/ male	Abdominal pain	13	Abdominal mass	Nil	<1 month
Pollack *et al*.[33]	Astrocytoma	6 months/ male	Abdominal distension	2	Ascites	Chemotherapy	>60 months
Jamjoom *et al*.[34]	Medulloblastoma	6 years/ male	Left scrotal swelling	10	Spermatic cord mass	Resection of mass	1 month
Ung *et al*.[35]	Pineal germinoma	13 years/ male	Abdominal pain	37	Abdominal mass	Chemotherapy, resection of mass	>24 months
Newton *et al*.[36]	Glioblastoma multiforme	13 years/ male	Abdominal distension, weight gain	NS	Ascites, thickened omentum	Intraperitoneal chemotherapy	1 month
Jiménez-Jiménez *et al*.[37]	Astrocytoma	4 years/ female	Abdominal distension	5	Ascites, peritoneal nodules	Nil	<1 month
Pallini *et al*.[38]	Pineal germinoma	15 years/ male	Abdominal pain, constipation	2	Pelvic mass, peritoneal nodules	Nil	4 months
Kim *et al*.[39]	Pineal germinoma	36 years/ male	Abdominal distension, vomiting	12	Ascites, peritoneal nodules	Chemotherapy	>6 months
Talamo and Mendelow[40]	Germinoma	25 years/ male	Asymptomatic, liver function abnormalities	10	Ascites, multiple abdominal masses	Nil	1 month
Bamberg *et al*.[41]	Endodermal sinus tumor	16 years/ male	Abdominal pain and distension, vomiting	3	Multiple abdominal masses	Nil	<1 month
Devkota *et al*.[42]	Pineal germinoma	12 years/ male	Abdominal pain and distension, vomiting	24	Pelvic mass, peritoneal nodules	Nil	<1 month
Trigg *et al*.[43]	Optic glioma	3 years/ male	Abdominal distension, constipation	4	Ascites	Chemotherapy	>12 months
Haimovic *et al*.[44]	Pineal germinoma	27 years/ male	Constipation	36	Abdominal mass	Radiotherapy	NS
Kun *et al*.[45]	Germinoma	14 years/ male	Abdominal pain, constipation	14	Pelvic mass	Radiotherapy	>38 months
Triolo and Schulz[46]	Pineal germinoma	15 years/ male	Constipation, weight loss	NS	Pelvic and abdominal mass, peritoneal, and omental nodules	Nil	21 months
Pasquier *et al*.[47]	Meningeal sarcoma	21 years/ female	Postmortem diagnosis	N/A	Liver nodules	Nil	NS
Oberbauer *et al*.[48]	Oligodendroglioma	2 months/ male	Postmortem diagnosis	N/A	Peritoneal and omental nodules	Nil	NS

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Table 1: Contd...

Author	Primary central nervous system tumor	Age/ gender	Presenting symptoms of IAM	Time to IAM after VPS insertion (months)	Nature of IAM	Treatment for IAM	Survival after diagnosis of IAM
Wood *et al*.[49]	Pineal germinoma	11 years/ male	Rectal discomfort	36	Pelvic mass	Chemoradiotherapy	>24 months
	Pineal germinoma	13 years/ female	NS	10	Pelvic mass	Nil	<1 month
	Pineal germinoma	15 years/ male	Abdominal pain and distension	36	Fluid collection	Chemotherapy	NS
Wilson *et al*.[50]	Endodermal sinus tumor	4 years/ male	Postmortem diagnosis	N/A	Peritoneal and omental nodules	Nil	<1 month
	Endodermal sinus tumor	19 years/ female	Abdominal pain and distension, vomiting, diarrhea	NS	Ascites	Chemotherapy	>30 months
	Endodermal sinus tumor	12 years/ female	Difficulty with urination	6	Abdominal mass	Nil	1 month

N/A – Not applicable; NS – Not specified; IAM – Intra-abdominal metastasis; VPS – Ventriculoperitoneal shunt

in five patients. Two patients presented with scrotal swelling due to tumor seeding of the spermatic cord through a concurrent hernia defect, and two patients were asymptomatic at the time of diagnosis – one had liver function test abnormalities which led to further imaging and the other was diagnosed incidentally on follow-up spinal imaging. IAM most commonly manifested as ascites (39.5%, 17 out of 43), peritoneal/omental nodules or thickening (34.9%, 15 out of 43), abdominal masses (32.6%, 14 out of 43), and pelvic masses (25.6%, 11 out of 43). Treatment after diagnosis of IAM varied, with 42.1% (16 out of 38) of patients receiving chemotherapy, of which two patients received intraperitoneal chemotherapy. Eight patients (21.1%) underwent surgery to remove the metastatic tumors, of which four of them received prior chemotherapy, radiotherapy, or both. Two patients received combined chemoradiotherapy, two patients received radiotherapy alone, and 15 patients (39.5%) had no further treatment after diagnosis of IAM. Survival outcomes in this particular group of patients were poor, with 45.7% (16 out of 35) of patients dying within 1 month and 57.1% (20 out of 35) dying within 1 year of diagnosis of IAM. There is neither particular histological subtype, nature of IAM, nor treatment modality that rendered a superior survival outcome. However, it is evident that in our review of the literature, a higher incidence of VPS-related IAM is associated with pediatric patients, male gender, and primary intracranial germinomas or medulloblastomas. Our analysis is consistent with an older review by Rickert *et al*., published in 1998, where patients with VPS-related IAM mainly belonged to the pediatric age group with an overall male predominance and that the most common histological subtypes of primary CNS tumors were germinomas (25.7%) and medulloblastomas (22.9%).[30]

With the VPS serving as a conduit for CSF flow from the ventricular cavity into the abdomen, it is a rare but possible means for dissemination of tumor cells, especially in patients with highly aggressive primary tumors that recur or are not completely removed, or tumors that tend to spread through CSF. There have even been cases reported of primary abdominopelvic tumors seeding into the CNS in a retrograde fashion through the VPS.[51,52] In an attempt to prevent the metastasis of tumor cells, a Millipore filter can be placed at the distal peritoneal end of the VPS. However, obstruction due to plugging of the shunt was a commonly reported complication.[21,53] A CSF irradiation filter, which exposes draining CSF to a localized high-intensity radiation field adequately shielded from surrounding tissue, did not produce promising results either.[54] Alternative forms of CSF drainage have also been explored. Placement of only an external ventricular drain can prevent IAM, but patients are at high risk of developing drain-related infections such as ventriculitis or meningitis, which may result in significant morbidity and mortality.[55] Endoscopic third ventriculostomy is a safe and effective method to relieve obstructive hydrocephalus caused by midline tumors and can be a substitute for a VPS in selected cases.[56] Matsumoto *et al*. developed a percutaneous long-tunneled ventricular drainage (PLTVD) to be used for highly malignant intracranial tumors such as germ cell tumors (GCTs) and medulloblastomas. This involved the cannulation of the frontal horn of the lateral ventricle with a ventricular catheter that was connected to a flow-controlled CSF reservoir. A peritoneal catheter proximally connected to the reservoir was then subcutaneously tunneled, exiting at the upper abdomen, and connected to a drainage system. A study of 13 patients who presented with medulloblastoma or GCT showed zero cases

of extraneural metastasis or infections using this method; however, PLTVD had to be converted to a VPS in one case of communicating hydrocephalus due to dissemination and two cases of adhesive aqueductal stenosis not related to the tumor.[57]

Conclusion

We report the index case of a rhabdoid meningioma metastasizing through the VPS into the peritoneal cavity causing widespread intra-abdominal carcinomatosis. As IAM through a VPS is an unusual phenomenon, whether or not the placement of a VPS increases the risk of a patient with a primary intracranial tumor developing IAM remains controversial. The lack of statistical evidence means that the management and treatment of patients who require a VPS should not be changed. Alternative means of CSF drainage or diversion such as PLTVD merit further discussion and risk-benefit assessment. Meanwhile, clinicians should maintain a high index of suspicion in patients with a primary intracranial tumor and VPS presenting with abdominal symptoms for IAM, upon which a low threshold for abdominopelvic imaging should be maintained.

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Conflicts of interest

There are no conflicts of interest.

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