

CASE REPORTS

Anesthesiology

1996; 85:923-5

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Lippincott-Raven Publishers

Spontaneous Intracranial Hypotension: A Series

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SPONTANEOUS intracranial hypotension is a syndrome identical in presentation to "spinal headache," but it occurs in the absence of dural puncture.¹ The most common cause is spontaneous rupture of an arachnoid cyst with persistent leak of cerebrospinal fluid (CSF). We report eight cases of spontaneous intracranial hypotension, two of which are described in detail. All patients had an incapacitating postural headache and were successfully treated with an epidural blood injection.

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Case 1

A 42-y-old occupational therapist in excellent health came to her primary physician seeking treatment for a severe occipital headache that worsened significantly on standing and was accompanied by nausea, vomiting, and tinnitus. Incapacitated by the headache, the patient had become bedridden. She had no reported history of dural puncture or trauma to the spinal cord. Physical examination was remarkable only for bilateral visual field defects. Lumbar puncture revealed an opening pressure of 40 mm H₂O (normal, 70 to 180 mm H₂O). Six weeks after the onset of headache, a computed tomograph (CT) of the head demonstrated a "ptotic brain" (defined by caudal displacement and accompanying venous engorgement). Radionuclide cisternography revealed an arachnoid cyst at the level of T12-L1, with CSF leak. The leak was believed to result from spontaneous rupture of the cyst.

An epidural blood injection was performed using 15 ml blood drawn aseptically from the left antecubital space and mixed with 1 ml Omnipaque 240 (Nycomed, Puerto Rico), a water-soluble dye. Immediate relief of headache occurred and a CT of the lumbosacral area obtained 1 h after the epidural blood patch revealed spread of the blood-dye mixture within the epidural space from T6 to L5. Twenty-four hours after the epidural blood patch, a CT of the head

was obtained that showed resuspension of the brain within the cranium and decreased venous engorgement. The patient was discharged 2 days after the blood patch and has remained asymptomatic for 18 months.

Case 2

Severe headache developed in a 38-y-old postal worker in excellent health immediately after he bent down. The headache was diffuse, slightly worse over the posterior cranium, postural, and accompanied by tinnitus and hyperacusis but was associated with no visual symptoms. He had no reported history of dural puncture or trauma to the spinal cord. The headache persisted for 6 weeks and kept him from working. Physical examination revealed normal neurologic function, except for the presence of bilateral peripheral visual field deficits. Computed tomographs of the head showed a small left subdural hematoma and diffuse dural enhancement. Radionuclide cisternography demonstrated an opening CSF pressure of 17 mm H₂O and indicated a CSF leak at T12-L1.

An epidural blood patch was performed using the same technique described in Case 1, resulting in immediate pain relief. However, a follow-up lumbosacral CT revealed that the blood-dye mixture was in the paraspinous muscles at L1-L2. Pain relief lasted only 12 h, after which the blood patch was repeated at L1-L2. Another CT demonstrated spread of the blood-dye mixture in the epidural space from T6-S1. A postprocedure CT of the head was not obtained. The patient was discharged home 24 h later and has remained asymptomatic for 18 months.

Additional Cases

In six additional cases, patients had similar symptoms (table 1). Radiocisternography was not performed in the additional six patients. An epidural blood injection was performed with subsequent resolution of the symptoms and signs of low CSF pressure; the patients have remained symptom free for at least 6 months.

Discussion

Although spontaneous intracranial hypotension may result from CSF leak, reduced CSF production, or increased CSF absorption, CSF leakage is the most common cause. In our series, leakage was demonstrated in two cases and is suggested by the positive response to epidural blood injection in the remaining six patients. Similar to postdural puncture headache, pain probably

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Received from the Department of Anesthesia, University of California, San Francisco. Submitted for publication March 15, 1996. Accepted for publication June 16, 1996.

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Key words: Epidural, Spontaneous intracranial hypotension.

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Table 1. Symptoms of Spontaneous Intracranial Hypotension on Initial Presentation

	Patient 1	Patient 2	Patient 3	Patient 4	Patient 5	Patient 6	Patient 7	Patient 8
Age (yr)	42	38	63	57	29	45	32	71
Sex	Female	Male	Male	Male	Female	Female	Female	Male
History of dural puncture	—	—	—	—	—	—	—	—
Postural headache	+	+	+	+	+	+	+	+
Diplopia	—	—	+	—	—	—	—	+
Nausea/vomiting	+	—	+	+	+	+	—	+
Tinnitus	+	+	+	+	—	+	—	—
Hyperacusis	—	+	—	+	+	+	—	—
Visual field defect	+	+	+	+	+	—	—	+
Epidural blood patch (EBP)	+	+	+	+	+	+	+	+
Resolution of headache after EBP	+	+	+	+	+	+	+	+

results from displacement or stretching of pain-sensitive structures in the cranial vault, rather than low CSF pressure.² Because the volume of the cranium is fixed, the decrease in CSF results in venous engorgement of the bridging veins to compensate for the loss of volume. These features can be readily appreciated in the CT scan from the first patient which reveals an increased space between the brain and the cranium, and enhancement of the bridging veins secondary to venous engorgement. The CT scan from the second patient also revealed a subdural hematoma, a complication that may arise from rupture of bridging as the brain retracts from the dura secondary to the decreased CSF volume.^{1,3}

Treatment options for spontaneous intracranial hypotension are essentially the same as those for postdural puncture headache: oral caffeine, epidural saline infusion, and epidural blood patch.

The efficacy of autologous epidural blood patch in treating postdural puncture headache is well documented^{4,5} and has been reported for spontaneous intracranial hypotension.⁶⁻¹⁰ The concept of "patching the hole" in the dura is supported by data showing that formation of an organized clot can tamponade CSF leak.^{11,12} Fibroblastic activity may occur within 48 h of the epidural blood patch, collagen deposit at 2 weeks, and scar formation at 3 months.

Case reports of patients who were treated successfully with an epidural injection of blood or saline in whom no leak was identified have been published.^{13,14} Although an epidural injection of blood works, it may not patch a "hole," thus raising concerns about the mechanism of this technique.

The immediacy of pain relief after an epidural blood patch may be attributed to an increase in epidural and,

therefore, CSF pressure.^{15,16} The effects of the epidural blood patch on epidural and CSF pressure can be appreciated by CT scan obtained in the first case 24 h after treatment, which showed resuspension of the brain and decreased venous engorgement. Our second patient obtained immediate relief after epidural blood patch, but for only 12 hours. Computed tomographs of the lumbosacral spine indicated blood in the paraspinal muscles. One possible explanation for the relatively transient relief is the placebo effect. More likely, depositing a large volume in the paraspinal muscles immediately adjacent to the epidural space may increase CSF pressure. However, as with epidural saline injection, this increase in pressure may be transient and insufficient to correct the underlying defect, resulting in the need for additional intervention to provide sustained relief of symptoms. In this case, the second epidural blood patch provided long-term relief.

Fifteen milliliters has been recommended as the optimal volume of blood for use in performing autologous epidural blood patch, because this volume appears to spread bidirectionally — on average six segments cephalad and three caudad.¹⁷ Consequently, bolus placement of blood within one interspace of the dural tear has been suggested for treatment of postdural puncture headache.¹⁵ The spread of blood within the epidural space demonstrated by CT scan in the present two cases is similar to the spread previously described.¹⁷

We report effective treatment of spontaneous CSF leak with an epidural blood patch in eight cases, two presented in detail. All eight patients had severe postural headache. In two patients, pretreatment studies confirmed the diagnosis and showed the distortion of the brain that occurs with loss of CSF volume. After

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treatment by epidural blood patch, the symptoms and signs of low CSF pressure resolved; in one case, symptom resolution was documented by CT scan to coincide with brain resuspension within the cranium.

The authors thank Robert Fishman, M.D., and William Dillon, M.D., for their insightful discussions regarding these patients and the syndrome of spontaneous intracranial hypotension.

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