ORIGINAL ARTICLE

Endoscopic Third Ventriculostomy for Hydrocephalus Due to Tectal Glioma

Roberto Jose Diaz, Fady M. Girgis, Mark G. Hamilton

ABSTRACT: ***Background:*** Tectal gliomas commonly present with hydrocephalus from obstruction of the aqueduct of Sylvius. The creation of a ventriculostomy in the floor of the third ventricle (ETV) has been previously reported to by-pass aqueduct obstruction. The goal of this study was to determine the safety and efficacy of ETV in the presence of an obstructing tectal glioma. ***Methods:*** We retrospectively reviewed the clinical presentation, management, and clinical outcome after ETV in patients diagnosed with tectal glioma and obstructive hydrocephalus in our institution over a period of 15 years. Shunt freedom at follow-up was the main outcome variable. Long-term clinical outcome was assessed at the most recent clinic visit. Clinical outcome was ranked as excellent, good, or poor according to resolution of symptoms and patient functional status. ***Results:*** The median age at presentation was 16.5 years (range: 6.4 to 59 years) and the most common presenting symptom was headache. Eleven patients had ETV as a primary procedure and three patients underwent ETV as a substitute for shunt revision at the time of shunt failure. At follow-up (median 3.9 years, range: 2.2 to 7 years) 13 of 14 patients remain shunt independent with excellent (n=9) or good outcomes (n=5). ***Conclusions:*** In patients with tectal glioma causing obstructive hydrocephalus, ETV can be performed safely in the primary setting or as a substitute for shunt revision. A high rate of shunt freedom (78%-100%) at prolonged follow-up can be expected in this patient population.

RÉSUMÉ: Ventriculostomie du troisième ventricule par voie endoscopique pour une hydrocéphalie due à un gliome tectal. ***Contexte :*** Les patients atteints de gliomes tectaux présentent souvent une hydrocéphalie due à une obstruction de l'aqueduc de Sylvius. Selon des études antérieures, la création d'une ventriculostomie dans le plancher du troisième ventricule (ETV) contourne l'obstruction de l'aqueduc. Le but de cette étude était de déterminer la sécurité et l'efficacité de l'ETV en présence d'un gliome tectal obstructif. ***Méthode :*** Nous avons revu rétrospectivement le mode de présentation clinique, la prise en charge et l'issue clinique après une ETV de patients chez qui un diagnostic de gliome tectal accompagné d'hydrocéphalie obstructive avait été posé dans notre institution au cours d'une période de 15 ans. L'absence de shunt au moment du suivi était le critère d'évaluation principal. Le résultat clinique à long terme a été évalué au moment de la visite la plus récente à la clinique. Le résultat clinique était considéré comme étant excellent, bon ou mauvais selon la présence ou l'absence de symptômes résiduels et selon le statut fonctionnel du patient. ***Résultats :*** L'âge médian au moment de la consultation était de 16,5 ans (écart : 6,4 à 59 ans) et la céphalée était la principale manifestation initiale. Onze patients ont subi une ETV comme traitement principal et trois patients ont subi une ETV comme substitut à une révision de shunt au moment où le shunt a fait défaut. Au moment du suivi (médiane 3,9 ans ; écart 2,2 à 7 ans) 13 des 14 patients ne dépendaient pas d'un shunt avec d'excellent résultats (n = 9) ou de bons résultats (n = 5). ***Conclusions :*** Chez les patients atteints d'un gliome tectal causant une hydrocéphalie obstructive, l'ETV peut être effectuée avec sécurité dans un contexte primaire ou comme substitut à une révision de shunt. À long terme, on peut s'attendre à ce qu'un taux élevé de patients ne nécessitent pas de shunt (78% - 100%).

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The development of neuroendoscopic techniques for the management of obstructive hydrocephalus has provided an alternative to shunt systems for cerebrospinal fluid (CSF) diversion. Endoscopic third ventriculostomy (ETV), pioneered by Mixter in 1923 and later Vries,[1] involves the use of a transmitted light source in the form of a flexible or rigid endoscope to visualize the intraventricular spaces of the lateral and third ventricle. This permits the positioning of blunt instruments or a coagulating laser for the creation of an opening in the tuber cinereum. Such an opening allows the free flow of cerebrospinal fluid generated in the lateral and third ventricles directly into the basal subarachnoid space, obviating passage through the aqueduct of Sylvius and the fourth ventricle. The most common application of ETV has been in the setting of obstructive hydrocephalus to provide for a physiologic correction of hydrocephalus[2] potentially avoiding the complications of ventriculoperitoneal shunting such as recurrent infection and obstruction. The role of ETV has been expanded to include the management of acquired causes of hydrocephalus in the pediatric and adult hydrocephalus population.[3] Intrinsic tumors of the tectal plate, which produce obstructive hydrocephalus due to compression of the aqueduct of Sylvius, have been previously managed by ventriculoperitoneal shunting. However, growing support for ETV as the first choice modality

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for management of these tumors has emerged in the literature.[4,5]

In the present study we evaluate the role of ETV in the management of obstructive hydrocephalus in patients with a clinical-radiologic diagnosis of tectal glioma. By reviewing our experience in this patient population we sought to ascertain the degree of symptom improvement with ETV and the risk of treatment failure as defined by the need for ventriculoperitoneal shunting.

Methods

Approval for review of patient clinical information and imaging was obtained from the University of Calgary health research ethics board. A database of endoscopic procedures conducted in Calgary, Alberta, Canada was searched from 1994 to 2008. Patients who had undergone ETV in the setting of tectal glioma were selected. Demographic information, symptoms at presentation, age at surgery, history of ventriculoperitoneal shunting, extent of symptom resolution, and surgical outcome were extracted from the data set. Patient outcome was reviewed through outpatient clinic chart review. Surgical outcome was judged based on degree of symptom resolution and lack of requirement for a ventriculoperitoneal shunt. Post-operative outcome was graded subjectively by an unbiased observer into four categories, with 'poor' being a worsening of symptoms, 'moderate' being no change in symptoms, 'good' being an improvement of symptoms, and 'excellent' being complete resolution of symptoms.

In our institution ETV is performed via right frontal burr hole positioned at the coronal suture. An endoscope sheath is inserted into the lateral ventricle after canulation with a brain needle to assess depth of insertion. We prefer a flexible endoscope to maneuver through the Foramen of Monro and into the third ventricle. A blunt ended probe is used to open a hole in the tuber cinereum. We routinely inspect the subarachnoid space for any

Table 1: Symptoms at presentation to medical care in patients with tectal glioma

Symptom	Number of Patients
Headache	10 (71%)
Gait disturbance	5 (36%)
Impaired cognition	5 (36%)
Nausea or vomiting	4 (29%)
Depressed level of consciousness	4 (29%)
Syncope or seizure	1 (7%)
Diplopia	1 (7%)
Weakness	1 (7%)
Tremor	1 (7%)

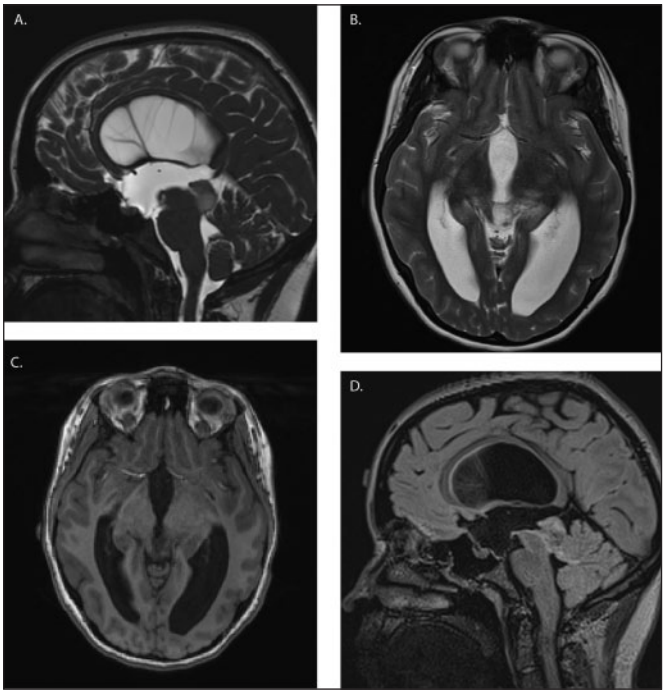

Figure 1: *MRI appearance of a tectal glioma. A) T2-weighted sagittal MRI showing a hyper-intense lesion in the tectal plate causing obstructive hydrocephalus. B) T2-weighted axial MRI of the same lesion. C) T1-weighted axial MRI showing a hypo-intense lesion in the superior colliculus. D) FLAIR MRI showing heterogeneous signal surrounding and within the lesion.*

Table 2: Magnetic resonance imaging features of tectal gliomas

Patient	T1W	T1W +C	T2W	FLAIR
1	Iso	-	Iso	Hyper
2	Iso	N/A	Hyper	Hyper
3	Iso	N/A	Hyper	Hyper
4	Iso	-	Hyper	Hyper
5	Iso	-	Hyper	Hyper
6	Hypo	N/A	Hyper	Hyper
7	Iso	-	N/A	N/A
8	N/A	N/A	N/A	N/A
9	Hypo	-	Hyper	Hyper
10	Hypo	-	Hyper	Hyper
11	Hypo	-	Hyper	Hyper
12	Mixed	+	Hyper	Hyper
13	Hypo	-	Hyper	Hyper
14	Hyper	-	Hypo	Hypo

"Iso" isointense to grey matter, "hypo" hypointense to grey matter, "hyper" hyperintense to grey matter, "+" positive contrast enhancement, "-" no contrast enhancement, "N/A" not applicable

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obstructing membranes. Pulsatile flow through the hole in the floor of the third ventricle is visually confirmed. An external ventricular drain (EVD) is seldom used. However, patients undergoing ETV at the time of ventriculoperitoneal shunt malfunction frequently have an EVD placed at the time of surgery. Magnetic resonance imaging (MRI) of the brain with cine phase contrast imaging of the floor of the third ventricle is routinely obtained to define the degree of CSF flow through the ventriculostomy site. Magnetic resonance imaging cine phase contrast has been used previously to determine the patency and quantitative flow across ETV stoma.[6]

Results

A total of 300 patients were recorded as having endoscopic procedures during the period from 1994 to 2008. There were 40 patients who underwent an endoscopic procedure for a diagnosis of brain tumor. Fourteen patients (nine male and five female) were diagnosed pre-operatively with tectal glioma and the clinical features for each patient are presented in Table 1. The median age at time of surgery was 16.5 years (range: 6.4 to 59 years). The most common presenting symptom was headache. Table 2 summarizes the frequency of all presenting symptoms documented. Five patients had previous ventriculoperitoneal shunting.

Table 2 summarizes the imaging findings available in all 14 patients. The MRI signal characteristics of the tectal lesion were variable; however, the majority demonstrated T1 hypo-intensity with fluid attenuated inversion recovery (FLAIR) and T2 hyper-intensity and no gadolinium enhancement. Figure 1 demonstrates a typical tectal glioma with obstructive hydrocephalus before ETV. In Figure 2, the anatomical defect in the floor of the third ventricle after ETV is visualized on T1-weighted MRI with CSF flow across the floor of the third ventricle demonstrated by cine phase contrast.

Endoscopic biopsy was not attempted in any of the cases presented. Symptoms described on admission improved or resolved in all 14 patients and remained so after a median follow-up of 3.9 years (range: 2.2 to 7 years). Four of the five patients with prior ventriculoperitoneal shunts remain shunt-free. None of the patients developed permanent endocrine dysfunction after ETV. Surgical outcome was deemed "excellent" in nine patients and "good" in five patients by an

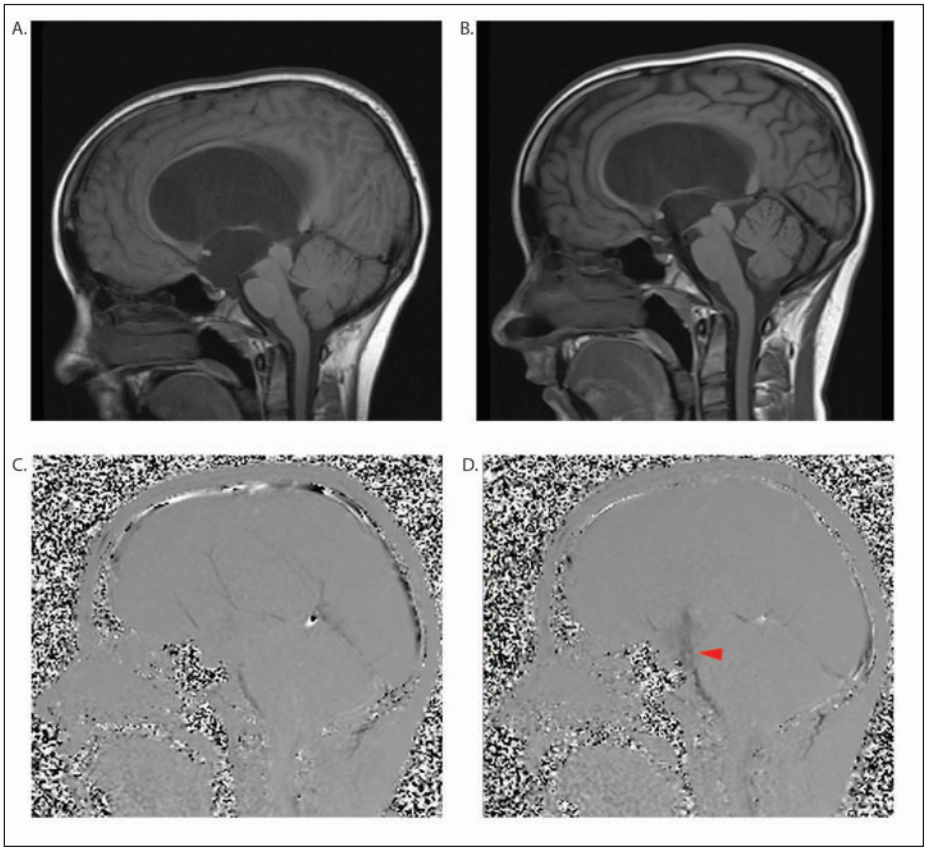

Figure 2: *A) T1-weighted sagittal MRI of an 11 year-old male presenting with syncopal episodes in the setting of obstructive hydrocephalus secondary to tectal glioma. B) T1-weighted sagittal MRI of the same patient three years after ETV. Note the defect in the floor of the third ventricle. C) Pre-operative MR cine phase contrast image in the sagittal plane. D) Post-operative MR cine phase contrast image in the sagittal plane demonstrating CSF flow (red arrow) through the floor of the third ventricle.*

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unbiased observer. Only one patient has a ventriculoperitoneal shunt at present, which was present prior to ETV for management of an isolated lateral ventricle. One patient had his initial ETV at age 11 when he presented with pre-syncopal symptoms and prominent ventriculomegally. He returned six months later with recurrence of presyncopal symptoms, headache, and papilledema at which time a repeat ETV was performed. Follow-up imaging three years after repeat ETV showed reduction in ventricular size and a persistent opening in the floor of the third ventricle. He remains alive with no recurrence of hydrocephalus for the past eight years.

Discussion

The most common presenting symptoms in patients with tectal glioma in our series were headache, gait disturbance, and cognitive impairment. Headache was also found to be a common presenting symptom in a series of ten patients of mixed ages with hydrocephalus secondary to obstructing tectal tumor.[7] Symptoms of raised intracranial pressure are also frequent in children with tectal gliomas.[8-10] Intrinsic tumors of the brainstem are more common in the pediatric population,[11] which explains the young age distribution of the study population presented. Tectal gliomas comprise a subgroup of all pediatric and adult brainstem tumors, which have been reported to have a more favourable prognosis than diffuse lesions in multiple case series.[9,12-18] Hamilton *et al*[14] reported a group of 16 patients with focal midbrain gliomas with mean follow-up of 84 months in which only two patients demonstrated tumor progression. These patients were treated with a variety of strategies, including observation, radiation, chemotherapy and surgery (one biopsy and four debulking). Conservative management involving CSF diversion and no surgical biopsy or resection of tectal lesions has been the favoured approach when such lesions demonstrate characteristic imaging findings of low-grade tumor, indolent growth, and the absence of progressive neurological deficit.[5] While focal tectal and tegmental midbrain lesions may have a benign clinical course, there is still a risk for tumor growth or transformation to higher grade necessitating continued surveillance after surgical intervention to relieve obstructive hydrocephalus. In a series of seven patients under the age of 25, increase in tumor size was observed in six patients and the magnitude of increase in size ranged from 1.6 to 3.8 times the original size.[17] Despite the increase in tumor size in six patients, only one of these had new symptoms related to tumor progression. In a series of 32 children with tectal tumors, 12 demonstrated increase in tumor size or worsening symptoms that triggered further intervention (excision and/or radiotherapy).[19] Sanford *et al*[20] described tectal gliomas as low-grade tumors arising in the tectum surrounding the cerebral aqueduct and producing obstructive hydrocephalus without the brainstem nuclei or long-tract signs observed in diffuse brainstem glioma. A low mitotic index, a feature of low grade gliomas, would favour a slow growing tumor and, in comparison with other low-grade astrocytomas, tectal gliomas may have molecular features which would permit long periods of stable disease or even regression.[21] Nevertheless, great caution should be exerted in follow-up of patients with a non-biopsied lesion presumed to be a low-grade glioma as 18-31% of tumors will continue to grow.[22] It is important to note that tectal gliomas have characteristic imaging findings and therefore biopsy in this delicate area is often not required. Typical lesions cause asymmetric expansion of the tectal plate causing tectal distortion and narrowing of the aqueduct of Sylvius.[15] Computed tomography demonstrates a hypodense lesion which may have calcification in 9-25% of cases, but with no contrast enhancement.[23] Tectal gliomas were previously described as T2 hyper-intense and T1 hypo-intense lesions on MRI,[24] but T1 iso-intensity has also been reported.[10,23] Gadolinium enhancement on T1-weighted MR images has been reported in a minority of patients[15,23] and development of enhancement may suggest a more aggressive lesion.[13,19,25] Both low and high-grade histology has been reported for lesions that show gadolinium enhancement but a lack of correlation between contrast enhancement and tumor grade cannot be fully established given small sample size and the possibility of biopsy sampling error.[23] It is important to note that pilocytic astrocytomas of the brainstem characteristically enhance and are considered low-grade tumors.[26] Larger lesion size (> 1.5 cm) and volume greater than 4 cm^3 is associated with propensity for tumor progression.[15,22] Furthermore, the only clinical or imaging parameter that has been found to predict future lesion enlargement is tumor volume.[22] For lesions with unusual imaging features, especially large lesions with mixed intensity on T1-weighted imaging and contrast enhancement, endoscopic biopsy may be an option if the lesion is visible and prominent. Alternatively, a stereotactic biopsy can be considered if the lesion is not accessible from within the third ventricle. For very large (>10 cm^3) or rapidly growing lesions, early surgical excision should be considered in order to avoid progressive

Table 3: Summary of studies evaluating shunt-freedom after ETV in patients with tectal glioma

Study	Number of Patients	Patient Age Range (years)	Follow-up Range (months)	Shunt Free at Follow-up
Diaz *et al*., 2013	14	6 - 59	14 - 72	13 (93%)
Ramelli *et al*., 2011	10	0.9 - 18	30 - 132	10 (100%)
Ternier *et al*., 2006	26	0 - 17.6	Not specified	26 (100%)
Stark *et al*., 2005	9	0.08 - 16	3 - 336	7 (78%)
Li *et al*., 2005	18	2 - 18	Not specified	16 (89%)
Javadpour & Malluci, 2004	11	9 - 59	2 - 45	9 (82%)
Wellons *et al*., 2002	13	4 - 16	2 - 64	13 (100%)

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neurological deficit and establish an histologic diagnosis which can guide adjuvant therapy, as noted by Ternier *et al*.[22]

In the present patient series, 13 of 14 patients have demonstrated excellent or good clinical outcome, after an average follow-up of 4.4 years, when ETV is used in the setting of acute hydrocephalus secondary to a tectal lesion with radiologic features characteristic of low-grade glioma. Complications were limited and 13 of 14 patients have been spared future shunt-related complications. Our assessment of surgical outcome was determined by an independent observer using clinical follow-up assessments and as such is prone to error by observer bias. However, the finding that only 1 out of 14 patients required ventriculoperitoneal shunting for management of hydrocephalus after ETV adds additional support to our conclusion that ETV can be considered as a first choice for treatment in patients with hydrocephalus secondary to a tectal glioma.

Our results are in agreement with those of Yeh *et al*[27] who demonstrated shunt independence in patients with midbrain gliomas, and Javadpour and Malluci who showed 82% shunt independence in patients with tectal glioma.[28] The observed response rate to shunt freedom (four of five patients with prior shunt) is also in keeping with a prior series demonstrating 76.7% success rate of ETV in the setting of shunt malfunction or infection.[29] Table 3 summarizes all studies pertaining to ETV in the setting of tectal glioma found using a PubMed primary search using the terms "endoscopic third ventriculostomy OR third ventriculostomy OR ETV" AND "tectal glioma OR glioma OR brainstem tumor".[27,28,30-32] While long-term shunt freedom rates are high, patients with ETV are at risk of ETV failure, which can result in rapid neurological decline.[33] Stark *et al*[32] reported five patients who underwent ETV as initial treatment of hydrocephalus due to tectal glioma, with two of these patients requiring ETV revision and one requiring shunt during follow-up. Four patients with tectal glioma have been reported in the literature with late deterioration from an occluded ETV, which resulted in death in three cases.[33,34] Given the potential for ETV failure, family members and patients should be educated about the importance of seeking urgent medical care in the setting of acute symptoms of hydrocephalus, as well as informing their primary care physician of their history of hydrocephalus. Larger studies with longer follow-up are needed, but will be challenging due to the rarity of tectal gliomas (1.3% of pediatric tumors).[13] Multicentre cooperation will likely be needed to accomplish such a study. Furthermore, future studies should focus on a detailed assessment of neurocognitive outcomes after ETV as cognitive impairment is a common presenting symptom of obstructive hydrocephalus and cognitive assessment is often limited in routine clinical follow-up. Cognitive improvement on neuropsychological assessment is observed in a majority of patients after ETV for obstructive hydrocephalus;[35,36] however, some impairment in memory and executive domains may persist in a subset of patients.[37]

Conclusions

In the setting of a compressive tectal glioma, ETV can be successfully used to relieve obstructive hydrocephalus. We have also demonstrated the use of ETV in the setting of ventriculoperitoneal shunt malfunction when the underlying cause of obstructive hydrocephalus is a tectal glioma. However, it is recommended that a ventricular drainage catheter remain until a working ventriculostomy can be confirmed with MR cine phase contrast imaging and clinical improvement is maintained when a sufficient trial of clamping of the ventricular catheter has been undertaken.

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