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CASE REPORT

Spontaneous intracranial hypotension and single entry multi-site epidural blood patch

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ABSTRACT

The syndrome of spontaneous intracranial hypotension is often difficult to treat. Unfortunately, cerebrospinal fluid leaks are often numerous and difficult to detect radiologically. Multiple entries to the spinal epidural space, in an effort to alleviate symptoms, are therefore sometimes necessary. This case report details two patients treated successfully with a single lumbar entry point and the administration of a continuous multi-site epidural blood patch via a mobile catheter and their subsequent follow-up. These procedures are based on that first published by Ohtonari *et al.* in 2012. It is, to our knowledge, the first undertaken in Australasia.

Key words: Epidural blood patch, headache, spontaneous intracranial hypotension

Introduction

The syndrome of intracranial hypotension, initially published in 1938,[1] has classically been described as an orthostatic headache, low cerebrospinal fluid (CSF) opening pressure on lumbar puncture and diffuse pachymeningeal gadolinium enhancement on magnetic resonance imaging (MRI).[2] However, many clinical variations on this exist.[3] It is thought to occur due to leaking of the CSF through the dura. Localizing the site (s) of the CSF leak is often difficult and frequently requires repeated entries to the epidural space, with the risks associated with this. Conservative treatment options include hydration, bed rest, analgesia and caffeine. Gormley[4] carried out the first epidural blood patch (EBP) in 1960 (undertaken to alleviate postlumbar puncture headache). Since then its use has been extended, it is now commonly used for those suffering from syndrome of spontaneous intracranial hypotension (SSIH). Unfortunately, this treatment has yielded disappointing results.[5] Ohtonari *et al.*[6] proposed a novel method for single entry multi-site EBP administration. Their paper details the use of a single lumbar access point and a mobile catheter from which autologous blood is introduced at multiple spinal levels in a caudal fashion. This report documents our experience with this procedure and subsequent follow-up. To our knowledge, these are the first such cases undertaken in this manner in Australasia.

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Case Report

Patient A was a 44-year-old female who presented following a 3 weeks history of intermittent headaches. She described a diffuse, constant, sharp pain, worse on standing. This was associated with blurring of vision, nausea and vomiting. Neurological examination was normal. A computed tomography (CT) brain demonstrated bilateral thin isodense subdural collections and mild mass effect with transtentorial herniation. A Magnetic resonance venography showed marked smooth T2-weight hyperintense supratentorial and infratentorial pachymeningeal thickening and enhancement overlying both cerebral hemispheres and posterior fossa. There was descent of the midbrain and iter of the cerebral aqueduct below the incisural line. An enlarged pituitary was also noted. MRI of the spine revealed extensive collection of extradural fluid extending from C2 down to the sacrum. The origin of the fluid was not apparent, but imaging was suggestive of a thoracic origin. A trial of conservative measures was ineffective. An EBP was done with 20 ml autologous blood introduced at L2/L3. No resolution of symptoms occurred. A second blood patch was undertaken at T4/T5. Although this produced a better result, her symptoms remained debilitating. She presented for multi-site EBP.

Patient B was a 43-year-old lady who first presented with new onset, recurrent, severe, bifrontal headaches. The headaches occurred 30 min after she mobilized, gradually increasing in

severity over a period of approximately 5 min. The headaches were relieved by lying down. Each episode was associated with mild photophobia and nausea. Of note, she had suffered a fall from her bike landing on her buttocks 5 days prior to the onset of any headaches. She suffered four such headaches before presenting to hospital. There was no neurological deficit on examination. A CT scan was undertaken which was normal. Her symptoms resolved, and she was discharged home. She suffered two further episodes and represented to the hospital where a lumbar puncture was performed, this was nondiagnostic. An MRI demonstrated a prominent extradural space posteriorly along the greater length of the thoracic spine but failed to show any evidence of a CSF leak. The history was suggestive of SSIH, therefore, two lumbar EBP's were undertaken at the L3/L4 level. The first afforded her a single symptom-free day and the second 3 days although the severity of the headaches diminished. She presented for multi-site EBP.

Procedure

Informed consent was obtained from both patients who underwent the procedure on an angiography table with single-plane fluoroscopy. Patient A was positioned prone. Patient B was initially positioned in the lateral position right side up. Local anesthetic (1% lignocaine) was administered and then an extradural puncture, using the paramedian approach, was performed with an 18-guage Touhy needle at L2/L3 in the interlaminar window on patient A and at L4/L5 on patient B. A guide wire (150 cm Bentson Starter guide wire, Boston Scientific) was then advanced into the epidural space with removal of the Tuohy needle and placement of a four French introducer sheath (10 cm Radiofocus introducer II, Terumo Co.). A 4 French catheter (150 cm Radiofocus Guidewire M, Terumo Co.) was initially advanced to T5/T6 for patient A, but could not be advanced further so a 2.7 French micro catheter (130 cm Progeat micro-catheter, Terumo Co.) was used and advanced to the C3/C4 disc level [Figure 1]. For patient B the 2.7 French micro-catheter was solely used and advanced cranially to C5/C6. A volume of 2 ml of Visipaque 270 was used to identify the catheter position. At this stage, approximately 2 ml of autologous blood was introduced at each segmental level as the catheter was withdrawn. This was obtained from each patient from a previously secured, sterile venepuncture in their forearm. Patient A received 50 ml extending from C4/C5 to L1/L2. Patient B received a total of 41.5 ml extending from C5/C6 to L1/L2. Patient A experienced slight back discomfort and right leg pain with the passage of the guide wires in the cephalad direction. She also experienced discomfort when the blood was injected in the thoracic region only. Patient B experienced moderately intense cervical discomfort with the introduction of the blood. Both patients' symptoms settled with simple analgesia in the recovery room. Patient A was instructed to remain on bed rest for 24 h. Patient B remained on bed rest for 48 h with graded ambulation over the next 24 h. Both patients were discharged 3 days postprocedure with no complications evident.

Follow-up

Patient A was seen in outpatients 2 months following discharge. Apart from intermittent fleeting headaches she was much improved. She went on to have a late follow-up MRI of her spine and head and was reviewed again 9 months following discharge. At this stage she was symptom-free and active. Her imaging demonstrated an improvement in her brain position compared to pretreatment scan. The upper end of the aqueduct had risen approximately 3–4 mm relative to the incisural line, the cerebellar tonsils had risen 4 mm, the enlarged pituitary had assumed a normal shape and the interpeduncular fossa and suprasella system had resumed a normal size. The MRI spine showed no abnormalities

Patient B was not formally seen in an outpatient clinic but was followed-up via telephone at 6 months. She described suffering from neck and shoulder pain for a period of about 2 weeks postprocedure and a 3 weeks window of fatigue. She

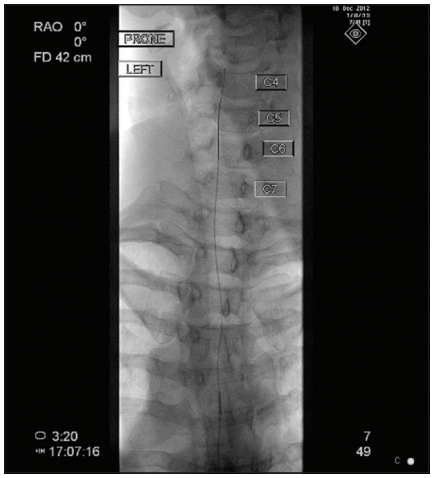

Figure 1: Advancement of Progeat micro-catheter to cervical region

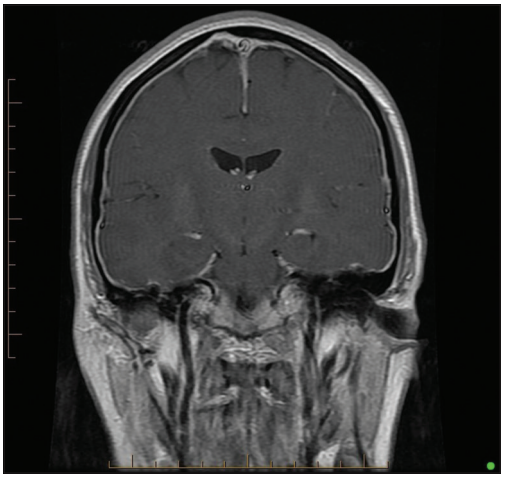

Figure 2: T1-weight coronal magnetic resonance imaging head postgadolinium pretreatment with diffuse dural enhancement

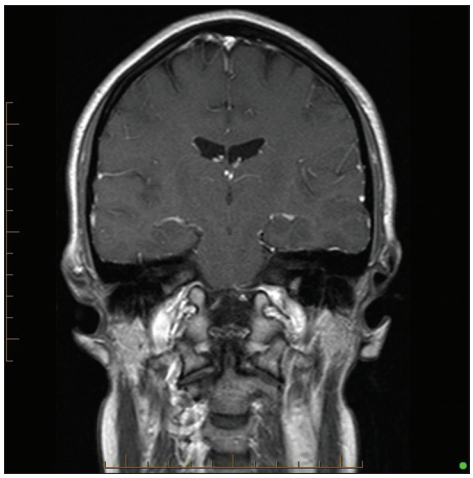

Figure 3: T1-weight coronal magnetic resonance imaging head postgadolinium posttreatment with resolution of the dural enhancement

then returned to normal activities of daily living and has been symptom-free since [Figures 2 and 3].

Discussion

A number of diagnostic tools have been described to establish CSF leakage points including MRI, CT myelography and radiosiostope cisternography. Starling *et al*.[7] demonstrated a sensitivity of 91.7% for spinal MRI when compared to CT myelography and given its less invasive nature; it may be suitable as a first-line diagnostic tool for the detection of CSF leaks. If a readily identifiable single leakage point is demonstrated on imaging, then a single EBP is the treatment of choice. However, single entry multi-site EBP may have a role in those cases where single site EBP procedures have failed. Given the slidable nature of the catheter technique used in this procedure then theoretically any leakage point identified above the access site is amenable to blood patching from the safer lumbar entry point.[6] In addition, the volume of autologous blood introduced at each spinal level can be altered.

Both patients who presented for this procedure had a consistent history and suggestive MRI findings for the diagnosis of SSIH. Conservative measures were inadequate, and two attempts at localized EBP proved unsuccessful. The procedure undertaken proved safe, efficient and effective as a treatment option for SSIH. In addition, both patients were symptom-free at 6 months. Clearly more clinical information needs to be obtained regarding the wider use of this novel technique.

Conclusion

The SSIH is often an overlooked diagnosis. Identifying the CSF leak(s) is often difficult, and EBP administration often fails. Continuous EBP administration may offer a viable solution.

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