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Pediatric pineal and tectal region tumors: the use of neuroendoscopy

Pediatrik pineal ve tektal bölge tümörleri: nöroendoskopi kullanımı

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Abstract

Purpose: In this article, we aimed to contribute to the literature by describing our approach to pediatric pineal and tectal masses treated with endoscopic interventions in our clinic.

Materials and methods: We retrospectively reviewed the records of 26 patients with lesions in the pineal and tectal regions who underwent endoscopic procedures between 2012 and 2023 at Gazi University Department of Pediatric Neurosurgery. Demographic data, blood tumor markers (alpha fetoprotein-BETA AFP, alpha fetoprotein-β-hCG), pathological diagnoses, reasons for clinical presentation, presence of hydrocephalus and additional interventions were analyzed.

Results: Of the 26 patients who underwent endoscopic procedure, 6 had lesions in the pineal region and 20 had lesions in the tectal region. The success rate for ETV was 75-80%. The success rate for ETV was 78.3% for patients with tumors in the pineal region and 84% for tectal tumors.

Conclusion: Pineal or tectal region tumors are a rare group of tumors. In addition, these regions are among the most heterogeneous regions in the central nervous system in terms of tumor type and histology. Endoscopic intervention through a single burr hole using rigid endoscopy is a comfortable and safe minimally invasive method for both pathologic sampling and management of hydrocephalus in indicated cases in the same session.

Keywords: Pediatric tumors, pineal tumors, tectal tumors, neuroendoscopy.

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Öz

Amaç: Bu yazıda kliniğimizde endoskopik girişimlerle tedavi edilen pediatrik pineal ve tektal kitlelere yaklaşımımızı anlatarak literatüre katkıda bulunmayı amaçladık.

Gereç ve yöntem: Gazi Üniversitesi Çocuk Nöroşirürji Bilim Dalı'nda 2012-2023 yılları arasında pineal ve tektal bölgede lezyonu olan ve endoskopik girişim uygulanan 26 hastanın kayıtları retrospektif olarak incelendi. Demografik veriler, kan tümör belirteçleri (alfa fetoprotein-BETA AFP, alfa fetoprotein-β-hCG), patolojik tanılar, klinik başvuru nedenleri, hidrosefali varlığı ve ek girişimler analiz edildi.

Bulgular: Endoskopik işlem uygulanan 26 hastanın 6'sında pineal bölgede, 20'sinde ise tektal bölgede lezyon vardı. ETV için başarı oranı %75-80 idi. ETV için başarı oranı pineal bölgede tümörü olan hastalar için %78,3 ve tektal tümörler için %84 idi.

Sonuç: Pineal veya tektal bölge tümörleri nadir görülen bir tümör grubudur. Ayrıca bu bölgeler tümör tipi ve histolojisi açısından santral sinir sistemindeki en heterojen bölgeler arasındadır. Rijit endoskopi kullanılarak tek bir burr deliğinden endoskopik girişim, endikasyonu olan vakalarda aynı seansta hem patolojik örnekleme hem de hidrosefali yönetimi için konforlu ve güvenli bir minimal invaziv yöntemdir.

Anahtar kelimeler: Pediatrik tümörler, pineal tümörler, tektal tümörler, nöroendoskopi.

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Introduction

Pineal or tectal region tumors are relatively rare [1]. Tumors in the pineal region represent 1.5% to 8.5% of pediatric brain tumors and 1.2% of all central nervous system tumors [2, 3]. Brainstem gliomas, on the other hand, account for 10% to 20% of childhood primary brain tumors [4]. Due to the anatomical location of lesions in this region and their mass effect, patients typically present with obstructive hydrocephalus symptoms such as headache, increased intracranial pressure, urinary incontinence, altered consciousness, and seizures [5]. Advances in neuroendoscopic techniques have allowed for the safe and effective management of these region lesions in terms of diagnosis and obstructive hydrocephalus, with the opportunity for cerebrospinal fluid (CSF) sampling and concurrent histopathology [6]. This article aims to contribute to the literature by describing our approach to the pediatric pineal and tectal region masses treated with endoscopic interventions in our clinic.

Material and method

Permission was obtained from the local Ethics Committee of Gazi University. Records of 26 patients with lesions located in the pineal and tectal regions who underwent endoscopic procedures were retrospectively reviewed between the years 2012 and 2023 within the Pediatric Neurosurgery Department at Gazi University. Demographic data of the patients, blood tumor markers (alfa fetoprotein-BETA AFP, alfa fetoprotein-β-hCG), pathological diagnoses, reasons for clinical presentation, presence of hydrocephalus, and additional interventions performed were examined.

The article was scanned by including pediatric-age (<18 years old) human studies published in English through the MEDLINE/PubMed and EMBASE library electronic databases. The primary search was performed using the keywords “Endoscopy”, “pineal,” “Tectal,” and “Hydrocephalus.” The criteria for inclusion were: 1. Pediatric age group and who underwent endoscopic procedures with pineal and tectal region lesions in the cases 2. reports or case series in which sufficient clinical, histological, and surgical information specific to the patient is available. Studies in which pediatric patients could not be distinguished and mixed age groups were presented were not included.

Surgical procedure

Patients were positioned supine, with the head extended at approximately 20-25 degrees, to center the surgical table under the general anesthesia. The entry point was selected as 1 cm anterior to the coronal suture and 2-3 cm lateral to the sagittal suture on the non-dominant side (Figure 1). After determining the incision site, the periosteum was visualized with a vertical incision of approximately 2-3 cm. When the dura was exposed, a linear incision was made using bipolar cautery. Advancement was achieved with a blunt-tipped trocar through inferoposterior and medial openings, descending into the ventricular cavity. During this process, the trocar’s stylet was removed, allowing for the observation of pressurized CSF flow and the collection of CSF samples for cytological and microbiological analysis. In our clinic, controlled irrigation with Ringer’s lactate or normal saline is used to fill the ventricular cavity, preventing the closure of the field of view. Additionally, this method assists in hemostasis for minor bleeding during the procedure while maintaining a clear field of vision.

Subsequently, a 0° camera is introduced through the trocar to ensure orientation. For orientation purposes, the foramen of Monro, thalamostriate vein, septal region, and choroid plexus are identified, and the advancement is guided such that the foramen of Monro is anterior, the choroid plexus is posterior, the septal vein is medial, and the thalamostriate vein is lateral [7]. Care should be taken not to damage the fornix, which forms the anterior and medial part of the foramen of Monro. The anatomical structures to be identified on the floor in the third ventricle include the bilateral mamillary bodies, basilar artery, dorsum sellae, and infundibular recess (Figure 1) [7]. After achieving orientation, if a biopsy is to be taken, 8-12 biopsy samples are obtained as long as the tumor’s bleeding status allows. Suppose an endoscopic third ventriculostomy (ETV) is performed. In that case, the point farthest from the basilar artery, which is the central point between the two mamillary bodies extending from the infundibulum, dorsum sellae, and closest to the clivus, should be selected for stoma creation. Subsequently, the stoma is dilated with a double-balloon neuro balloon or a 3F or 4F Fogarty catheter. The structures of the basilar artery and other interpeduncular cistern structures should be visualized in this area, and the presence of any additional membranes must be confirmed. After the procedure, the endoscope is slowly withdrawn, and the ventricular cavity is checked for potential bleeding. After removing the trocar, the cortical opening is closed with Spongostan or Surgicel, and the subcutaneous tissue and skin are sutured to conclude the operation.

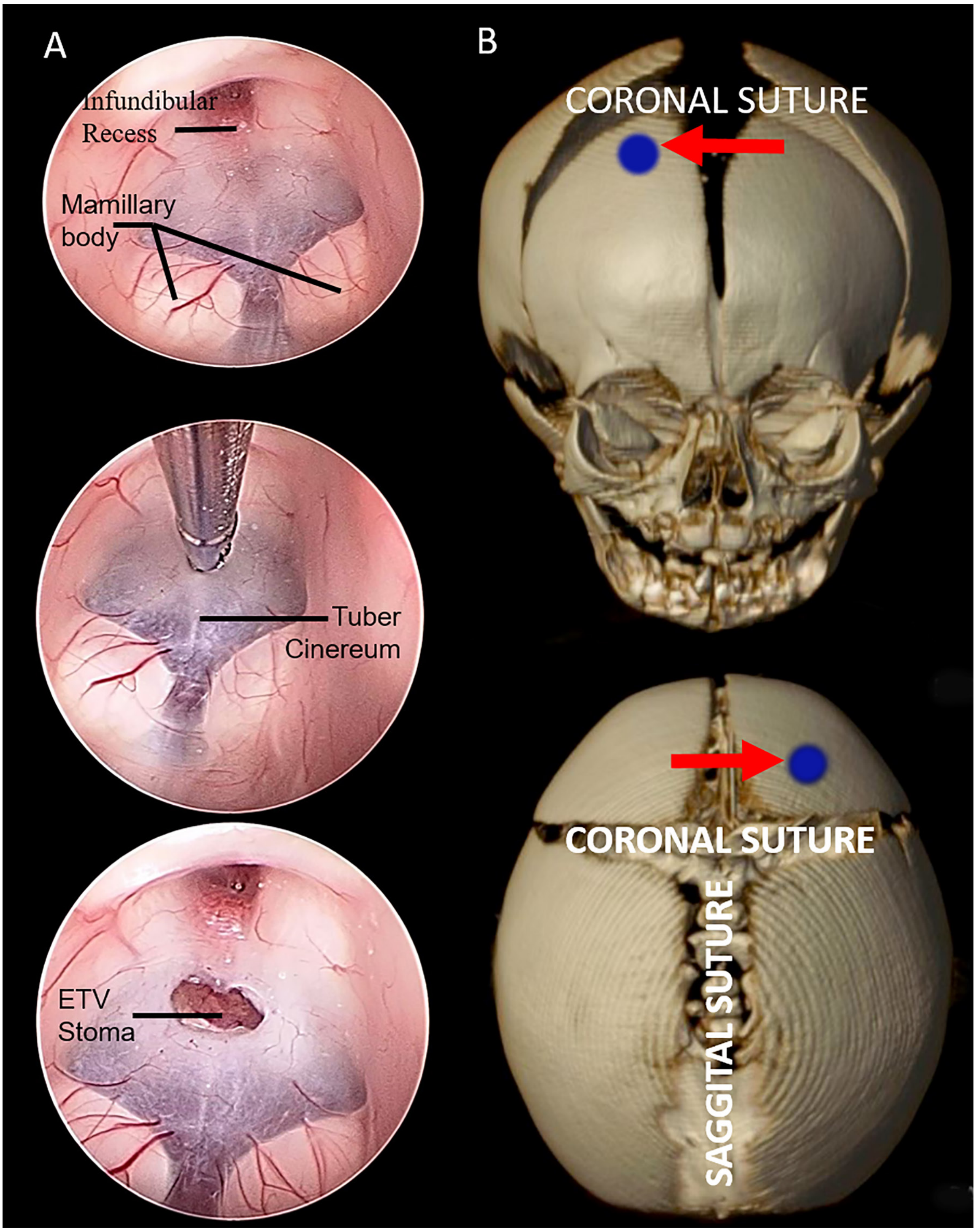

Figure 1. A. Three-Dimensional Computed Tomography (3D CT) scan of the appropriate entry point to be selected in endoscopic surgery, B. The appearance of the surgical area and stoma endoscopically in intraoperative ETV surgery

Results

Among the 26 patients who underwent endoscopic procedures, 6 had lesions in the pineal region, while 20 had lesions in the tectal region. Of the patients, 17 were male and nine were female, with an average age at the time of surgery of 9.5 years (range: 1.1-17.9 years). The most common presenting symptom was headache (46.1% or 12/26), and the most common findings in patients were, in order, unsteadiness 23% (6/26), nausea-vomiting 19.2% (5/26), seizures 11.5% (3/26), visual loss 11.5% (3/26), and spasticity 7.6% (2/26). The follow-up period ranged from a minimum of 15 to a maximum of 126 months, and there were no fatalities among the patients.

All 26 patients had MR findings and clinical symptoms related to hydrocephalus, which led to the performance of ETV. Simultaneous endoscopic biopsies were performed in 6 patients (2 pineal and four tectal). The blood tumor markers of the patients we performed a pineal region-based biopsy were 1-AFP 8.4, β-hCG 2, 2- AFP 182, and β-hCG 135.6. Based on biopsy results, histopathological diagnoses for tectal tumors included Pilocytic Astrocytoma Grade 1 and subependymal astrocytoma, while pineal tumors were diagnosed as germ cell tumors (Figure 2). and pineoblastoma. For two patients with tectal tumors who underwent biopsy, a histopathological diagnosis could not be established. The diagnostic rate for patients who underwent endoscopic biopsy was 66.6% (4/6).

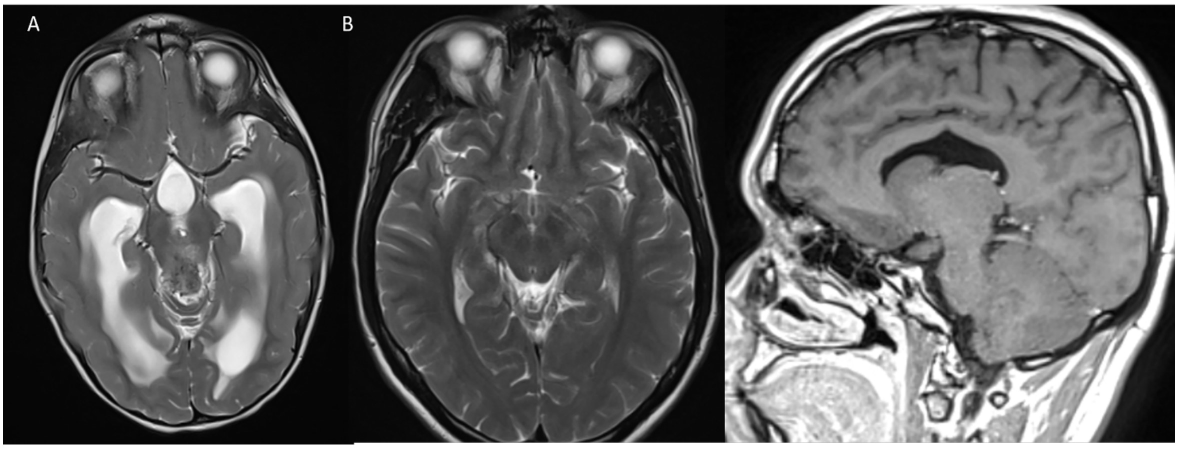

Figure 2. Radiological images of a patient with germ cell tumor pathology ETV+ETB+Adjuvant therapy performed due to pineal mass

A. Preoperative axial T2 MRI image, B. Control axial and sagittal MRI images after eight years postoperative

Among patients who did not undergo simultaneous biopsy and only had ETV (4 pineal and one tectal tumor), subsequent open surgery was performed, resulting in histopathological diagnoses of papillary tumor grade 2-3 in one patient, pilocytic astrocytoma in two patients, oligodendroglioma grade 2 in one patient, and mature cystic teratoma in one patient. ETV was applied to all 26 patients (Figure 3). with 6 (6/11) receiving endoscopic diagnoses and 5 (5/11) undergoing open surgery for diagnosis and treatment. The success rate for ETV in patients was 75-80%. For patients with tumors in the pineal region, the ETV success rate was 78.3%, while it was 84% for tectal tumors. Two patients (2/26) had preexisting shunts before ETV, and two patients (2/26) had VP shunts placed within the first year after ETV (Figure 4). One patient, who initially underwent open tumor resection and did not experience tumor recurrence, developed urinary incontinence three years later, leading to repeat ETV.

No major complications were observed after endoscopic procedures. Two patients had fornix injury, and minor bleeding, which was controlled with irrigation, occurred in six patients after ETV. Long-term follow-up did not reveal radiological progression or clinical deterioration requiring surgical intervention for tumor-related issues.

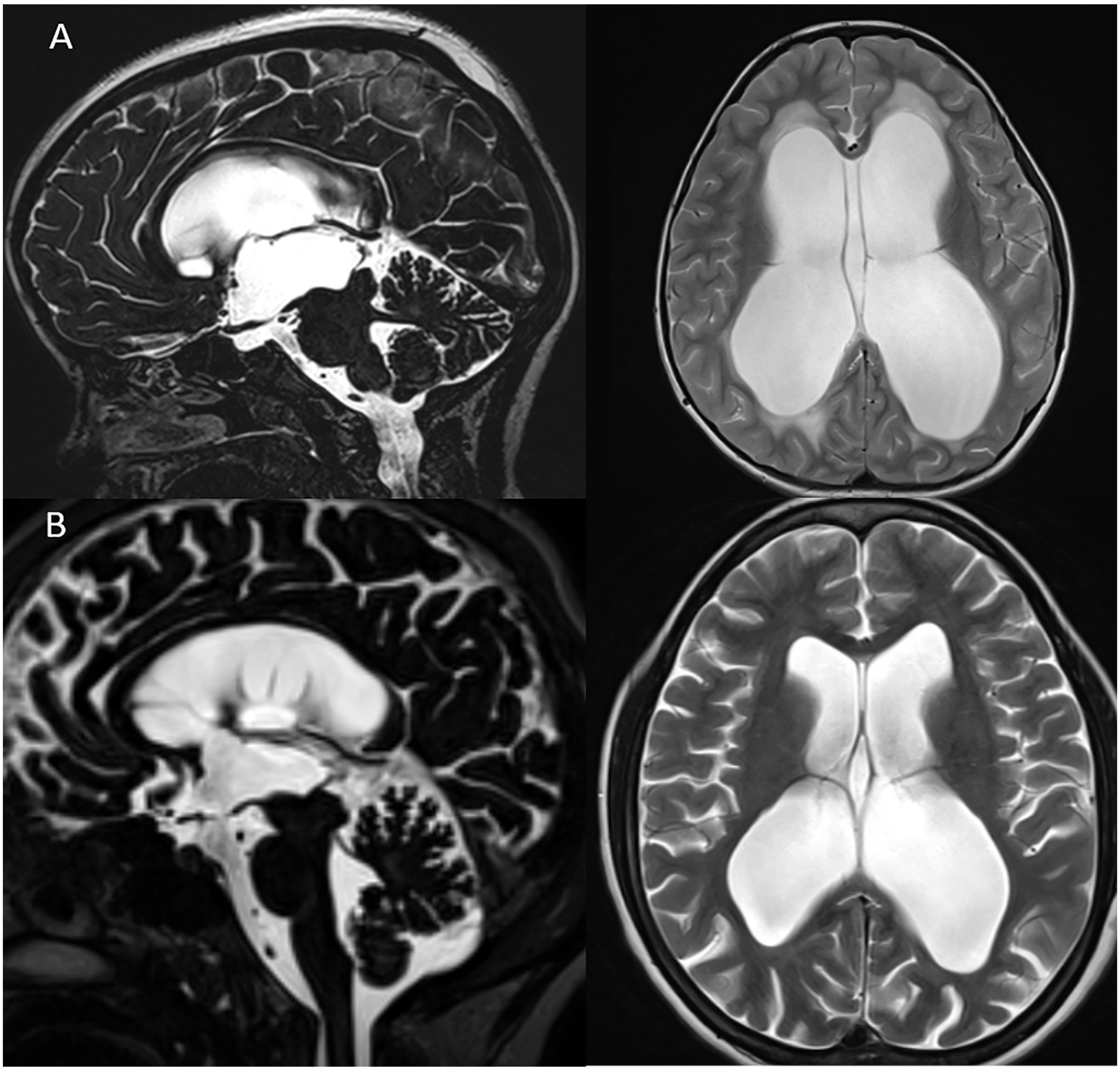

Figure 3. Radiological images of the patient who had only ETV performed due to a tectal tumor

A. Preoperative sagittal and axial MRI, B. Postoperative sagittal and axial MRI

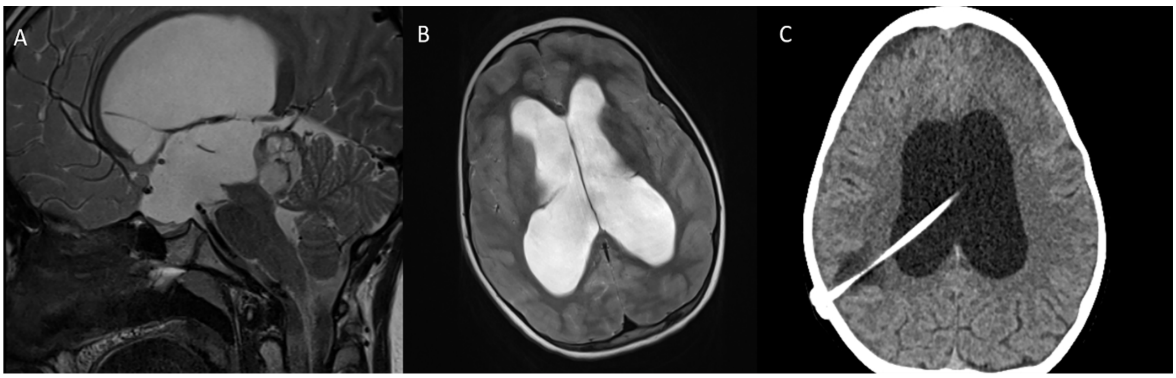

Figure 4. MRI images of a patient with pilocytic astrocytoma pathology with ETB+ETV performed due to a tectal tumor

A. Preoperative Saggital CSF flow MRI and the appearance of tectal-located tumor and hydrocephalus, B. Preoperative Axial T2 MRI image, C. CT image of a VP shunt implanted after ETV in a postoperative patient

Discussion

The pineal and adjacent tectal regions represent the most heterogeneous area in the central nervous system in terms of tumor type and histology [1]. In particular, imaging fails to characterize tumor type accurately in pineal region tumors [8], and appropriate treatment planning depends on establishing a histopathological diagnosis. Additionally, due to the high incidence of concomitant obstructive hydrocephalus in this patient group, urgent treatment is often required. In such cases, minimally invasive surgery using endoscopic techniques, which have evolved parallel to technological advancements since their description in the 1970s [9], can be employed to establish a histological diagnosis and alleviate the symptoms of obstructive hydrocephalus.

However, especially for tectal region tumors, if typical radiological findings such as hyperintensity on T2 and FLAIR sequences, iso/hypointensity on T1 sequences, and the absence of contrast enhancement on contrast-enhanced T1 sequences are present (Figure 5), many studies suggest that these tumors can remain stable for years without surgical debulking or radiotherapy [10, 11]. Tectal gliomas in children are often described as slow-growing, indolent tumors [12, 13]. However, it is emphasized that the management of concomitant hydrocephalus is necessary [14]. Some authors argue that a definitive histological diagnosis is crucial for tectal tumors [15]. In contrast, others suggest that biopsy is not necessary in cases where typical features of low-grade tectal gliomas are seen on magnetic resonance imaging (MRI) [12] (Table 1). Despite numerous publications on these tumors in this region, there is still no universally accepted algorithm [16, 17].

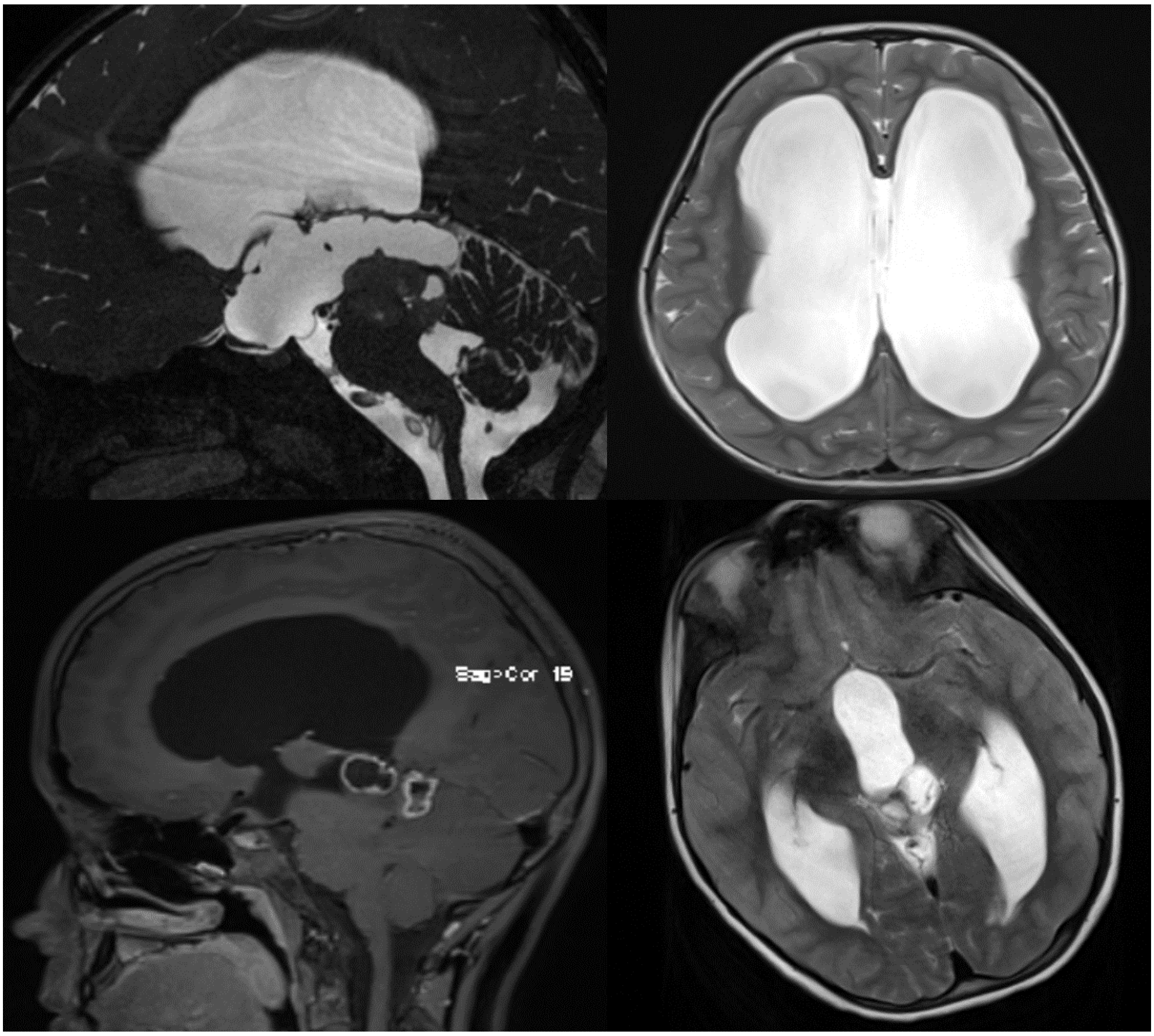

Figure 5. MRI images of a tectal tumor hyperintensity on T2 with hydrocephalus

Table 1. Literature review of ETV and endoscopic tumor biopsy (ETB) studies on tectal tumors

Study	Year	Number Of Patients	Mean Age (in years)	ETB Diagnostic Rate	ETV Success Rate
Al Tamini et al. [27]	2008	8	10	75%	88%
Wong et al. [28]	2011	25	13.5	84%	81%
Herrada Pineda et al. [29]	2015	28	8.5	96%	NA
Schulz et al. [30]	2021	28	12.4	96%	91.7%
Deopujari et al. [15]	2022	22	11	95.45%	85.7%
Present study	2023	4	6.8 (2.4,4.3,9.1,11.7 years-old)	50% (2/4)	82.5 %

In our study on ETV and ETB endoscopic treatment applied simultaneously to pediatric patients only, we found 6 cases series. According to these data, our diagnosis rate by biopsy for tectal regions was found to be low. The ETV success score in all studies was between 80-90%

In our patient follow-up, for pineal region tumors and similar cases, we initially perform blood tumor marker sampling like Deopujari et al. [15]. If the results are negative and there is no hydrocephalus, we proceed with CSF sampling through lumbar puncture. If tumor markers in the CSF are positive, we refer the patients for adjuvant treatment (Figure 6). If the results are negative, we oparete for simultaneous CSF management and CSF tumor marker sampling. Additionally, if the tumor location is suitable and the risk of bleeding is low, we plan an endoscopic biopsy during the same session [15]. Some authors advocate for performing a biopsy before conducting an ETV, particularly due to concerns like potential bleeding [18-20]. However, we support performing these procedures in a single session. Furthermore, we have not encountered scenarios in our cases that warrant an alternative approach, and minor intraoperative bleeding has not negatively affected the feasibility and outcomes of ETV.

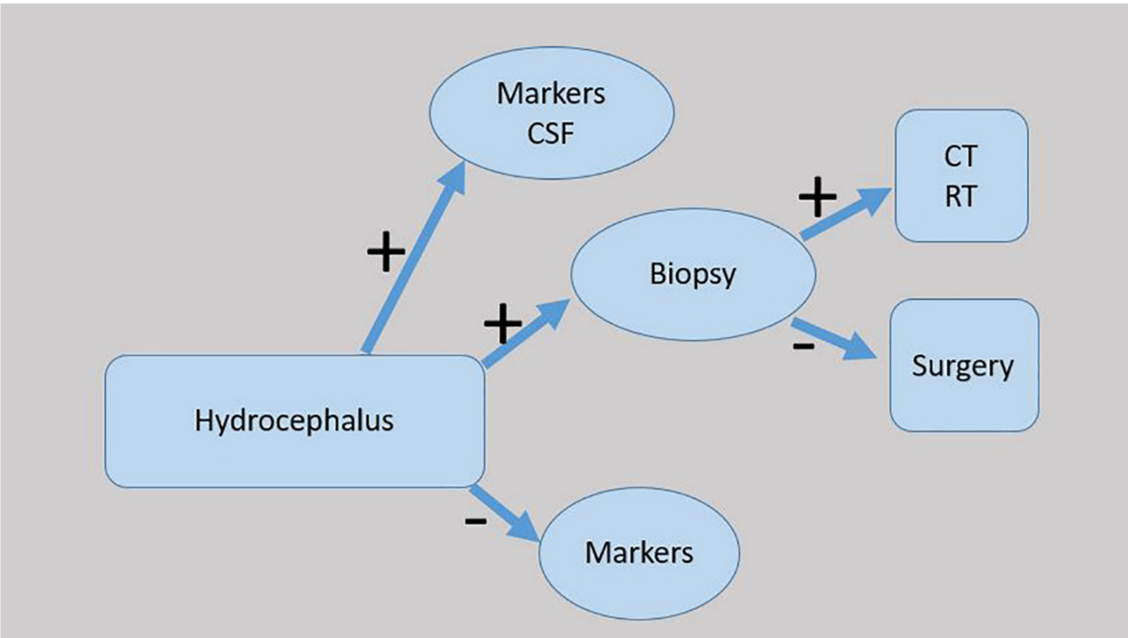

Figure 6. Treatment algorithm for pineal tumors

For pineal region tumors, we perform blood tumor markers AFP and BHCG. If the results are negative and there is no hydrocephalus, we proceed with CSF sampling through lumbar puncture for markers. If tumor markers in the CSF are positive, we refer to adjuvant treatment. If the results are negative, we operate for CSF management and CSF tumor marker sampling. If the tumor location is suitable, we take endoscopic biopsy samples

For tectal region tumors, especially when typical radiological findings are present, we prioritize ETV surgery for hydrocephalus management (Figure 7). However, in cases where unstable conditions such as radiological changes and clinical progression occur during follow-up, we plan for a biopsy. Indeed, in two of our cases, we conducted biopsy surgeries following the development of such conditions after ETV. ETV can be performed using both flexible and rigid endoscopes through one or two burr hole openings. Studies by Morgenstern et al. [21] have shown that there is no significant difference between single and double burr hole approaches. On the other hand, Ahn and Goumnerova found in their study that when flexible endoscopes were used, the biopsy success rate (45.5%) was lower compared to rigid endoscopy (81%) [22]. They attributed this to the limited sampling capacity of biopsy forceps with flexible endoscopes. In our series, we performed all endoscopic surgeries using rigid endoscopes through a single burr hole.

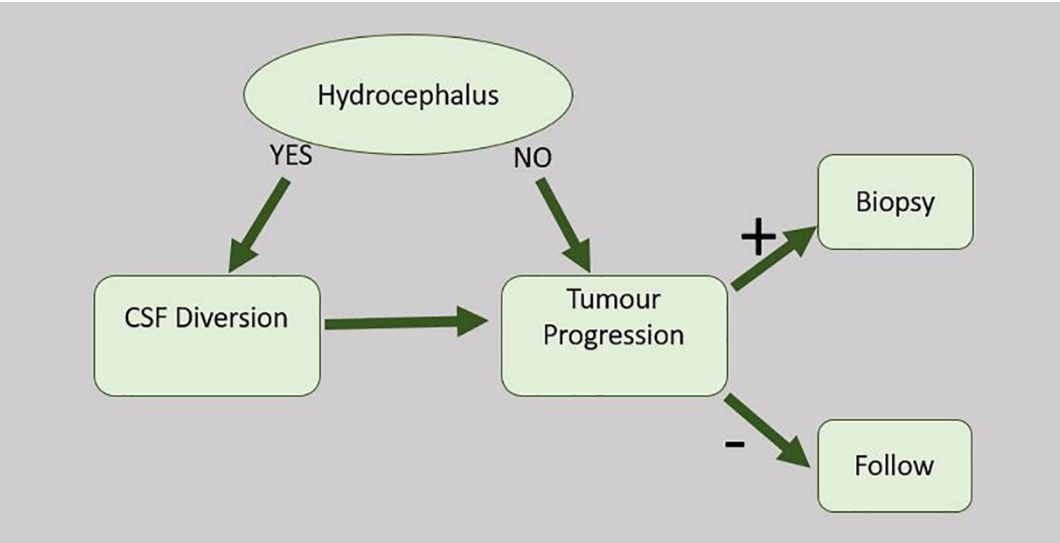

Figure 7. Treatment algorithm of tectal tumors

Tectal region tumors: If there are specific MRI markers and hydrocephalus, we usually perform endoscopic treatment to manage hydrocephalus without biopsying the tumor

In 2010, Abhaya et al. [23] developed the ETV success score (patient's age, cause of hydrocephalus, and VPS history) to predict the prognosis of ETV. According to our cases, the success score for patients with pineal region tumors was 78.3%, while it was 84% for tectal tumors. This suggests that ETV is a suitable option for hydrocephalus management, especially in all cases, including those with tectal lesions.

Yamini et al. [24] mentioned that among 54 patients who underwent ETV and biopsy, a definitive histopathological diagnosis was established in 89% of cases (Table 2). They also reported a 15% shunt placement rate after ETV [24]. Similarly, in a study conducted in 2013 by Mottolese et al. [25], they stated that the accuracy of endoscopic biopsy ranged from 61% to 100%, while the success rate of ETV ranged from 50% to 100%. In our series, the rate of histopathological diagnosis was 66.6%, and the rate of shunt placement after ETV was low, at 7.6%. Therefore, for patients with hydrocephalus due to pineal and tectal region tumors, treatment options include ETV and shunt placement [10].

Table 2. Literature review of ETB studies on pineal tumors

Study	Year	Mean Age (in years)	Number Of Patients	Biopsy Positivity Rate
Robinson et al. [31]	1997	15.7	3	100% (3/3)
Pople et al. [32]	2001	14.5	10	100% (10/10)
Haw et al. [33]	2001	14	1	100% (1/1)
Yamini et al. [24]	2004	8.5	6	66.7% (4/6)
Choi et al. [34]	2007	13	1	100% (1/1)
Al-Tamimi et al. [27]	2008	9.9	8	62.5% (5/8)
Ahn et al. [22]	2010	11.3	20	65% (13/20)
Wong et al. [35]	2011	13.5	21	95.2% (20/21)
Zhu et al. [36]	2012	17	2	100% (2/2)
Ahmed et al. [37]	2015	NA	18	NA
Abbassy et al. [38]	2018	10.7	11	81.8% (9/11)
Liu et al. [39]	2021	10.5	34	100% (34/34)
Schulz et al. [30]	2021	12.4	28	95.8% (23/24)
Cartmill et al. [40]	2000	9	1	0% (0/1)
Present study	2023	4.9 (1.1,8.8)	2	100% (2/2)

In general, ETV is considered to be the first choice for the treatment of hydrocephalus in cases with a tumor of the tectum [26]. Endoscopic aqueductoplasty (EAP) is thought to create a more physiologic state and provide long-term control. However, the long-term results of EAP have not been as successful as expected [27-31]. EAP has been shown to fail frequently. In addition, this procedure has a higher risk of damage to midbrain structures that may lead to neurologic deficits such as oculomotor or trochlear nerve palsy, Parinaud's syndrome, and periaqueductal syndrome [32]. Therefore, ETV, which has a higher long-term success rate and lower risk, is considered a better alternative for the occlusion of the aqueduct.

However, given that ETV is associated with minimal mortality and morbidity [33], we have concluded that neuroendoscopy is important in hydrocephalus management, especially for tumors in this region. Additionally, when a biopsy is indicated, endoscopy is a beneficial intervention that is safer than open microsurgical procedures in this region and facilitates radical resection.

In light of the recommendations seen in our clinical series' long-term results and literature review, as well as our clinical experience, we believe that endoscopic third ventriculostomy with simultaneous biopsy should be the first step in the management algorithm for pineal and tectal region tumors considering mortality and morbidity rates for indicated cases. Endoscopic intervention, performed through a single burr hole using rigid endoscopy, is a comfortable and safe minimally invasive method for both pathological sampling and hydrocephalus management in indicated cases during the same session.

In limitation the validation of this single-center study with a relatively small patient population with a multicenter and more extensive series in the future will be valuable in terms of contribution to the literature.

Conflict of interest: The authors declared no conflict of interest.

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Authors' contributions to the article

P.K. constructed the main idea and hypothesis of the study. P.K. and A.O.B. developed the theory and arranged/edited the material and method section. The article written by P.K. A.O.B. reviewed, corrected and approved. In addition, all authors discussed the entire study and approved the final version.