A case of recurrent flight-induced cerebrospinal fluid shunt overdrainage

Linda D'Antona (MD, MBBS)[a,b]*, Claudia Louise Craven (M.Sc., MRCS)[a], Huzaifah Haq (MBBS)[a], Lewis Thorne (FRCS Neurosurgery)[a], Manjit Singh Matharu (PhD, FRCP)[b,c], Ahmed Kassem Toma (MD, FRCS Neurosurgery)[a,b], Laurence Dale Watkins (MD, FRCS Neurosurgery)[a]

[a]*Victor Horsley Department of Neurosurgery, National Hospital for Neurology and Neurosurgery, London, United Kingdom;* [b]*University College London (UCL) Queen Square Institute of Neurology, London, United Kingdom;* [c]*Headache and Facial Pain Group, National Hospital for Neurology and Neurosurgery, London, United Kingdom*

Corresponding author:

Dr Linda D'Antona

UCL Queen Square Institute of Neurology and National Hospital for Neurology and Neurosurgery, Queen Square, London. WC1N 3BG.

Email: linda.d'antona@nhs.net

Case Report for the British Journal of Neurosurgery

Abstract: 100 words

Word count: 1561 words

Tables: 0

Figures: 4

References: 14

A case of air flight-induced shunt overdrainage

Abstract

Shunted patients often complain of headaches after flights. The effect of air travel on shunt systems is unknown. We describe the case of a patient with longstanding hydrocephalus, who suffered flight-induced clinical deterioration and shunt overdrainage in two independent occasions. The patient, clinically stable for 1.5 and 5 years before each episode, reported severe headaches starting during the descent stages of the air travel. On both occasions, brain MRI imaging demonstrated pronounced ventricular size reduction. This case suggests that flight-induced shunt overdrainage can occur and should be suspected in patients with prolonged headaches and/or clinical deterioration triggered by air travel.

Keywords: air travel; cerebrospinal fluid; hydrocephalus; shunt overdrainage; ventriculoperitoneal shunt

Introduction

Beside a survey conducted by Lo Presti et al., there is paucity of evidence on the effects that air travel might have on shunted patients (Lo Presti et al. 2015). Patients with neurosurgical conditions such as colloid cysts and intracerebral tumours can suddenly deteriorate during commercial flights (Buttner et al. 1997, Goldberg and Hirschfeld 2002, Mahdavi et al. 2010, Meulen et al. 2006, Zrinzo et al. 2006). The mechanisms causing these deteriorations are similar to the high-altitude cerebral oedema (HACE) that affects climbers (Zrinzo et al. 2006). Climbing and air travel have in common a low-atmospheric pressure environment that can lead to hypoxia and ultimately raised intracranial pressure (ICP) (Lawley et al. 2016). In their survey Lo Presti et al. reported that 72% of shunted children experience transient symptoms after a flight (most commonly headache) and that these symptoms last for 1 to 14 days after the flight (Lo Presti et al. 2015). The results of this survey and the reports of

increased ICP during flights, raise the important question of what the effect of air travel on shunt systems is.

This article describes the case of a patient with longstanding hydrocephalus, who experienced two separate episodes of flight-induced shunt overdrainage.

Case Report

A 37-year-old man, with a ventriculoperitoneal shunt inserted for the treatment of decompensated longstanding hydrocephalus, had two episodes of flight-induced headache associated with evidence of shunt over-drainage in brain MRI.

The diagnosis of decompensated longstanding hydrocephalus was made in 2008, when the patient complained of severe headaches with features consistent with high cerebrospinal fluid (CSF) pressure. He had ventriculomegaly on brain imaging. ICP monitoring results demonstrated high ICP and reduced brain compliance (median 24-hour ICP 12 mmHg, median 24-hour pulse amplitude 8 mmHg). The patient was treated with a ventriculoperitoneal shunt that incorporated an adjustable differential pressure valve (Miethke ProGAV®) and the headaches subsided (Figure 1). Beside longstanding hydrocephalus, the patient also had a history of migraines controlled with propranolol prophylaxis.

Air flight-induced shunt overdrainage: first episode

The patient had been headache-free since shunt insertion and was considering discontinuing his propranolol migraine prophylaxis. In July 2010 he travelled by air from London to Scotland and suffered unusual severe episodes of headache, which started during the descent portion of both the outward and return flights. The patient reported that the headaches had a sudden onset, were severe and localised to the left frontal area; behind the left eyebrow,

radiating around the eye. The headache associated with the first flight settled spontaneously after several hours. Following the return flight, it took several days for the headache to resolve. To exclude a possible shunt malfunction, a brain MRI was performed after few weeks (August 2010). The MRI demonstrated reduction of the ventricular size and reduction of the cerebellar tonsils' descent compared to baseline (Figure 2A-B). Additionally, new signal changes within the splenium of the corpus callosum were noticed. An expert neuroradiologist suggested that these changes were in keeping with precipitous change in intraventricular pressure. A CSF sample analysis did not show any abnormality that could provide a different explanation for the splenial signal changes (normal cytology, no organisms, and negative CSF culture). The valve setting was unchanged compared to baseline (pre-flight).

The shunt overdrainage was treated with a valve adjustment (ProGAV increased from 5 to 10 cmH2O). A follow-up brain MRI performed in December 2010 demonstrated a slight re-expansion of the ventricles and partial regression of the abnormal signal within the splenium (Figure 2C-D). The patient was well after the valve setting adjustment and he remained headache-free for the following years.

Second episode

After being generally well and headache-free for several years, a second episode of air flight-induced headache occurred in August 2019, when the patient flew from London to Croatia. The headache started during the descent phase of the flight and had similar characteristics to the previous episode: severe, sudden, left frontal pain that spontaneously resolved after a few hours. The post-flight brain MRI demonstrated a dramatic reduction of the ventricular size when compared to previous imaging suggesting overdrainage (Figure 3A-B). During the following months, the patient reported feeling unwell with intermittent episodes of severe

headache and had to take a break from work. His shunt valve setting was increased (ProGAV from 10 to 12 cmH2O), but this seemed to worsen the symptoms. The valve was re-adjusted to its original setting after a few days (10 cm H2O).

In February 2020, the patient spontaneously started feeling better and a new brain MRI (Figure 3C) demonstrated re-expansion of the ventricles. The patient is currently neurologically intact, with no evidence of papilloedema and is closely monitored. His current ventricular size is very similar to the baseline brain MRI performed when he was well in 2015 (Figure 3).

A summary of events and changes in Evan's index is provided in Figure 4.

Discussion

Shunted patients are often affected by transient headaches after a flight (Lo Presti et al. 2015), but there is lack of evidence on how flights may affect shunt drainage. This is the first report of flight-induced shunt overdrainage. We hypothesise that this shunt overdrainage may have been caused by an imbalance between ICP and intrabdominal pressure during the descending phases of the flights.

The mechanisms believed to be responsible for increasing ICP during flight are cerebral oedema and vasodilation. Commercial flight cabin pressurisation achieves lower pressures than sea level and, in these conditions, the inspired partial pressure of oxygen significantly declines (Cottrell 1988). Hypoxemia can occur and 54% of passengers have SpO2 lower than 94% during flights (Humphreys et al. 2005). This hypoxemia could trigger cerebral vasodilatation and cause cerebral oedema similarly to the way this occurs in HACE (Zrinzo et al. 2006, Lawley et al. 2016). In addition, the concentration of air carbon dioxide (CO2) present in airplanes is higher than usual; in fact, concentrations of CO2 up to 0.5% are

allowed (Federal Aviation Administration; 1996). The resulting hypercarbia could cause cerebral vasodilation and further ICP increases (Zrinzo et al. 2006).

Cerebral oedema and vasodilation are initially balanced by CSF drainage but can eventually lead to raised ICP when/if autoregulation is exhausted. This hypothesis is supported by the finding that subjects with more compliant systems (atrophic brains and larger ventricles) are less likely to develop HACE (Wilson and Milledge 2008, Ross 1985). Interestingly, the unexplained splenium signal changes found in our patient's brain MRI were similar to what Hackett et al. found in patients affected by HACE (Hackett et al. 1998). This supports the hypothesis that a certain degree of flight-induced vasogenic cerebral oedema occurred in our patient.

Further considerations need to be made for patients flying with a shunt, more specifically about intrabdominal pressure during flights. It should be noted that at cabin pressure of 575 mmHg gas expands to 132% of its baseline volume (Goldberg and Hirschfeld 2002). This causes an increase in the intrabdominal pressure that could counterbalance the increase in ICP and provide resistance to CSF drainage through the shunt during the flight.

We hypothesise that during the descent portion of the flight, our patient's intrabdominal pressure reduced rapidly but the ICP reduction lagged behind, as a longer time was required to reverse the cerebral oedema. Therefore, for a certain period of time, raised ICP overcame the resistance of his intrabdominal pressure to draining CSF and causing shunt overdrainage. Subsequently the cerebral oedema resolved, and the patient responded in two different ways after each flight (Figure 4). After the first flight he was left with the effects of a mild temporary shunt overdrainage (smaller ventricles) and his symptoms resolved with a valve setting increase. In the second episode, the shunt overdrainage caused slit ventricles and obstruction of the ventricular tip of the shunt catheter leading to a temporary shunt

obstruction. This could explain why the patient did not respond to the valve setting increase and indeed actually worsened.

The possibility that his headache and the observed shunt overdrainage were not linked should be considered. The initial headache characteristics were not typical of shunt overdrainage and were actually in keeping with what the International Headache Society defines as “headache attributed to aeroplane travel” (ICHD-3, 2018). However, a typical airplane headache will settle after 30 minutes whereas this patient had headaches persisting for hours in each occasion. Moreover, the initial headaches, were followed by weeks of unusual intermittent headaches. While the initial symptoms suggest that the patient might have suffered episodes of airplane headache during the flight, the headaches which occurred in the days following airplane travel could be the result of the shunt overdrainage or could represent new migraine episodes.

This report has the obvious limitation of describing only one case. Moreover, it is not possible to establish with absolute certainty whether the shunt overdrainage episodes were flight-induced or in fact coincidental. However, that shunt overdrainage occurred after air travel on two separate occasions does support causality.

Further investigations of this phenomenon would be particularly relevant for patients prone to shunt overdrainage and development of subdural collections. Future studies could investigate the effect of flight on shunt systems utilizing telemetric ICP measuring devices (e.g. M.scio®, Miethke) to test the frequency of flight-induced shunt overdrainage.

Conclusions

This case suggests that flight-induced shunt overdrainage can occur and should be suspected in patients with prolonged headaches and/or clinical deterioration triggered by air travel.

Further studies are required to understand the mechanism of this event and assess its frequency.

Funding details: No funding was received for this research.

Disclosure statement: No funding was received for this study. LD is supported by an NIHR Academic Clinical Fellowship and was the recipient of a research fellowship sponsored by B.Braun. MSM serves on the advisory board for Allergan, Novartis, Eli Lilly, Autonomic Technologies Inc and TEVA and has received payment for the development of educational presentations from Allergan, electroCore, Eli Lilly, Novartis and TEVA. AKT research time was supported by the National Institute for Health Research University College London Hospitals Biomedical Research Centre. LDW has received honoraria from and served on advisory boards for Medtronic, B.Braun and Codman. The other authors have no disclosures to report.

Data availability statement: The authors confirm that the data supporting the findings of this study are available within the article.

Informed consent: The patient has consented to the submission of this case report for publication.

References

Buttner, A., *et al.* 1997. Colloid cysts of the third ventricle with fatal outcome: a report of two cases and review of the literature. *Int J Legal Med,* 110, 260-266.

Cottrell, J. J. 1988. Altitude exposures during aircraft flight. Flying higher. *Chest,* 93(1), 81-84.

Federal Aviation Administration 1996. Allowable Carbon Dioxide Concentration in Transport Category Airplane Cabins [online]. Federal Register. Available from: https://www.federalregister.gov/documents/1996/12/02/96-30525/allowable-carbon-dioxide-concentration-in-transport-category-airplane-cabins [Accessed 15/09/2020].

Goldberg, C. R. and Hirschfeld, A. 2002. Hemorrhage within brain tumors in association with long air travel. *Acta Neurochir (Wien),* 144, 289-293.

Hackett, P. H., *et al.* 1998. High-altitude cerebral edema evaluated with magnetic resonance imaging. *JAMA,* 280(22), 1920-1925.

Headache Classification Committee of the International Headache Society (IHS) 2018. The international classification of headache disorders 3rd edition (ICHD-3). *Cephalalgia,* 38(1), 1-211.

Humphreys, S., *et al.* 2005. The effect of high altitude commercial air travel on oxygen saturation. *Anaesthesia,* 60, 458-460.

Lawley, J. S., *et al.* 2016. Cerebral spinal fluid dynamics: effect of hypoxia and implications for high-altitude illness. *J Appl Physiol (1985),* 120(2), 251-262.

Lo Presti, A., Weil, A. G. and Ragheb, J. 2015. Flying with a shunt. *J Neurosurg Pediatr,* 15(2), 223-224.

Mahdavi, A., *et al.* 2010. Sudden deterioration due to intra-tumoral hemorrhage of ependymoma of the fourth ventricle in a child during a flight: a case report. *J Med Case Rep,* 4, 143.

Meulen, B. C., Kros, J. M. and Jacobs, B. C. 2006. Sudden death after air travel in a patient with colloid cyst. *Neurology,* 67, 998-1005.

Ross, R. T. 1985. The random nature of cerebral mountain sickness. *The Lancet,* 27, 990-991.

Wilson, M. H. and Milledge, J. 2008. Direct measurement of intracranial pressure at high altitude and correlation of ventricular size with acute mountain sickness: Brian Cummins' results from the 1985 Kishtwar expedition. *Neurosurgery,* 63(5), 970-974; discussion 974-975.

Zrinzo, L. U., *et al.* 2006. Commercial flight and patients with intracranial mass lesions: a caveat. Report of two cases. . *J Neurosurg,* 105, 627-630.

Figure 1. Postoperative CT brain performed 2 days after ventriculoperitoneal shunt insertion (November 2008).

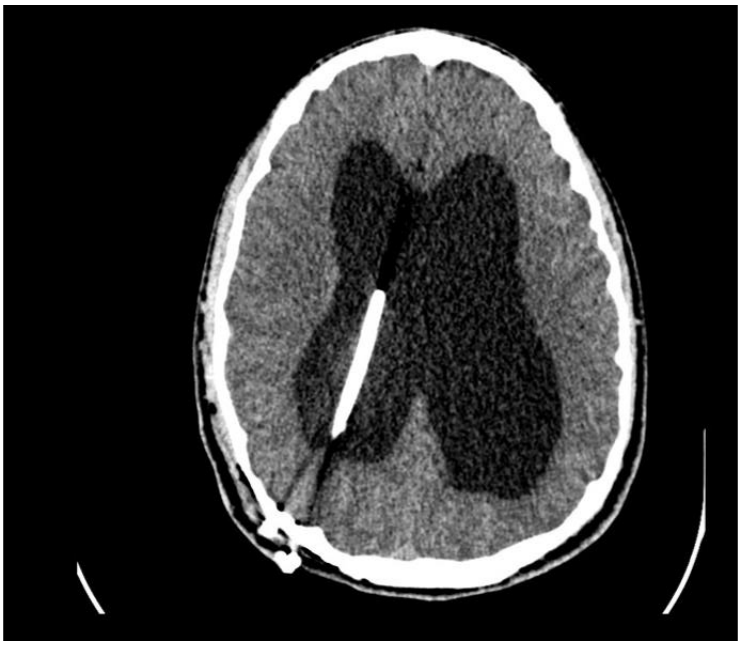

Figure 2. Brain imaging for the first episode of flight-induced shunt overdrainage. A. Baseline brain MRI (March 2009); B. Post-flight brain MRI (August 2010); C. and D. Follow-up brain MRIs (December 2010 and August 2011 respectively).

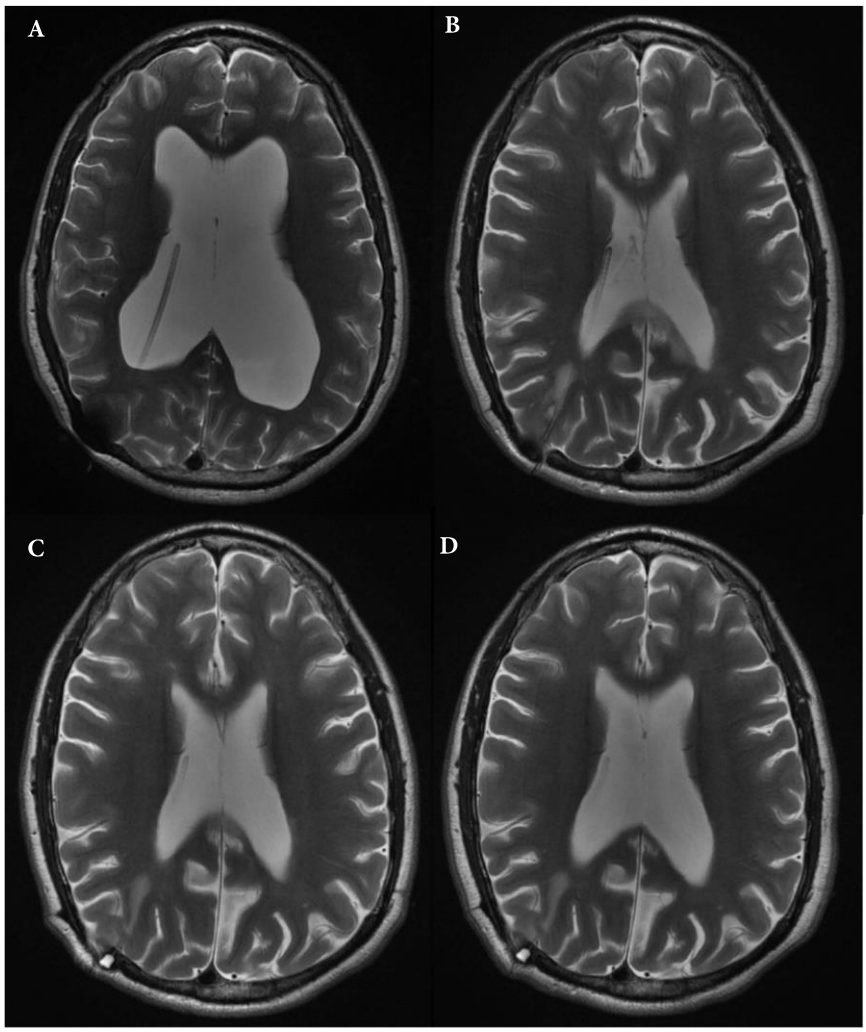

Figure 3. Brain imaging for the second episode of flight-induced shunt overdrainage. A. Baseline brain MRI (November 2015); B. Post-flight brain MRI (September 2019); C. and D. Follow-up brain MRIs (beginning and end of February 2020).

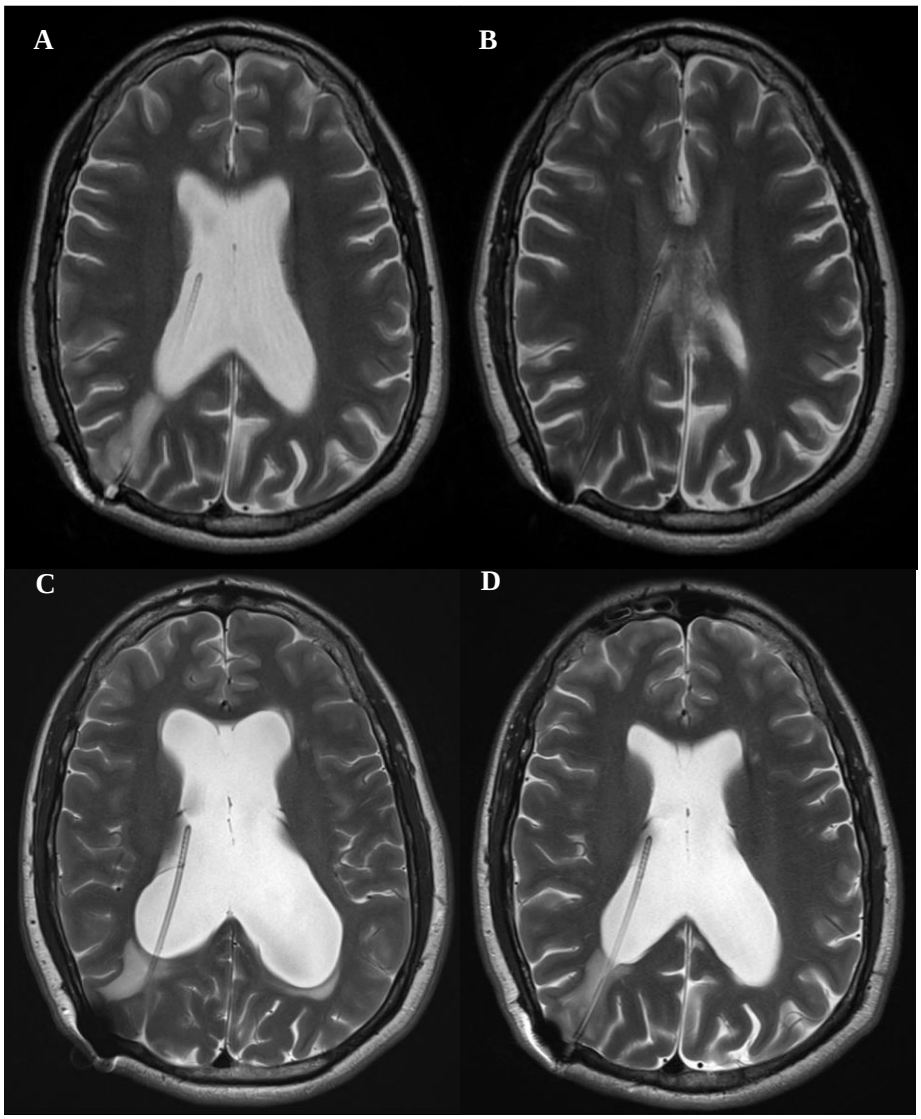

Figure 4. Summary of events and changes in Evan's index.