Case Report

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Haemmorhage into a Septum Pellucidum Subependymoma: An Unusual Complication of a Benign Glial Neoplasm

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Abstract

Subependymomas (SE's) are infrequent benign glial neoplasms closely related to the ventricular system. Rarely do they present with symptoms or acute haemorrhage. This case reports a 76-year-old male presenting with severe headache and decreased consciousness in the setting of a known septum pellucidum (SP) lesion and a synchronous meningiothelial meningioma. CT revealed a right lateral intraventricular haemorrhage extending to the third and fourth ventricles with associated acute hydrocephalus. CTA established the SP lesion as the source of the acute bleed. Emergency external ventricular drainage was performed followed by stereotactic fronto-parietal craniotomy and sub-total resection. Histology was notable for a WHO Grade I subependymoma. Our patient represents only the third reported case with haemorrhage into an SP subependymoma, and the first reported case of a co-existent meningiothelial meningioma. The pertinent diagnostic and management aspects of this rare lesion in an extraordinary location with an uncommon complication are herein discussed.

Keywords: Subependymal glioma; Subependymoma; Intraventricular haemorrhage; Low grade glioma; Benign tumour; Neurosurgery; Brain tumour; Septum pellucidum

Introduction

Originally described in 1945 by Sheinker, SE's are rare benign (WHO grade I) glial neoplasms representing 0.2-0.7% of all intracranial tumours [1,2]. Approximately 40% are symptomatic and classically present with symptoms of obstructive hydrocephalus, ataxia, visual changes and impaired cognition [3-5]. Rarely do they present with symptoms or signs of acute haemorrhage. To date there have been only thirteen reported cases of haemorrhage arising from subependymomas, with just two located at the septum pellucidum (SP). This report presents only the third case of SP haemorrhage and the first reported case of a co-existent meningiothelial meningioma.

Case Report

A 76-year-old male presented to the emergency department following a sudden-onset severe headache associated with nausea and unsteadiness of gait. His past medical history was significant for a complete resection of a right frontal lobe WHO Grade I meningiothelial meningioma two years prior, and a diagnosed synchronous intraventricular anterior septum pellucidum lesion. He also had significant history ischaemic heart disease, metallic aortic valve replacement (on warfarin) and an implanted defibrillator.

On initial presentation he had a left hemi-sensory neglect with reduced power in left arm flexion. Significant laboratory findings included an international normalized ratio (INR) of 2.5. Shortly thereafter, he experienced a rapid decline in his Glasgow Coma Score (GCS) necessitating intubation. Computed tomography (CT) brain showed a right lateral intraventricular haemorrhage extending to the third and fourth ventricles with associated acute hydrocephalus and transependymal CSF leak (**Figure 1a**). Concurrent CT angiography revealed increased density at the SP raising suspicion for the known lesion as the causative agent (**Figure 1b**). A magnetic resonance imaging scan was not possible as the patient's prior metallic valve replacement was incompatible.

A left frontal external ventricular drain was placed. The following week the patient underwent a stereotactic fronto-parietal craniotomy and sub-total resection via an interhemispheric approach. Intraoperative frozen specimens identified an ependymal tumour. Formal histological analysis was compatible with a WHO Grade I SE, confirmed on specialist neuropathological review (**Figure 2**).

Postoperatively the patient showed gradual neurological recovery. On day 32, he underwent insertion of a percutaneous endoscopic gastrostomy tube for maintenance of adequate nutritional intake. On day 50, the patient was discharged to rehabilitation. Whilst at rehabilitation, he succumbed to a hospital-acquired pneumonia with subsequent decline and passed away peacefully three months after initial presentation.

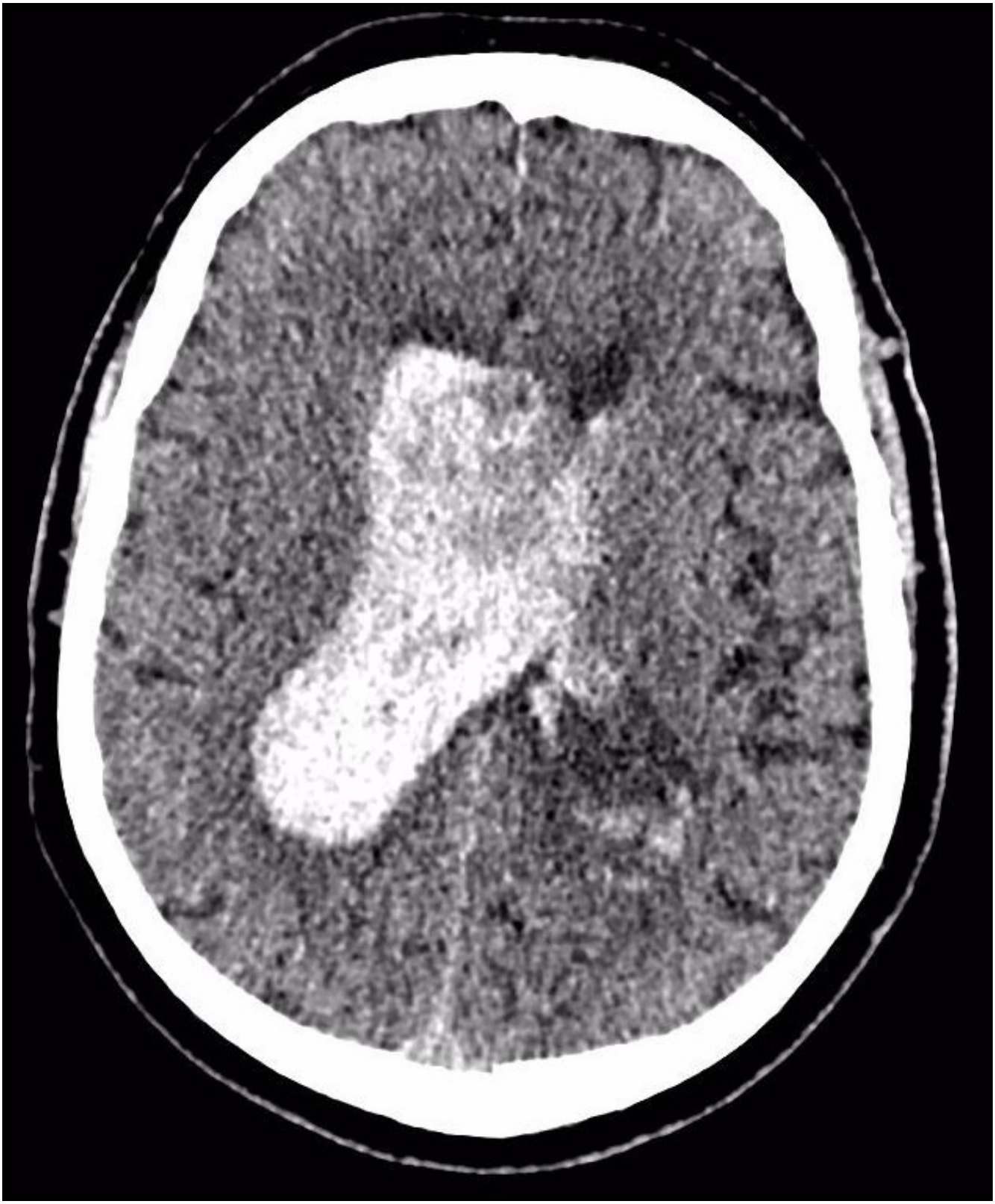

Figure 1a Axial CT brain showing interventricular haemorrhage into the right and left lateral ventricles.

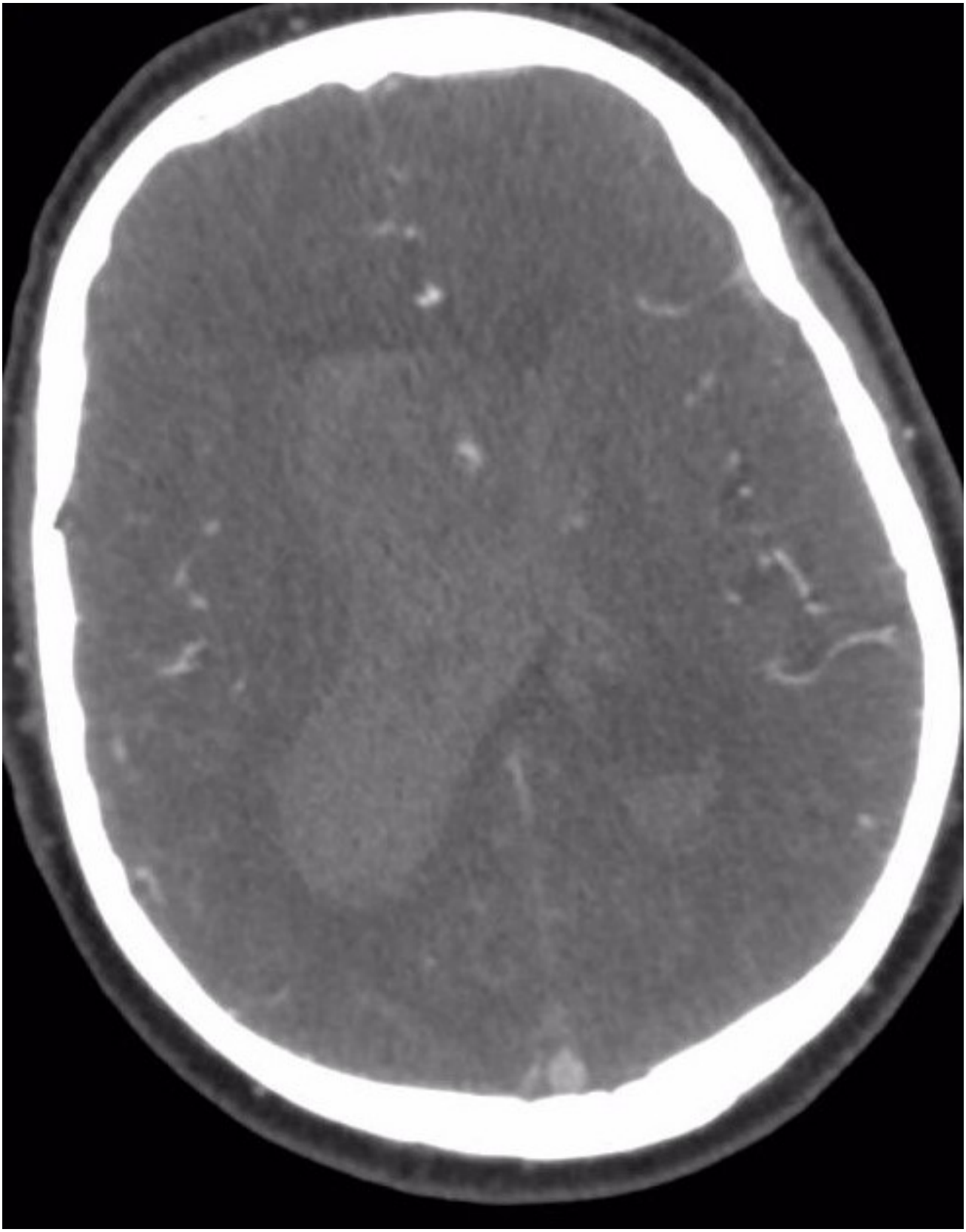

Figure 1b Coronal CTA revealing increased density at the SP.

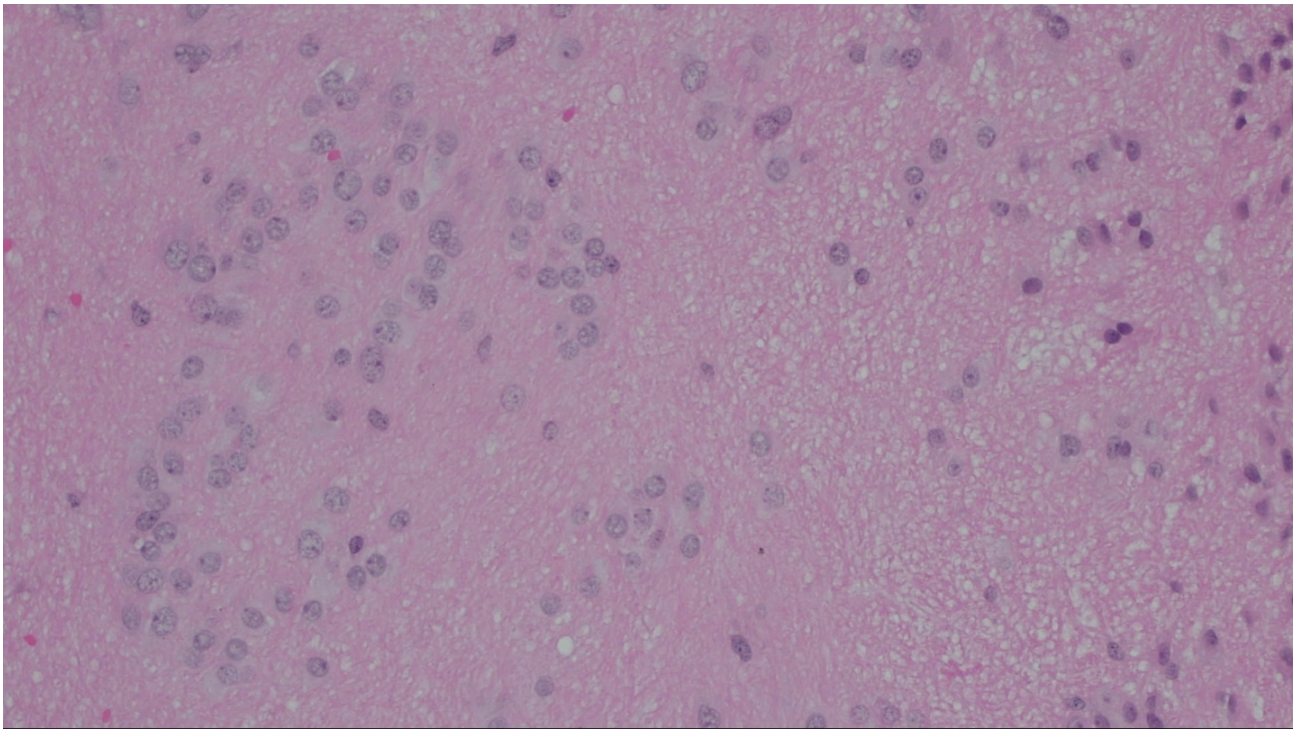

Figure 2 Haematoxylin & Eosin stained sections showing characteristic architecture with paucicellular areas separating clusters of irregularly arranged cells with oval nuclei (20x magnification).

Discussion

SEs are infrequent glial neoplasms typically described as being benign [3]. Demographically, symptomatic lesions most commonly manifest in middle-aged and elderly males however they have been described in all decades of life [1-7]. SE's are prevalently located within the fourth ventricle, followed in order by the lateral ventricles, SP, third ventricle and spinal cord [3,5]. Synchronous or metachronous lesions are extremely rare; to the author's knowledge this is the first reported case of a synchronous SE and meningioma.

Spontaneous haemorrhage subependymomas is unusual due to their hypovascularity [5]. Of the thirteen cases of SE haemorrhages identified in the literature **(Table 1),** only two prior cases involved the SP [4,8]. Moreover, these had not been diagnosed prior to the acute event and interestingly, cases had presented in considerably younger individuals **(Table 1)**. A definitive mechanism of intratumoural bleeding has yet to be elucidated; However, stretching of adjacent ependymal and subependymal veins secondary to tumour growth is implicated [5]. This patient had a significantly increased risk due to arterial hypertension, therapeutic INR and antiplatelet therapy.

The crucial features of this case are thus highlighted; a rare lesion in an extraordinary location with the unexpected complication of haemorrhage. This raises several compelling issues. Whilst it is rare for SE's to bleed, it is an understood possibility and in lesions of the SP there is no tamponade effect from surrounding brain parenchyma, leading to catastrophic sequelae. Antiplatelet and anticoagulant therapies are commonly encountered in neurosurgical patients and questions our level of caution when encountering such patients with SE's. Traditionally considered clinically benign, should we in fact be far more cautious in our monitoring and management within the context of anticoagulation and antiplatelet therapies?

Secondly, was earlier resection warranted? It is the view of Cunha and colleagues that surgical management is necessary in symptomatic patients, a notion supported by multiple groups [3,6]. The behaviour of SE's to expand into rather than infiltrate surrounding brain parenchyma and their low growth rate is thought to be optimal for curative resection [3,4]. In retrospect, a conservative approach to this asymptomatic elderly patient with multiple co-morbidities was appropriate. Evidently, a patient-specific and balanced approach is required.

In this case, investigations and subsequent management were influenced strongly by the prior knowledge of the SP lesion. Hypertensive haemorrhages can be similarly located, are far more common, and are managed conservatively. The question is raised whether more aggressive measures should be employed in order to identify an underlying cause of ictus in similarly-aged patients with no known history. This is a

complex question that deserves consideration, keeping in mind the relative benefit, yield and cost.

Table 1 Summary of previous cases of subependymoma (SEs) presenting with intratumoral haemmorhage. TC=Transcallosal, IH=interhemispheric, SAH=subarachnoid haemorrhage.

Author	Gender	Age	Predisposing Factor	Presentation	Haemorrhage	Tumour Location	Treatment	Outcome
Scheithauer	F	81	Unknown	Decreased conscious state	Tumoural	Left Ventricle	Nil operative	Haemorrhage, death
Changaris et al.	M	16	No	SAH	Tumoural, subarachnoid	Right atrium, occipital horn	Parieto-occipital transcortical, total resection	Homonymous hemianopia, survived
Seiki et al.	F	33	No	SAH	Ventricular, subarachnoid	Left atrium	Parieto-occipital transcortical, total resection	Unknown
Yamasaki et al.	F	54	No	Chronic headache	Tumoural	Left frontal horn	Transcallosal, total resection	Transient memory impairment, survived
Marra et al.	F	42	Unknown	SAH	Ventricular, subarachnoid	Right frontal horn	Transcallosal, total resection	Nil deficits, survived
DiLorenzo et al.	M	46	HTN	SAH	Ventricular, subarachnoid	Septum Pellucidum	Frontal Transcallosal, total resection	Nil deficits, survived
Lindboe et al.	M	63	Highly vascular tumour	Confusion, memory loss, urinary incontinence	Tumoural	Left frontal horn	Transcallosal, partial resection	Haemorrhage, death
Viale (awaiting PDF)	M	52	Unknown	Decreased conscious state	Tumoural	Right frontal horn	?approach, total resection	Nil deficits, survived
Furie et al.	M	46	Unknown	Chronic headache	Tumoural	Right atrium	?	Unknown
Carrasco et al.	M	71	HTN, anticoagulation	Decreased conscious state	Tumoural, ventricular	Left frontal horn	Frontal transcallosal	Permanent memory impairment, survived
Akamatsu et al.	M	32	No	Headache, decreased conscious state	Ventricular	Septum Pellucidum	Anterior TC, IH. Total resection	Nil deficits, survived
Landriel et al.	M	32	No	Headache	Cisterna Magna	Fourth Ventricle, spinal canal	Midline suboccipital, velotonsillar. Total resection	Nil deficits, survived
Present Case	M	76	HTN, anticoagulation	Headache, decreased conscious state	Septum pellucidum	Right lateral ventricle	Fronto-parietal, IH. Partial resection	Decreased conscious state, urinary incontinence, death

Conclusion

In conclusion, intraventricular haemorrhages secondary to subependymomas are extremely rare. Our patient represents only the third reported case with haemorrhage into a septum pellucidum lesion, and the first reported case of a co-existent meningiothelial meningioma. Investigation and management should be case-specific however further research is required, particularly in older patients with multiple comorbidities.

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