Archives of Pediatrics
Palis M, et al. Arch Pediatr 8: 275
www.doi.org/10.29011/2575-825X.100275
www.gavinpublishers.com

OPEN ACCESS

GAVIN PUBLISHERS

Case Report

Intracranial Granuloma and Unbalanced Epilepsy as Complications of a Subdural Peritoneal Shunt

Palis M[1], Lassalle A[1], Boetto S[2], Antherieu P[2], Raveneau M[3], Uro-coste E[4], Baudou E[1], Cances C[1*]

[1]Neuropediatric Department, Toulouse University Hospital, Toulouse, France

[2]Neurosurgery Department, Toulouse University Hospital, Toulouse, France

[3]Neuroradiology Department, Toulouse University Hospital, Toulouse, France

[4]Laboratory of Anatomy and Pathological Cytology, Toulouse University Hospital, Toulouse, France

***Corresponding author: Cances C**, Neuropediatric Department, Toulouse University Hospital, Toulouse, France

Citation: Palis M, Lassalle A, Boetto S, Antherieu P, Raveneau M, et al. (2023) Intracranial Granuloma and Unbalanced Epilepsy as Complications of a Subdural Peritoneal Shunt. Arch Pediatr 8: 275. DOI: 10.29011/2575-825X.100275

Received Date: 20 June 2023; **Accepted Date:** 28 June 2023; **Published Date:** 3 July 2023.

Abstract

Many children require placement of a subdural peritoneal shunt for congenital or acquired hydrocephalus, certain cystic malformations, or subdural hematomas. The device may be placed temporarily or permanently. As many complications have been reported, the indications for placement are limited.

We report the case of a male child who required a subdural peritoneal shunt because of a subdural hematoma as a complication of abusive head trauma in early childhood. For technical reasons, the intracranial portion could not be extracted. The child's development was marked by residual hemiparesis and balanced epilepsy. Eleven years later, the epilepsy had worsened, and a left frontal nodular formation in contact with the intracranial portion of the drain was identified, surrounded by perilesional edema. Surgical excision was performed. The histopathological examination revealed a chronic inflammatory infiltrate with superinfection by multisensitive Staphylococcus epidermidis. Antibiotic therapy was associated for 8 weeks. The control cerebral MRI at 8 weeks revealed a scarred cavity with a marked reduction in the left frontal perilesional edema, along with improvement in his epilepsy. Few similar cases have been reported in the literature. Maintaining exogenous material in the intracranial space carries a risk, even many years later, which suggests it should be systematically removed when no longer indicated or functional.

Keywords: Granuloma; Edema; Complication; Subdural peritoneal shunt; Epilepsy; Pediatric

Abbreviations

MRI: Magnetic Resonance Imaging

EEG: Electro Encephalo Graphy

RCBV: Relative Cerebral Blood Volume

Introduction

A subdural peritoneal shunt may be placed to treat congenital or acquired hydrocephalus, certain cystic malformations, or subdural hematomas. The main complications of these devices are obstruction, infection, migration, rupture of the tube, cerebrospinal fluid leak, and skin ulceration. In most cases, they are left in for a limited time. Nevertheless, when the drain adheres to the cerebral parenchyma, thus posing a significant risk of major hemorrhage or brain damage, it can be permanently left in [1].

Shaken baby syndrome is a subset of inflicted head injuries or abusive head trauma, in which shaking, alone or in combination with an impact, causes the traumatic brain injury [2]. It most often occurs in infants under 1 year old, and in two thirds of the cases

before 6 months. Intracranial injuries as subdural hematoma are most frequently observed, and some of them require neurosurgical management: the placement of a subdural peritoneal shunt is the most frequently used technique [3]. The drain is generally removed 6 months later if resolution of the hematoma is observed [4].

We report the case of a young boy who presented a secondary complication related to an intracranial drain left in place after treatment for a subdural hematoma.

Case presentation

The male child, with no personal or family medical history, was hospitalized at the age of 12 months for treatment of status epilepticus secondary to a right subdural hematoma. The etiological assessment confirmed that he had been the victim of abusive head trauma. The initial management included resuscitative measures and the placement of a subdural peritoneal shunt. Initial cerebral magnetic resonance imaging (MRI), performed at 16 months old, identified temporoparietal-occipital ischemic sequelae associated with compensatory dilation of the occipital horn of the right lateral ventricle, petechiae within the rolandic cortex and the right prefrontal cortex, and a subdural collection in the right frontal lobe and the bilateral parieto-occipital region, suggesting a chronic encysted subdural hematoma (Figure 1A). The shunt was removed 9 months later except for the intracranial portion, which was firmly adhered to the parenchyma and not extractable.

The child's neurological development was marked by residual left hemiparesis that was not markedly disabling, appropriate learning capacity in school, and non-drug-resistant structural epilepsy for many years. However, his epilepsy became drug-resistant around the age of 12 years, with no new neurological deficit. A pre-surgical assessment of the epilepsy was therefore performed: a new video electroencephalography (EEG) revealed clear focal slow wave activity with wave spikes in the right temporal region; independently, denser left fronto-temporal wave spikes; and three paradoxical left fronto-temporal seizures. Cerebral MRI revealed the formation of a 19 mm x 9 mm left frontal tissue nodule in contact with the subdural probe with voluminous perilesional edema, suggesting a granulomatous reaction to contact with the shunt device and indicating the possibility of superinfection onset (Figure 1B-E).

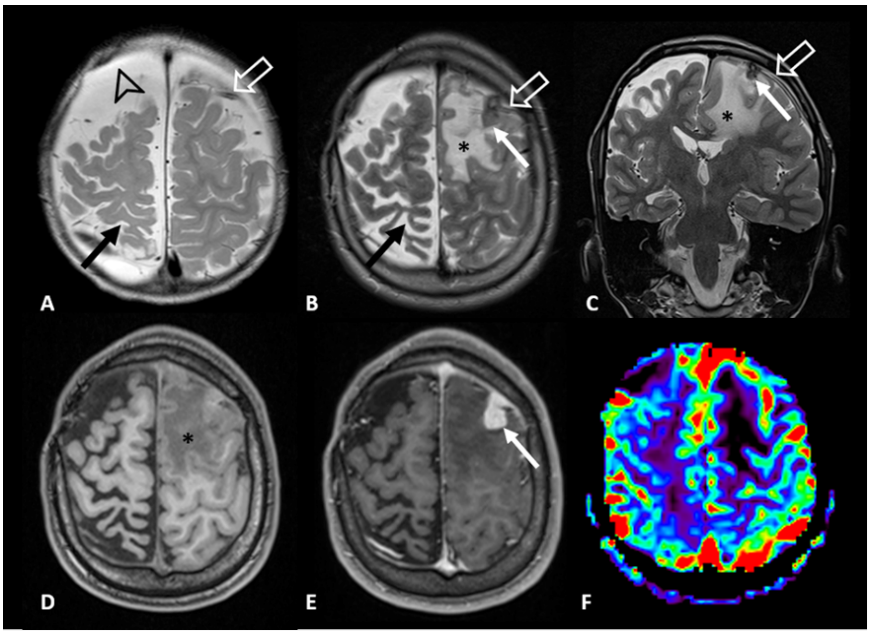

Figure 1: MRI at 16 months (A) and 12 years (B to E) with T2-weighted (B,C) and T1-weighted images before (D) and after (E) gadolinium injection; corrected rCBV perfusion mapping (F); Right frontal subdural hematoma (arrow) and T2 hyperintensity in the right parietal parenchymal lesion progressing to cortical atrophy (black arrows) related to sequelae of the shaken baby syndrome; Left frontal subdural probe (hollow arrows); Appearance of a T2 hypo intense tissue lesion with intense contrast uptake, with no neoangiogenesis in contact with the probe (white arrows); Extensive perilesional edema of the left frontal white matter (asterisk).

Management consisted of removal of the drain and the adherent lesion (Figure 2A), followed by initiation of antibiotic therapy for 8 weeks. Perioperative samples were positive for multisensitive Staphylococcus epidermidis, mycobacterial and mycological findings were negative. Histopathological examination found sclerotic scar changes associated with a significant chronic and subacute inflammatory infiltrate, sometimes purulent, with no sign of malignancy (Figure 2B-D).

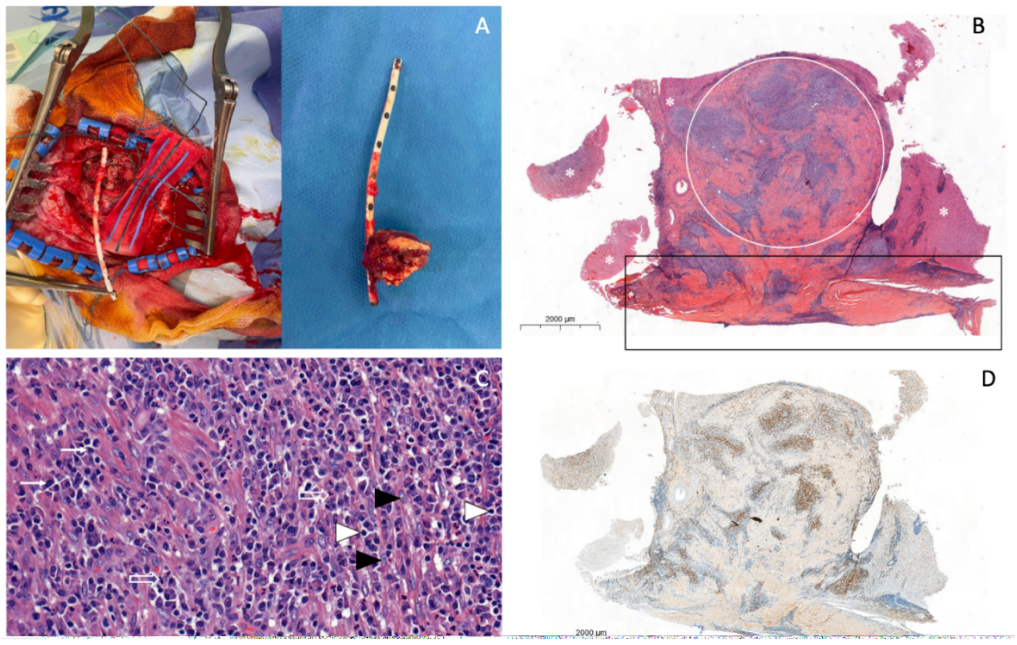

Figure 2: (A) Perioperative and postoperative pictures showing the granuloma in contact with the subdural peritoneal shunt probe; (B) Inflammatory fibrinous granuloma (white circle), in brain tissue (white asterisk). Fibrous and inflammatory meningeal thickening corresponding to pachymeningitis (black rectangle); (C) Polymorphic infiltrate of the granuloma: lymphocytes (white triangle), plasma cells (white arrows), neutrophils (hollow arrows), histiocytes (black triangle); (D) Anti CD68 marks macrophages in brown.

At 3 weeks postoperative, follow-up cerebral MRI revealed a significant decrease in the left frontal perilesional edema with peri-cavitary contrast enhancement, and no postoperative residue.

At 8 weeks postoperative, cerebral MRI confirmed a clear decrease in the perilesional edema and left frontal contrast enhancement, and antibiotic therapy was thus discontinued. At 6 months postoperative, the MRI showed residual scar tissue and the disappearance of the edema.

The child's epilepsy improved over the middle term, with more than 50% reduction in seizures following removal of the granuloma and initiation of antibiotic therapy.

Discussion

The occurrence of a granuloma in contact with a shunt in its intracranial portion, causing an unbalanced epilepsy, is rare and has rarely been described in children. In the context of a subdural hematoma, Korosue and al. (1981) described a 26-month-old child who presented with an intracranial granuloma in contact with a subdural peritoneal shunt, which was left in place due to the adherences between the material and brain parenchyma [5]. The material was removed following an intra-abdominal and then a retro-auricular infection. This complication was revealed by papilledema and left hemiparesis at the age of 6 years. The EEG revealed diffuse slow waves in the right hemisphere. Skull films disclosed bilateral parietal burr holes and the tip of the subdural shunt in the frontal region. Injected cerebral MRI showed a hyperintense homogeneous round zone in the right fronto-parietal region. A right lesionectomy was performed. The general pattern was consistent with the histological diagnosis of a typical granulomatous process. Although the bacteriological culture of the mass was negative, the hypothesis of an infectious origin was raised. In 1983, Ono Jiro and al. described the case of a 3-year 8-month-old child, who presented with bilateral intracranial granulomas in contact with the tips of subdural-peritoneal shunts left in place in the context of a subdural hematoma [6]. This complication was revealed by a status epilepticus at the age of 13 months, followed by epilepsy, and finally by an episode of acute bacterial meningitis. A brain computed tomography scan showed bilateral areas of low density in the frontal regions. Infectious etiology was confirmed by the presence of an isolated Staphylococcus epidermidis at the site of the material. Other similar cases have been reported in adult patients [7].

From a pathophysiological point of view, the mechanism of occurrence is probably immuno-inflammatory and infectious. Sarkiss and al. described the various types of biological reaction between a drain and the cerebral parenchyma [9]. Mechanical trauma during insertion seems to cause inflammatory processes inducing cell proliferation and early tube obstruction (<6 months). The prolonged and delayed immune reaction to the foreign body via the action of macrophages leads to chronic inflammation and the formation of astroglial scars, altering the biological properties of the silicone drain and contributing to later obstruction (6 months to 3 years). Activated macrophages are unable to digest the foreign body, creating multinucleated giant cells with the release of pro-inflammatory mediators, leading to granuloma formation. Tissues with significant proliferative capacity (choroid plexuses, leptomeninges, hemosiderin and blood clots), trapped inside the tube, also seem to be involved. Later effects are drain mineralization and susceptibility to failure (>3 years). Regarding the infectious mechanism, the drain is frequently found to be colonized. Germs from the cutaneous bacterial flora are often involved, but more atypical germs have also been reported: in 1997, Sharma and al. reported 32 cases of intracranial fungal granulomas due to Aspergillus, Cryptococcus, Candida and Mucoromycota [9].

Other locations have also been reported. In a 69-year-old man who presented with a subarachnoid hemorrhage following a ruptured aneurysm, Aoyama and Hida reported the formation of an abscess occurring on contact with a fragment of a lumbar drain accidentally left in place during removal and a granuloma 9 months later [10]. In a 7-year-old boy, Santos de Oliveira and al. reported an intracranial inflammatory myofibroblastic tumor following the placement of a ventriculoperitoneal shunt in the context of meningitis at the age of 6 months and complicated by hydrocephalus [11]. The treatment was surgical with removal of the granuloma and the material.

Exogenous material in the intracranial space exposes patients to potential complications, which must be evoked and investigated when neurological symptoms appear or worsen. In our case, the development of drug resistance in the context of this child's epilepsy was an expected potential risk given the initial lesions, but the emergence of seizures with contralateral onset was paradoxical, prompting us to look for a secondary complication. Thus, as Korosue and al. suggested, complete removal of the shunt device should be the treatment of choice in shunt revision surgery [5].

Conclusion

In conclusion, the long-term formation of an infectious granuloma due to an intracranial drain and causing an imbalance in known epilepsy is exceptional and an adverse effect not described in the literature.

Acknowledgment

The authors sincerely thank the patient and his parents for their participation.

Declaration of interest

The authors declare no conflict of interest.

References

1. Kurshel S, Puget S, Bourgeois M, Zerah M, Ofner P, et al. (2007) Factors influencing the complication rate of subduroperitoneal shunt placement for the treatment of subdural hematomas in infants. J Neurosurg 106: 172-178.
2. Recommandations de bonne pratique, syndrome du bébé secoué ou traumatisme crânien non accidentel par secouement, texte des recommandations, Haute Autorité de Santé, Juillet 2017
3. Choudhary AK, Servaes S, Slovis TL, Palusci VJ, Hedlund GL, et al. (2018) Consensus statement on abusive head trauma in infants and young children, Pediatric Radiology 48: 1048-1065.

4. Tude Melo JR, Di Rocco F, Bourgeois M, Puget S, Blauwblomme T, et al. (2014) Surgical options for treatment of traumatic subdural hematomas in children younger than 2 years of age. J Neurosurg Pediatr 13: 456-461.

5. Korosue K, Tamaki N, Matsumoto S, Yoshiyuki O (1981) Intracranial granuloma as an of subdural peritoneal shunt unusual complication Case report, J Neurosurg 55:136-138.

6. Ono J, Mimaki T, Okada S, Yamanishi Y, Morimoto K, et al. (1983) Bilateral Intracranial Granulomas as a Complication of Infected Subdural Peritoneal Shunt. Brain Dev 5: 499-503.

7. Iaquinandi A, Corrivetti F, Lunardi P, Ferlosio A, Giannini E, et al. (2017) Brain granuloma: rare complication of a retained catheter. World Neurosurg 110: 210-216.

8. Sarkiss CA, Sarkar R, Yong W, Lazareff JA (2014) Time dependent pattern of cellular characteristics causing ventriculoperitoneal shunt failure in children, Clin Neurol Neurosurg 127: 30-32.

9. Sharma BS, Khosla VK, Kak VK, Banerjee AK, Vasishtha RK, et al. (1997) Intracranial fungal granuloma. Surg Neurol 47: 489-497.

10. Aoyama T, Hida K (2010) Spinal Intradural Granuloma as a Complication of an Infected Cerebrospinal Fluid Drainage Tube Fragment- case report. Neurol Med Chir (Tokyo) 50: 165-167.

11. De Oliveira RS, Amato Marcelo Campos M, Brassesco MS, Valera EV, Barros Jucá CE, et al. (2009) Clinical and cytogenetic analysis of an intracranial inflammatory myofibroblastic tumor induced by a ventriculoperitoneal shunt. J Neurosurg Pediatr 4: 372-377.